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Tic Disorders and Tourette Syndrome
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Chapter 1

Georges Gilles de la Tourette in the history of tics

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Abstract

After a year spent working for Jean-Martin Charcot, Georges Gilles de la Tourette published an article in 1885 that would become a seminal work, reporting on nine cases of tic disease. However, only four observations meet the current criteria of the eponym, including echolalia and coprolalia, the latter term coined by Gilles de la Tourette. In 1886, Charcot asked Georges Guinon to complete the clinical picture by adding obsessional disorders temporary control of tics, and the premonitory sensation leading to their occurrence. In 1888, Charcot presided over the jury for Grégoire Breitman's thesis, then in 1890 for Jacques Catrou's thesis; both theses covered Gilles de la Tourette disease and were prepared with the namesake's help. Gilles de la Tourette put the finishing touch on the disease's description in 1900, which implicitly credited Guinon with completing the clinical picture and naming the disease at the time: "la maladie des tics convulsifs" (convulsive tic disease). That said, Gilles de la Tourette did not give much importance to his own nosographic contribution to neurology. After having been nearly forgotten, it was not until the 1960s that this pathology was definitively resurrected and validated by Arthur and Elaine Shapiro under the name of Gilles de la Tourette syndrome. This was a testament to its significant prevalence and its treatment possibilities.

FROM ÉTIENNE BOUTEILLE TO JEAN-GASPARD ITARD AND THE MARQUISE DE DAMPIERRE

In 1884 Jean-Martin Charcot (1825–93) asked his resident Georges Gilles de la Tourette (1857–1904) to study tics. At that time, the medical nosography was inaccurate and tics were not distinguished from chorea (Walusinski, 2022). In his treatise on chorea in 1810, Étienne Michel Bouteille (1732–1916) was the first to distinguish certain forms he called "pseudo-chorea" or "false chorea." Although he never used the word "tic," he described the case of a 10-year-old child: "The facial features are not simply altered; they express through their contortion—such as the grimacing of the lips and the impetuous rolling of the eyes—the most ridiculous and bizarre figures" (Bouteille, 1810). This description of abnormal movements already existed in 1790, authored by Joseph Sumeire (1735–1806), a physician in Marignane, Provence: "She continually agitated primarily her head; her eyes, lips, and facial muscles were in uninterrupted, convulsive motion" (Sumeire, 1790).

After citing Bouteille in his article, Gilles de la Tourette proposed as his first demonstrative clinical case, the observation made by Jean-Gaspard Itard (1775–1838) in 1825 (Itard, 1825), involving the famous case of the Marquise de Dampierre (1800–84), whose origins were recently discovered (Walusinski and Féray, 2020). She was in fact "Madame La Comtesse Picot de Dampierre," that is, Ernestine Émilie Prondre de Guermantes, born on August 22, 1800 ("4 Fructidor an VIII" according to the revolutionary calendar) at the Guermantes family castle in Seine et Marne, east of Paris. Ernestine was a source of worry for her mother, Eulalie de Brisay-Tholozan (1779–1866). Itard described her behavioral problems as follows: "At age 7, she was overcome with convulsive contractions in the muscles of her hands and arms, especially when she was practicing her writing; she would abruptly move her hand away from the letters she was forming. After shifting her hand, its movements became regular and could be controlled voluntarily, until another jolt once again interrupted her work. At first this was considered a sign of vivacity and harmless mischief, but since the behavior was repeated over and over, adults around the girl began to respond with reprimands and punishment. Soon, however, they became convinced that these movements were involuntary and convulsive, involving the shoulder, neck, and facial muscles. The result was extraordinary contortions and grimaces. The disease continued to progress, and when the spasms spread to the organs of the voice and speech, this youngster would utter

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bizarre cries and words devoid of meaning, but never in a state of delirium, without any disturbance of the mental faculties." Her physicians held out hope that the symptoms would improve with puberty, but in vain. They advised Ernestine's mother to find a husband for her, since marriage was an ancestral treatment for hysteria. Eulalie selected a first cousin, Louis Augustin Picot de Dampierre (1780–1841), as a potential son-in-law capable of accepting this young woman as his wife, despite her natural defects. Since he was Ernestine's third-degree relative, he had to apply for a dispensation for the marriage from the Bishop of Meaux, requested in these terms: "The most pressing reason is this young woman's health: for several years, she has suffered from an acute nervous and convulsive disease. All the medical remedies used by physicians in Paris, Brittany, and Switzerland have not improved her constitution. Her doctors agree that only a change in her civil status can bring an end to the fits she experiences. In this situation, it is improbable that she could rapidly find a husband appropriate to her rank and birth, and the idea of a delay in the consummation of the union proposed to her and approved by her, or the idea of some other obstacle, may worsen her condition" (Archives de Seine-et-Marne, 1817). Authorization was granted and the marriage contract approved by King Louis XVIII. The marriage was celebrated at the Guermantes castle on December 7, 1817 when she had just turned 17.

Itard was especially struck by the verbal tics: "Among the continual and chaotic movements resulting from these morbid contractions, those affecting the organs of the voice and speech are the only ones worthy of our attention, as they represent an exceptional phenomenon, and constitute a most disagreeable inconvenience that deprives the patient of all of the joys of society; the disturbance they create is due to the pleasure the subject takes in social interaction. For example, in the middle of a conversation in which she is most interested, she suddenly, without being able to restrain herself, interrupts what she is saying or hearing with bizarre cries and words that are even more extraordinary and contrast deplorably with her intellect and her distinguished manners. These words are mostly vulgar curses, obscene epithets, and, what is no less embarrassing for her and others listening, blunt expressions of judgment or unfavorable opinions of those present in society." During his Tuesday lesson on December 13, 1887, Charcot explained: "In Parisian high society, there was a person of the most aristocratic circles who was known for uttering vulgar words. I did not have the honor of knowing her; I only crossed paths with her one day on my way up the stairs from the Salon. I was surprised to hear her suddenly say 'shit' and 'fucking pig'" (Charcot, 1887). Gilles de la Tourette himself wrote: "On several occasions, Professor Charcot saw this patient who, even when she was quite elderly, continued to suffer from muscular incoordination and to utter obscene words in spite of herself, even in public places", as Mr. Charcot was able to witness. The political journals announced her death, which occurred in July or August 1884, and some of them treated their readers to a list of the obscene words.

Although virtually nothing is known about the course of Ernestine's disorder from 1825 until her death in 1884, we can appreciate that some of Ernestine's reported behaviors would currently be regarded as not only coprolalic, but also corresponding to what has been described as nonobscene, socially inappropriate symptoms (NOSISs) (Kurlan et al., 1996). There are significant social and emotional sequelae to living with tics and NOSISs, which can adversely affect quality of life (Eddy and Cavanna, 2013). However, these inconveniences do not seem to have hindered Ernestine's social life; for example, she was at an art exhibition when Charcot heard her swear (Fig. 1.1).

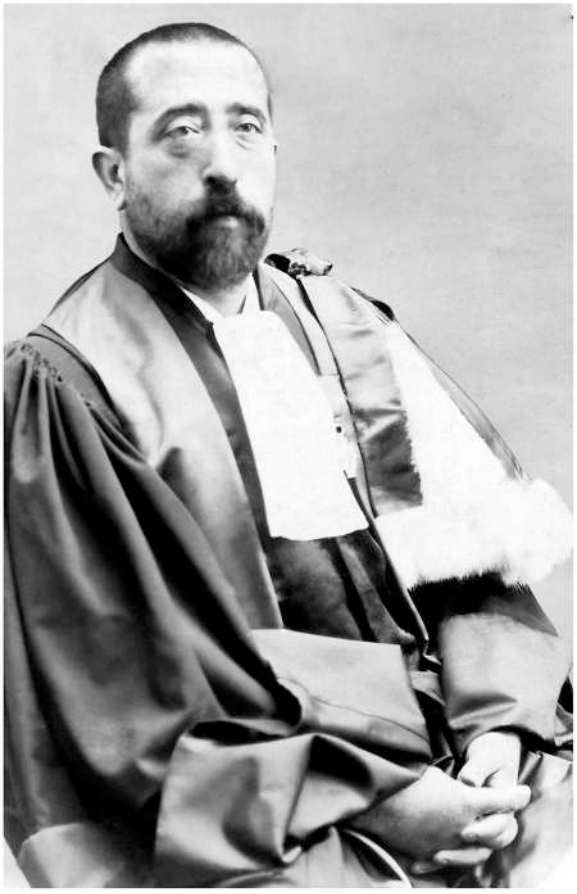

Fig. 1.1. Georges Gilles de la Tourette around 1900 (Collection OW).

CLINICAL CASES COMPILED BY GILLES DE LA TOURETTE

The second case discussed by Gilles de la Tourette involved a 17-year-old boy, hospitalized at La Salpêtrière for "sudden, involuntary movements" in the right leg and arm, accompanied by "a soft, inarticulate cry, a sort of hmm! or ahh! uttered loud enough to be perfectly audible for people around the boy." This condition had developed four years prior (Gilles de la Tourette, 1885a). These movements, which Gilles de la Tourette only qualified as tics after discussing them at length, secondarily spread to the face, then to the left limbs. They occurred in irregular fits and ceased during sleep. In addition, "the patient echoed words and even brief phrases he had heard uttered," without being able to restrain himself. Moreover, "the vulgar

nature of the word or phrase accompanying the movement is constant … He often couples his contortions with the word 'shit'." The treatments tested were not effective, but his condition improved spontaneously after he returned home. The patient "seems to have imperceptibly lost the habit of uttering vulgar words, but he remains echolalic."

The third case was that of an 11-year-old boy who, since the age of 8, had exhibited "rapid movements of flection and extension of the head and neck. Soon thereafter, these jolts spread throughout his body, his facial muscles took on various grimaces, and his arms moved, with alternating lifting of the shoulders." Then he began to utter "shit" and "damn" and became "echolalic." Follow-up until the end of 1884 showed the persistence of "frequent tics in the face with an echolalic tendency."

The fourth patient was 28 years old and complained of verbal phobic ruminations. Gilles de la Tourette did not use the word "tic" to describe his jumping movements. The absence of true echolalia and coprolalia makes it doubtful that the clinical picture he described was indeed the disease of interest, suggesting instead a case of hysteria. The fifth patient, aged 14, had facial and shoulder tics that were isolated and few in number, calling to mind transient benign tics of childhood. The sixth patient was 11 years old and had "small convulsive tics," including ocular and shoulder tics, sometimes accompanied by a few verbal tics that were only transient. The seventh patient was 21 years old. Since having been traumatized by the bombings of Paris in 1870, he had a few isolated ocular and shoulder tics that were spontaneously in the process of disappearing. Once again, this man, an anxious adult, did not really match the clinical picture for the disease. The eighth case was that of a 15-year-old girl with tics in her face and limbs. She involuntarily uttered "get away from me, get away from me you imbecile" soon replaced by "in the name of God, fuck, shit," typical of coprolalia, which she could briefly contain by inarticulate grunting. Albert Pitres (1848–1928), a former resident of Charcot who sent this patient to La Salpêtrière Hospital, reported echolalia involving dog barks and imitation of the movements and postures the patient observed around her. The ninth and last observation compiled by Gilles de la Tourette was that of a 23-year-old man with infantilism. At age 20, he developed "convulsive phenomena" consisting of "rhythmic movements" of the limbs, without echolalia or coprolalia. Once again, these symptoms had nothing in common with the disease Gilles de la Tourette was attempting to introduce into the neurologic nosography.

In total, only four observations out of the nine presented by Gilles de la Tourette correspond to the symptom complex of the disease, including the age of onset, motor and verbal tics, echolalia, and coprolalia. The others are either transient benign tics of childhood or hysterical-phobic disorders.

GILLES DE LA TOURETTE'S INITIAL INTERPRETATION

After presenting his clinical cases, Gilles de la Tourette wrote a long discussion in which he argued that the symptoms described in 1878 by the American alienist George M. Beard (1839–83), referring to the "Jumpers of Maine" (Beard, 1878), were similar. The translation of Beard's publications by Gilles de la Tourette, published in the *Archives of Neurologie* when he was a first-year resident (Walusinski, 2019), undoubtedly captured Charcot's attention and led the Maître to choose him to study these symptoms (Beard and Gilles de la Tourette, 1881). However, the Jumpers of Maine did not resemble the patients observed at La Salpêtrière (Gilles de la Tourette, 1884). Gilles de la Tourette's desire to underscore the similarity of these two disorders led him to avoid using the word "tic" in several contexts and in the title of his article. He instead employed the term "motor incoordination," unsuitable because the muscular actions involved in tics are perfectly coordinated. He saw a predominance of these tics in boys and was right to suggest that they were hereditary. He likened the suddenness and rapidity of the jumping to limb tics. Gilles de la Tourette pointed out the variability of tics over time, and how at times they could disappear totally for a while, in what can be called inexplicable "periods of remission." He also pointed out that a voluntary effort could block the appearance of tics, followed by an intense rebound. He underscored what he considered to be pathognomonic elements: "The patient cries out inarticulately," then "the cry becomes articulate and the word the patient pronounces, a variable word, is at times an echo, the subject becoming echolalic." This involuntary echolalia is never controlled by the patient. And beyond that, Gilles de la Tourette spoke of "echolalia of gestures or actions," what Charcot would name "echokinesia." In order to specifically identify "the obscene expression that their upbringing and mental state should have protected them against," he coined the word "coprolalia." "This coprolalia is not found in all subjects, in the same way that echolalia is not always present." Lacking sufficient perspective, Gilles de la Tourette wondered whether the disease could gradually disappear. He had never seen it disappear completely, but considered the prognosis to be good, with no impact on life expectancy. That said, he noted, "There is no denying that [this condition] is a deplorable companion." For him, the chronic progression "results in lazy habits and prevents intellectual development by eliminating work or resulting in serious obstacles to working." For Gilles de la Tourette, the differential diagnoses were easy to make out, especially true chorea. Facial motor tics without pain may be a way into the disease but may also spontaneously disappear. The complete clinical picture must combine "motor incoordination with articulate cries, … the utterance of articulate words with echolalia and coprolalia." Treatment was yet to be found: "How many curative procedures have been unsuccessfully tried; all sedatives of the nervous systems have failed." He recommended, not very strongly, isolation, iron, hydrotherapy, static electricity – the therapies used at La Salpêtrière to treat hysteria! As he lacked anatomic-pathologic data, he hoped that psychology would know how to interpret these disorders, and he recommended the book of the director of the *La Revue philosophique*, Théodule Ribot (1839–1916): *Les maladies de la volonté* (illnesses of the will) (Ribot, 1883) (Fig. 1.2).

Fig. 1.2. Georges Guinon in 1890 (Collection OW).

Fig. 1.3. Jean-Martin Charcot around 1881 (Collection OW).

DECISIVE ROLE OF GEORGES GUINON

Charcot quickly realized the shortcomings in the initial article by Gilles de la Tourette. Georges Guinon (1859–1932) succeeded him in 1885 as one of Charcot's residents (Walusinski, 2021). Since Charcot saw two new cases in 1885, brought back by his Italian student Giulio Melotti (1857–?) from Italy the following year (Charcot and Melotti, 1887; Kushner et al., 1999), Guinon published them in 1886 (Guinon, 1886): "Mr. Charcot has called our attention to the existence in our patients of a series of psychic phenomena not noted in other, similar observations and which, if we refer to the few cases we have been able to study, must be quite frequent in the serious forms of the tic disease. These phenomena are *idées fixes* [fixed ideas]." Also cited were the *folie du pourquoi* (compulsion to ask the reasons underlying the most insignificant things), *folie du doute avec délire du toucher* (compulsion to doubt leading to delirium), arithmomania (compulsion to calculate), and onomatomania (anxiety-producing need to find exactly the right word), recently described by Charcot and Valentin Magnan (1835–1916) (Charcot and Magnan, 1885). It was thus Guinon who added the "idea tic" WHAT IS THAT? to the clinical picture of motor tics, specifically the obsessional and compulsive disturbances that Gilles de la Tourette did not initially mention. Moreover, Guinon strengthened the overlooked significance of his contribution, highlighting two other specific criteria: the possibility for the patient to temporarily inhibit the exteriorization of motor tics and the premonitory sensation of their occurrence. Finally, he contested, and rightly so, the title Gilles de la Tourette gave his article, notably the term "motor incoordination," underscoring that tics were involuntary movements but perfectly coordinated. In addition, in 1887, Guinon authored the "Tics" chapter in the *Dictionnaire Encyclopédique des Sciences Médicales* directed by Amédée Dechambre (1812–86). In this chapter, he did a better job than Gilles de la Tourette at explaining the differential diagnoses, notably for dyskinesia, dystonia, and chorea. He noted the disease's course and its fluctuating nature with the possibility of periods of remission with few symptoms. However, and in contrast to Charcot, Guinon thought "convulsive tic disease" to be a form of hysteria. It is noteworthy that he suggested keeping the name "convulsive tic disease" whereas Charcot has coined the eponym "Gilles de la Tourette disease" to mask the approximation of the initial title, but also perhaps for the harmonious sound of the name and his affection for his unique student. Was Guinon concealing acrimony? Perhaps, but it should be noted that when Rose Kamper, a deranged patient, shot Gilles de la Tourette in 1893, Guinon was the first to help "his friend," as he warmly recounted in the article in *Progrès médical* covering this episode a few days later (Guinon, 1893) (Fig. 1.3).

In his Tuesday lesson on October 23, 1888, Charcot clearly contradicted Gilles de la Tourette: "The movements of ticcers, however complex and bizarre they may be, are not always and essentially disorganized, uncoordinated, or contradictory, as is too often believed. On the contrary, they are generally systematic, to the extent that they always recur in the same way in a given subject. Moreover, very often they reproduce, albeit exaggeratedly, certain automatic physiologic movements that have a purpose." It is during this lesson that Charcot credited Gilles de la Tourette with the coinage of the word "coprolalia": "This is the remarkable phenomenon, among all the others, that Gilles de la Tourette, in his interesting work on the tic disease, ingeniously designated using the term 'coprolalia'" (Charcot, 1889).

The last Tuesday lesson during which Charcot mentioned "convulsive tic disease" took place on June 4, 1889. At the time, he refined the semiologic description: "The complex tic movement is not absurd in itself. It is absurd and illogical because it is executed outside of a context, without an apparent reason. The act of scratching oneself occurs without an itch, the eyes blink in the absence of any foreign object, and so forth. In addition, tic movements are abrupt, rapid, momentary, and do

not have, for example, the slowness of choreic gesticulations. They are not continuous but supervene by fits repeated at greater or lesser frequencies and lasting for shorter or longer periods, fits that patients can often, for a time, stop by an effort of will. It is also often the case that while jerking and grimacing, patients utter exclamations and entire words, quite often vulgar words. You will accumulate such specific clinical characteristics that will enable you to distinguish with certainty between the tic disease and all other types of convulsive conditions" (Charcot, 1889). Charcot was once again teaching the difference between tics and chorea by presenting the students with a ticcer alongside a chorea patient (Huntington chorea), both female. By way of introduction, he declared: "You can see to what extent the electric jerks of the ticcer differ profoundly from the slow and permanent gesticulations of subjects with Sydenham's chorea. Between tics and chorea, the distance is vast. Do not forget this; these conditions are sometimes wrongly given the same name and the prognosis is quite different." Charcot showed the opposition between the speed of the movements and highlighted the constancy of choreic writhing, whereas tics stopped intermittently. The chorea patient made no sounds, whereas the ticcer "makes, from time to time, expressive laryngeal noises." The movements of the latter were chaotic and unpredictable, whereas in the former, everything was rhythmic, regular, and systematic. The ticcer started having tics at the age of 12, whereas the other patient was "over 51" at the time, with an age of onset at 33.

BREITMAN INTRODUCES DEGENERATION THEORY

Grégoire (Gregor) Breitman (1859–1914) was a physician from the city of Ananyiv, a part of Moldavia at the time, currently in the Ukraine (160 km north of Odessa). On November 28, 1888, he defended the first thesis on Gilles de la Tourette syndrome since the seminal article's publication, with Charcot presiding over his jury (Breitman, 1888). Breitman, a student alienist at the Vaucluse asylum near Paris (currently the Hôpital de santé mentale de Perray-Vaucluse, in Epinay sur Orge), wanted to study the heredity of involuntary imitation symptoms to clarify the role of "degeneration" in their genesis. This had been the accepted etiology of neuropsychiatric diseases since the work of Bénédicte-Augustin Morel (1809–73): "As the semiology of hereditary degenerates grows more detailed, exact, and manifest, we are relegating the search for the original flaw in the various forms of alienation to a secondary position"; given that "the aim is to find out whether, by studying the semiology of each psychic state and by grouping the signs into syndromes, we can, by deduction, demonstrate the manifestly degenerative hereditary origin" (Morel, 1857).

On March 6, 1890, with Charcot presiding over the jury, Jacques Catrou (1865–?), a former nonresident medical student, defended his thesis on convulsive tic disorder (Catrou, 1890), in which he included Jumping, Latah, and Myriachit, previously described by Gilles de la Tourette (Gilles de la Tourette, 1884). He started by summarizing (nearly copying) the entire beginning of the seminal article, then cited Charcot's lesson recorded by Melotti. Catrou added 28 unpublished observations that Charcot and Gilles de la Tourette had sent him. He then proceeded to analyze all of the 45 observations published to date. His thesis took a highly original turn when he argued against the theories attributing the etiology to degeneration: "We do not subscribe in any way to the current opinion whereby all nervous pathology falls under mental degeneration, aside from conditions where a physical lesion has been demonstrated."

GILLES DE LA TOURETTE'S FINAL CONTRIBUTION

Aside from the observations he gave Catrou, Gilles de la Tourette never published any other data on the disease named for him during Charcot's lifetime. It was not until 1899 that the transcript of one of his lessons at Saint-Antoine Hospital was published (Gilles de la Tourette, 1899). A 22-year-old woman came "to ask for treatment for chorea, which seemed to explain, at least in appearance, the agitation of the muscles of her face and the left side of her body." The abnormal movements began when she was 8 years old. Periods lasting a few weeks to a few months alternated with calm periods, without "muscular incoordination," but with a few residual tics in her face. Gilles de la Tourette once again employed this awkward expression, as if not to admit defeat, all while rejecting it in the lines that followed! All the physicians consulted had diagnosed chorea and prescribed arsenic and antipyrine. In addition to her motor tics, the patient had phonic tics (hum! hum! or oh ah!). "When she grows irritated with her little boy, who is very agitated, her jerks worsen and she calls him 'pig, ass', often uttering the word of Cambronne [shit] in her interjections. I was now sure; the unanimous diagnosis of chorea should be replaced by a diagnosis of convulsive tic disease with coprolalia." This young woman also exhibited phobias. In his commentary, Gilles de la Tourette recognized the contributions of Guinon and Charcot to his initial description and gave this recommendation: "If you wish to read a good survey of the question, I urge you to consult the thesis of Mr. Catrou, my student, prepared in 1890 on the convulsive tic disease, based on twenty-six unpublished observations I sent to him." Gilles de la Tourette admitted at this point that the expression "motor incoordination" was unsuitable and acknowledged the validity of Guinon's "convulsive tics": "Jerks, or tics, to use what has become the established expression." There followed a long section on coprolalia and echolalia, "which can truly be deemed psychic stigmata." Fourteen years after the initial article, this clarification in 1899 presented the disease's clinical picture and its course in a comprehensive fashion that remains relevant today. It was an occasion for Gilles de la Tourette to repeat the aphorism: "Once a ticcer, always a ticcer."

CONCLUSION

The introduction of tics into the neurologic nosography owes in large part to the La Salpêtrière School. Once he had obtained the Chair of Nervous System Diseases, created especially for him in 1881, Charcot no longer published new descriptions of

neurologic diseases in his own name, instead promoting the publications of his students. The convulsive tic disease is a demonstrative example of this strategy, which played an important role in the renown of the La Salpêtrière Master. Throughout the conceptual and observational development of Gilles de la Tourette syndrome, Charcot was the real mastermind, always in the background, but constantly vigilant that the work published by his students was relevant and thorough, as in the case of Gilles de la Tourette then Guinon, the latter being the one that the history of this creation forgot.

Gilles de la Tourette was one of Charcot's favorite disciples, and the publications that established his fame while he was alive are all linked to hysteria (Gilles de la Tourette, 1891–1895), or with the diagnosis of gait disturbances in neurology (Gilles de la Tourette, 1885b), the subject of his thesis. By contrast, those that have ensured his current fame were only considered as modestly original and innovative in his day. He himself seems to have attached little importance to them. Gilles de la Tourette disease disappeared almost totally from the medical literature from the early 20th century until the late 1950s. In the 1960s, the disease became a syndrome, firmly reassociated with Gilles de la Tourette's name after Arthur K. Shapiro (1923–95) and his wife Elaine Shapiro-Schlaffer (1925–2014), both American psychiatrists, successfully replicated Jean-Noël Seignot (1925-2022)'s observation that the disease could be treated by haloperidol (Seignot, 1961). In their article, which American medical journals refused to publish, they convincingly demonstrated the efficacy of haloperidol in calming the symptomatology of Gilles de la Tourette syndrome (Shapiro and Shapiro, 1968). Their work had a large impact, first in the community of American psychiatrists, and later among American neurologists. Shapiro thus became the instigator of a radical paradigm shift. This disease was henceforth seen as organic; psychotherapy, and to an even greater degree psychoanalysis, had no role in its treatment, aside from helping patients and their families tolerate its psycho-affective repercussions (Kushner, 1999).

Both Itard and Gilles de la Tourette "expressed their observations in beautiful, clear language, above all, in narratives, because one requires a narrative, as opposed to an itemization or checklist, to convey the full complexity and vicissitudes of a life" (Sacks, 1993). We believe that by publishing their work, Gilles de la Tourette and Charcot have pioneered a specialty still in the making, the neuroanthropology, studying not just an individual, but a society, the attitudes and reactions of which get internalized in each patient.

We conclude by noting that Gilles de la Tourette is a last name, and not a first name followed by a last name. As a result, the expression "Tourette syndrome" used in English-language articles, should be replaced with "Gilles de la Tourette syndrome."

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