MEMORY LANE

A Cursing Brain? The Histories of Tourette Syndrome, by Howard I. Kushner

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SUMMARY

Kushner's monograph (published in 2000) explores clinicians' developing understanding of a syndrome first described 200 years ago. This article highlights changing concepts of the disorder, its aetiology and treatment over its long history, as described by Kushner, and considers the current differential diagnosis of DSM-5 'Tourette's disorder'. It points out Kushner's astute observation that the clinical syndrome originally described by Itard and Gilles de la Tourette as 'maladie des tics' is not what is defined in the 21st century as Tourette syndrome.

KEYWORDS

Tic disorders; ethics; involuntary movements; expert by experience; Tourette syndrome.

Kushner's (2000) monograph *A Cursing Brain? The Histories of Tourette Syndrome* provides an in-depth insight into the clinical syndrome first described by Jean Marc Gaspard Itard in 1825 and later by Gilles de la Tourette in 1885, classified as 'maladie des tics'. Kushner takes us through a chronological journey of understanding the clinical condition and treatments, based mainly on case histories or retrospective analysis, dictated by expert opinion but thin on systematic assessment and scientific evidence of rationale or outcome.

Gilles de la Tourette syndrome and typology: many experts, differing views

Kushner refers to the case of Marquise de Dampierre, described by Itard and 60 years later by Gilles de la Tourette, their first case example of 'maladie des tics', brought to prominence by verbal tics, including coprolalia, and at the behest of Charcot renamed Gilles de la Tourette syndrome. Kushner notes Itard's description of Madame de Dampierre's presentation:

> 'in the midst of a conversation that interests her extremely, all of a sudden, without being able to prevent it, she interrupts what she is saying or what she is listening to with bizarre shouts and with inappropriate or obscene words (e.g., shit, fucking pig) which make a deplorable contrast with her intellect and her distinguished manners […] the more she was revolted by the words' grossness, the more she is tormented by the fear that she will utter them, and this preoccupation is precisely what puts them at the tip of her tongue, and she can no longer control it' (Kushner 2000: p. 10–1).

Salpêtrière contemporaries Guinon and Brissaud rejected Gilles de la Tourette typology, citing coprolalia and echolalia as symptoms presented in both chorea and hysteria, and hence not unique to the condition described by him. Charcot's student Catrou distinguished two conditions with similar symptoms. He surmised that a diagnosis of 'maladie des tics' was justified if a degenerative family history was present and interventions were unable to cure or ameliorate its course. Instead, if the patient improved without a family history laid out by Charcot and Gilles de la Tourette as typical of the syndrome described by them, a diagnosis of hysteria was appropriate.

In the late 1880s, Freud, in the case of Frau Emmy von N who had tics, was confident with his diagnosis of hysteria, consistent with Charcot's description.

Meige & Feindel's *Tics and their Treatment*, first published in 1907, posited that tics and vocalisations were a result of uncorrected infantile habits in a population with hereditary weakness and that a minority of people with tics fitted Gilles de la Tourette's description. This view contributed to the psychoanalytic views of Ferenczi and his student Margaret Mahler in the 1940s and it remained influential until the 1960s.

Outside the Salpêtrière sphere of influence, in the 1920s German observers such as Straus viewed tics as organic. Another aetiological explanation for tics was that they were the sequelae of rheumatic disease, due to infection rather that resulting from repressed childhood sexual conflict.

Kushner's review (2000) observed that, in essence, between 1885 and 1968, there were several schools of thought, none accepting Gilles de la Tourette's typology.

It is of note that a classification system based on symptomatology may describe a particular

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syndrome but that syndrome may have one or more aetiologies. Arthur and Elaine Shapiro resurrected Gilles de la Tourette's classification system, based on the effectiveness of haloperidol on presenting symptoms (Shapiro 1968). Taranta & Stollerman (1956) established the connection between the onset of Sydenham's chorea and prior infection with group A beta-haemolytic streptococcus. Immunological cross-reactive antibodies affecting the basal ganglia serve as a common factor in several movement disorders, including Tourette syndrome, Sydenham's chorea and obsessive–compulsive behaviours, setting the scene for an implication that hypersensitivity of dopamine receptors in the basal ganglia and their associated pathways and for a revision of the genetic neurotransmitter-based classification of disorders such as Tourette syndrome.

As regards outcome from remedies used, including psychoanalysis, neurosurgical interventions such as lobotomy and leucotomy, or use of haloperidol, it is unclear whether claims of cure over a period of time followed by relapse of symptoms or recurrence were reflecting the unpredictable waxing and waning course of Tourette's syndrome itself.

Kushner notes that 'if her doctors had listened to Marquise de Dampierre, rather than appropriating her symptoms to shore up their preconceived theories, much suffering may have been ameliorated' (Kushner 2000: p. 220). Kushner observed at the time of writing that the clinical condition 'maladie des tics de Gilles de la Tourette, as named by Charcot, is not the same thing as what today is called Tourette syndrome' (Kushner 2000, p. 213).

Some clinical learning points

Importance of clinical formulation based on accurate history taking and examination

In summary, Kushner describes the general views of scientists of different eras on this syndrome, from belief in enchantment by diabolical forces, an illness of the will (i.e. hereditary weakness of the will) and hysteria to a condition triggered and maintained by familial and psychosocial trauma within the psychoanalytical and psychosomatic frameworks. He notes that Tourette syndrome has been an elusive syndrome for clinicians, leading to different, often competing, perspectives regarding what defines it and what it is not, by stalwarts of the day. The case of the cursing Marquise is described, with the disputes between its proponents, many of whom had never met or examined the woman, trying to delineate the syndrome with symptomatology and presentation, rudimentary understanding of aetiology and function, and responses to treatment, each following their own school of thinking.

This book further illustrates influences of psychoanalysis, in particular those of Margaret Mahler based on the case of 'O', followed by the observations of Shapiro et al, whose inferences from patients' response to haloperidol in ameliorating symptoms of Tourette syndrome resulted in countering psychogenic frames of aetiopathogenesis, notwithstanding clashes of cultural perspectives, and the eventual triumph of the organic narrative. The book also discusses Oliver Sacks's *The Man Who Mistook His Wife for a Hat*, along with other notable essays and publications on Tourette syndrome.

From a movement disorder perspective, the book provides a detailed background on the evolution and classification of chorea, for example Sydenham's chorea, St Vitus' dance, chorea as a result of an earlier attack of rheumatic fever, chorea of rheumatic affliction, chorea as one of the symptoms of rheumatism and psychogenic chorea. Historical, at least initially, Tourette syndrome was considered as part of the continuum of chorea, and Kushner explains why Tourette syndrome was initially labelled by scientists as a variable chorea or a subtype a variation of chorea.

Contemporary differential diagnosis

DSM-5 (American Psychiatric Association 2013) refers to the condition as Tourette's disorder, classifying it under the broader category of tic disorders, describing and delineating diagnostic criteria.

Diagnosing Tourette's disorder presenting with chorea would need to take into consideration other differential diagnoses, such as autoimmune and hereditary diseases, including Huntington's disease. Indeed, Tourette's disorder has been reported in those with juvenile- or adult-onset Huntington's disease as the initial manifestation of the disease or as Huntington's disease mimicking Tourette's disorder (Alonso 2004; Chandarana 2021).

Clinically, Huntington's disease is progressive and patients' other motor skills deteriorate over time, leading to premature mortality; symptoms of Tourette's disorder wax and wane. In both Huntington's disease and Tourette's disorder, the involuntary movements are jerky, fast, sudden, recurrent and non-rhythmic. Although chorea is generally regarded involuntary and some people with Huntington's disease lack insight into their involuntary movements, in Tourette's disorder there is likely to be an element of transitory control over the motor tics and patients can have insight

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into their involuntary movements, making attempts to mask them.

Facial, tongue and body tics, eye blinking and involuntary utterances/noises/grunting can be mistaken for chorea, whereas elaborate swear words or offensive gestures and comments are more in line with a diagnosis of Tourette syndrome than Huntington's disease. Echolalia in Tourette's disorder may be mistaken for perseveration in Huntington's disease, but in the latter there are generally no reports of the echopraxia, copropraxia or coprolalia seen in Tourette's disorder.

Patients, carers, healthcare and social care professionals, epidemiologists, government departments, commissioners of services, medico-legal professionals, and basic and clinical researchers all rely on accurate diagnosis that stands over time for identifying disorder, understanding the implications of a diagnosis, planning best treatment and guidance, and setting out pathways to alleviate ill-health, physical and/or emotional pain and discomfort.

Conclusion

Kushner's book has wide ramifications in that it provides much insight into how an illness, syndrome, disease or disorder is described, evolves and becomes conceptualised and defined over time as perspectives change, influenced by scientific and technological advances in understanding and explanation of phenomenology/symptomatology, aetiopathology and emerging treatments, and by other confounding factors such as the personalities and professional standing of those propounding them.

It also highlights that ethical questions regarding whether some interventions may have caused more suffering (maleficence) than benefit (beneficence) to the patient, whether the patient was able to provide informed consent (autonomy) and whether interventions were they appropriate (justice) nonetheless went unchallenged owing to practices accepted at the time.

Patients have so much to inform us about their disease, notes Kushner, enabling discerning practitioners and researchers to provide the best holistic treatment, support carers and families, and enable individuals to participate in social life and work, with reasonable adjustments. This also resonates with current thinking on learning from patients and carers, and teaching and training in partnership with patients, as experts with lived experience of their own clinical condition (Biswas 2009).

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