



Tourette's: Syndrome, disorder or spectrum? Classificatory challenges and an appraisal of the DSM criteria



Mary May Robertson^{a,b,1}, Valsamma Eapen^{c,d,*}

^a University College London, United Kingdom

^b Department of Neurology, Room 148, Atkinson Morley Wing, St. Georges Hospital & Medical School, Blackshaw Road, London SW17 0QT, United Kingdom

^c School of Psychiatry & Ingham Institute, University of New South Wales

^d Academic Unit of Child Psychiatry, South West Sydney (AUCS), ICAMHS, Mental Health Centre, L1, Liverpool Hospital, Elizabeth Street, Liverpool, NSW 2170, Australia

ARTICLE INFO

Article history:

Received 29 March 2014

Received in revised form 29 April 2014

Accepted 25 May 2014

Keywords:

DSM-5

Classification

Diagnosis

Tics

Neurodevelopmental disorder

Tourette Syndrome

ABSTRACT

The fifth version of the Diagnostic and statistical manual of mental disorders (DSM-5) was released in May 2013 after 14 years of development and almost two decades after the last edition DSM-IV was published in 1994. We review the DSM journey with regards to Tourette Syndrome from the original publication of DSM 1 in 1952 till date. In terms of changes in DSM 5, the major shift has come in the placement of Tourette Syndrome under the 'Neurodevelopmental Disorders' alongside other disorders with a developmental origin. This review provides an overview of the changes in DSM-5 highlighting key points for clinical practice and research along with a snap shot of the current use of DSM as a classificatory system in different parts of the world and suggestions for improving the subtyping and the diagnostic confidence.

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1. Introduction

1.1. The history of Gilles de la Tourette Syndrome (GTS) – where the DSM and America are situated

DSM was initiated in 1952 with the publication of DSM-1. Since then there have been several editions which have been numerically named or designated (DSM-1 to DSM-5), with several text revisions (e.g. DSM-III-R; DSM-IV-TR). DSM-5 was published in May 2013 and was not only eagerly awaited internationally, but has drawn much comment from practitioners since publication.

Before discussing DSM (or indeed any diagnostic criteria for Gilles de la Tourette Syndrome [GTS], e.g. the WHO ICD or the Chinese Criteria one must acknowledge that GTS is not a unitary condition as was once thought (see below for details) and there is no diagnostic test for GTS either, unlike other disorders (e.g.

Huntington's Disease, Dystonia, cystic fibrosis). As such, it is more difficult to be 100% certain what the syndrome is and who does or does not have it (for full discussion see below).

Regier et al. (2013) suggest that DSM-5 changes have been driven by (i) advances in neuroscience (ii) clinical and public health needs (iii) inherent problems in DSM-IV (iv) need for better alignment with the upcoming ICD-11 of the WHO. Blumenthal-Barby (2013) suggests that the main changes in DSM-5 include (i) expanded nosology (ii) taking into account the role of claims about societal impact in changes in nosology (iii) categorisation nosology to spectrum nosology. Nemeroff et al. (2013) examined the changes, controversies and future directions of the DSM criteria. There have been numerous letters and papers about the DSM-5 and they include some specific "syndromes, disorders, spectrum disorders". For example, in the case of Autism Spectrum Disorder (ASD), there have been significant changes in DSM-5 leading to numerous comments and discussions about the DSM-5 approach to autism and ASD (e.g. Volkmar and McPartland, 2013; Guthrie et al., 2013; Hazen et al., 2013). With regards to GTS, the diagnostic criteria are quite similar to DSM-IV-TR and work well.

What the DSM (APA, 1952, 1968, 1980, 1987, 1994, 2000, 2013) first called "tics" and now calls Tourette's Disorder (TD) was in fact established first as a "European syndrome"; the first descriptions being from the Salpêtrière and allied hospitals in Paris, France. In

* Corresponding author at: Academic Unit of Child Psychiatry, South West Sydney (AUCS), ICAMHS, Mental Health Centre, L1, Liverpool Hospital, Elizabeth Street, Liverpool, NSW 2170, Australia. Tel.: +61 2 96164205/96164364; fax: +61 2 96012773.

E-mail addresses: m.robertson@ucl.ac.uk (M.M. Robertson), v.eapen@unsw.edu.au (V. Eapen).

¹ Tel.: +44 208 725 2470; fax: +44 207 226 9979.

1825 the French physician Itard is highly likely to have first documented what we know as GTS today, writing his observations of a French noblewoman the Marquise de Dampierre in: “Memoire sur quelques fonctions involontaires des appareils de la locomotion, de la prehension et de la voix” (Itard, 1825). Sixty years later, George Eduard Brutus Gilles de la Tourette described nine cases of the syndrome in 1885, earning him eponymous fame (Gilles de la Tourette, 1885). Soon after, Guinon (1886) also from France described “Sur la maladie des tics convulsifs”. In this paper and that of a fourth Frenchman, Grasset, as well as another paper of Gilles de la Tourette (translated by Robertson and Reinstein, 1991), the tic phenomenology and early psychopathological features were exquisitely described including obsessions and compulsions, which were suggested to be integral to and an essential part of the syndrome. The World Health Organization has referred to it as a “syndrome – de la Tourette syndrome” (ICD-10, 1993, F95.2) and has thus remained consistent for over 20 years. The only stipulations were that (i) multiple motor and one or more vocal tics must have been present at some time during the disorder, but not necessarily concurrently (ii) the frequency of tics must be many times a day, nearly every day, for more than 1 year, with no period of remission during that year lasting longer than two months (iii) onset is before the age of 18 years. Numerous papers from predominantly Europe marked the beginnings of GTS documentation and research, with British John Corbett contributing substantially.

The first American to publish in the area of GTS was Stevens (1965) and Stevens and Blachly (1966) including the suggested treatment of GTS and also describing the “Jumping Frenchman of Maine” (Myriachit, also described by others in USA/Canada). It was perhaps after the Frenchman physician Seignot (1961) description of the successful use of haloperidol in GTS that the physical “treatment” of GTS “took off” and in this regard the USA took the lead, with papers by Chapel et al. (1964, haloperidol), Lucas (1964, 1967, phenothiazines, haloperidol) and Shapiro and Shapiro (1968, haloperidol). The evolution of the study of GTS was then “fathered” (our word) in the main, by the well known American pioneers (Arthur and Elaine Shapiro [PubMed publications 1968–1992], including the first book (Shapiro et al., 1978), Arnold Friedhoff [publications 1966–1977], Donald Cohen [publications 1978–2003]), all of which led the way to GTS becoming an “American led syndrome”. Ruth Bruun of the Shapiro group, also a pioneer, still works in the New York area [publications starting from 1972]. Other noted American neurologists in the field were Tom Chase [publications 1984–2002] and then those who still actively contribute such as Joseph Jankovic [publications starting from 1983] and the Harvard geneticist David L. Pauls (also from the Yale Child Study centre “stable” [publications starting from 1981] and Bradley Peterson, the latter two working on the genetic and neuro-anatomical substrates of GTS. Importantly the notable clinician-researchers from the “Yale Stable” who in fact really led the way included the mentor Donald Cohen, as well as James Leckman, Lawrence Scahill and Robert King. James F. Leckman “succeeded” Donald Cohen at the Yale Child Study Centre taking over the clinical and academic “reign” of GTS, and is currently the highest international publisher [publications 1982–2013; $n = 195$] on this subject. In a recent paper entitled “The most cited works in Tourette Syndrome” (Mariam and Cavanna, 2012), no less than 72% were authored solely by American groups. Thus understandably America has taken the lead, and that is why the American Psychiatric Association (APA) criteria (DSM) and their history with regards to GTS are important to know and understand.

When discussing the DSM criteria for GTS, it might be worth mentioning the important role of the American Tourette Syndrome Association (TSA) in the development of the notion of the phenotype of GTS. The TSA, while located in the USA, always

tried to emphasise the international perspectives of GTS, including both clinical work and the basic sciences. This was undertaken in several forums including the sponsoring and holding of international GTS scientific meetings (the first held in New York City in 1981), international scientific consortia for research (e.g. the TSA International Genetic Consortium) and also the awarding of substantial competitive scientific grants, which are given to many members of the international community, and who then conduct important GTS research world-wide. The TSA also published the proceedings of the conferences as a book after each major International Meeting: the first in the “GTS Advances Journey” was Advances in Neurology in 1982 (Volume 35; edited by Arnold J Friedhoff and Thomas N Chase Raven Press, NY) which was followed by Volumes 58 (1992), 85 (2001) and 99 (2006), covering all aspects of GTS, for example clinical phenomenology, co-morbid disorders (ADHD OCD, depression, behavioural disorders), new findings in basic sciences of GTS, neuroimaging and neurophysiology of GTS, genetics and molecular biology, immunology, epidemiology, and advances in treatment in GTS. It is easy to see how all this influenced the developing notion of GTS, its recognition world-wide and thus helping understand the GTS phenotype and almost certainly influencing the DSM–GTS debate.

2. The DSM criteria: a critical appraisal

The American Psychiatric Association (APA) drew up criteria (DSM) for GTS, as did the World Health Organization (WHO), but for the ease of comparing research data with the majority of publications which were from the USA, DSM criteria became the preferred classificatory system for GTS research internationally.

2.1. DSM-1

In 1951 the American Psychiatric Association (APA) was first established and the following diagnoses relevant to our current knowledge of GTS were included as diagnostic categories (DSM-1 APA, 1952): “echolalia” (326.2), “interjectional speech” (326.2), “habit spasm” (780.4), “spasm nutans” (nodding of the head, 780.4), and “tic” (irregular muscle contraction, 780.4). Interestingly from an historical perspective, other “disorders” in category 780.4 included athetosis, choreoathetosis, combined forms of involuntary movements, dystonic movements, hemiathetosis, hemiballismus, myoclonus and neurotic excoriations, Trichotillomania and other obsessional disorders were included in 313.

2.2. DSM-11

In 1968 DSM-11 was published and tic was included under 306.2 (special symptoms not classified elsewhere).

2.3. DSM-111

DSM-111 was published in 1980 and with it the “birth” of Tourette’s Disorder (307.23) and the specification of diagnostic criteria and essential features. The diagnostic criteria included (a) age at onset between 2 and 15 years (b) presence of recurrent, involuntary, repetitive, rapid, purposeless motor movements affecting multiple muscle groups (c) multiple vocal tics (d) ability to suppress movements voluntarily for few minutes to hours (e) variations in the intensity of the symptoms over weeks or months (f) duration of more than one year.

It must be noted that the upper age limit at onset was suggested to be 15 years. It was noted that tics typically involve the head and, frequently, other parts of the body, such as the torso and upper and lower limbs. The vocal tics include various sounds such as clicks, grunts, yelps, barks, sniffs, coughs and/or words. Coprolalia, an

irresistible urge to utter obscenities, is present in around 20 to 40% of cases. It was stated that all of the symptoms are exacerbated by stress. They disappear, however during sleep, and may become attenuated during some absorbing activities. Although the tics can be voluntarily suppressed, they eventually reappear. Associated features were suggested as echokinesis, palilalia, mental coprolalia, obsessive thoughts of doubting, and compulsive/impulsive urge to touch things or to perform complicated movements, such as squatting, deep knee bends, retracing steps, and twirling when walking. Non specific EEG abnormalities, soft neurological signs, subtle central nervous system or psychological test abnormalities, hyperactivity or perceptual problems during infancy and childhood, or organic stigmata occur in about half of the individuals with the disorder. The course is usually life-long, with brief periods of remission. It was stated that “there are no known predisposing factors” and the suggested prevalence was between 0.1 and 0.5 per thousand.

2.4. DSM-111-TR (text revised)

DSM-111-TR (text revised) was published in 1987. The diagnostic criteria for Tourette’s Disorder (307.23) were as follows: (a) both multiple motor and one or more vocal tics have been present at some time during the illness, although not necessarily concurrently (b) the tics occur many times a day (usually in bouts), nearly every day or intermittently throughout a period of more than one year (c) the anatomic location, number, frequency, complexity, and severity of the tics change over time (d) onset before age 21 (e) occurrence not exclusively during Psychoactive Substance Intoxication or known central nervous system disease, such as Huntington’s Chorea and post-viral encephalitis.

2.5. DSM-IV

DSM-IV was published in 1994. The diagnostic criteria for Tourette’s Disorder (307.23) included the following: (a) both multiple motor and one or more vocal tics have been present at some time during the illness, although not necessarily concurrently (a tic is a rapid sudden, recurrent, non-rhythmic, stereotyped motor movement or vocalisation) (b) the tics occur many times a day (usually in bouts) every day or intermittently throughout a period of more than one year, and during this time there was never a tic-free period of more than 3 consecutive months (c) the disturbance causes marked distress or significant impairment in social, occupational, or other important areas of functioning (d) the onset is before age 18 years (e) the disturbance is not due to the direct physiological effects of a substance (e.g. stimulants) or a general medical condition (e.g. Huntington’s Disease or post-viral encephalitis).

2.6. DSM-IV-TR (text revised)

DSM-IV-TR (text revised) was published in 2000. The diagnostic criteria for Tourette’s Disorder (307.23) included the following: (a) both multiple motor and one or more vocal tics have been present at some time during the illness, although not necessarily concurrently (a tic is a rapid sudden, recurrent, non-rhythmic, stereotyped motor movement or vocalisation) (b) the tics occur many times a day (usually in bouts) every day or intermittently throughout a period of more than one year, and during this time there was never a tic-free period of more than 3 consecutive months (c) the onset is before age 18 years (d) the disturbance is not due to the direct physiological effects of a substance (e.g. stimulants) or a general medical condition (e.g. Huntington’s Disease or post-viral encephalitis).

2.7. DSM-5

DSM-5 was published in May 2013. GTS is included as a “Neurodevelopmental Disorder” and is alongside/after (i) Intellectual Disabilities (ii) Communication Disorders (iii) Autistic Spectrum Disorder (ASD) and (iv) Attention Deficit/Hyperactivity Disorder (ADHD). GTS is included as one of the “Motor Disorders” and is further included in the group of “Tic Disorders”.

The diagnostic criteria for Tourette’s Disorder (307.23 DSM-V; F95.2 ICD-10) include: (i) both multiple motor and one or more vocal tics present at some time during the illness, although not necessarily concurrently (ii) the tics may wax and wane in frequency but have persisted for more than one year since first tic onset (iii) onset before the age of 18 years (iv) the disturbance is not attributable to the physiological effects of a substance (e.g. cocaine) or other medical condition (e.g. Huntington’s Disease, post-viral encephalitis).

3. The Tourette Syndrome Study Group criteria (1993)

The Tourette Syndrome Study Group criteria (coded as A-1 or A-2 depending on the source of information) are as follows: (i) both multiple motor and one or more vocal tics have been present at some time during the illness, although not necessarily concurrently (ii) the tics occur many times a day, nearly every day, or intermittently throughout a period of more than one year (iii) the anatomic location, number, frequency, complexity, type, or severity of tics changes over time (iv) the tics have their onset before age 21 years (v) involuntary movements and noises cannot be explained by other medical conditions (vi) motor and/or vocal must be witnessed by a reliable examiner directly at some point in the illness or be recorded by videotape or cinematography (for definite TS, A-1) or (for tics not witnessed by a reliable examiner), tics must be witnessed by a reliable family member or close friend, and description of tics as demonstrated must be accepted by reliable examiner (for TS by history, A-2). This was used by many during the DSM-IV period, which many found unacceptable, especially because of the impairment and distress stipulation (see later).

4. The ICD-10 criteria (WHO, 1992/3) F95.2

The ICD-10 criteria (WHO, 1992/3) were also used by many particularly during the DSM-IV period because of the objection to the impairment and distress stipulation (see later). In addition and importantly F95.2 used the term “Combined vocal and multiple motor tic disorder” (de la Tourette’s syndrome), with the emphasis being on the *syndrome* (which we suggest is an acknowledgement of the fact that GTS includes a group of observable symptoms, and has no diagnostic test). The ICD-10 criteria include: (a) multiple motor tics and one or more vocal tics have been present at some time during the disorder, but not necessarily concurrently (b) the frequency of tics must be many times a day, nearly every day, for more than one year, with no period of remission during that year lasting longer than 2 months (c) onset is before the age of 18 years.

5. The Chinese GTS Diagnostic Criteria

The Chinese have their own Diagnostic Criteria for GTS which include both impairment and distress and this may be one of the reasons that GTS is found to be somewhat less common in China than the rest of the world (Robertson, 2008a,b).

6. Gilles de la Tourette Syndrome (Tourette's Disorder, DSM-5 criteria): the current status

Gilles de la Tourette Syndrome/Disorder (GTS) is a complex neuropsychiatric disorder with onset in childhood characterised by multiple motor tics and one or more vocal/phonic tics, lasting longer than a year (DSM-5, APA, 2013; WHO, 1992).

GTS has been described almost worldwide. Males are more commonly affected, with the male: female ratio being 3/4:1 (Robertson, 2012). Clinical characteristics are similar irrespective of the country of origin, highlighting the biological nature of GTS. Evidence from family genetic studies suggests that within families, the affected males more often have tics, whereas females have OCB (Eapen et al., 1993). GTS was once considered to be rare, but in one comprehensive review, it was shown that at least eight studies with similar multi-staged methods documented remarkably consistent rates of occurrence and suggested a global prevalence of between 0.4% and 3.8% for youngsters between the ages of 5 and 18 years with a calculated prevalence (from raw data) of 1% worldwide apart from Sub-Saharan Black Africa (Robertson, 2008a,b). Aetiological suggestions for GTS include genetic factors and environmental influences. The latter might include infections and neuroimmunological effects, pre- and/or peri-natal difficulties, psychosocial stressors and/or androgen influences. The idea that the aetiology of GTS was psychological has now been discredited (Robertson and Eapen, 2013).

What is particularly relevant to the discussion in this paper is that there firstly is no diagnostic test for GTS and secondly that GTS is now recognised to have many subtypes/phenotypes (Robertson and Eapen, 2013) and it is not a unitary condition as has always been stipulated/suggested by both the American Psychiatric Association DSM Criteria and the World Health Organization ICD criteria.

7. Generally accepted clinical characteristics of GTS

Many clinicians who have been involved with GTS both in a clinical and research capacity have seen DSM and ICD changes over time as well as other classificatory suggestions (e.g. Tourette Syndrome Study Group). Most have opted for DSM as the majority of GTS research has come out of the USA, but when criteria were felt to be not in keeping with the expert consensus for GTS (e.g. the distress and impairment criteria in DSM-IV), several experts expressed their concerns (e.g. Freeman et al., 1995; Comings, 1995; Kurlan et al., 1997) and DSM-IV-TR changed the criteria in several aspects to address these concerns (see later). In the intervening times researchers used DSM-III-R criteria, those of the Tourette Syndrome Study Group, or ICD-10 Criteria (as references for research projects), etc. but in the main clinicians just used a pragmatic approach and clinical features as described below.

The age at onset of GTS ranges from 2 to 21 years, with a mean of 7 years commonly reported. The onset of vocal tics is usually some months to years later, many studies reporting at about 11 years. Tics usually begin in the head and face, and eye blinking is often the first and one of the most common first tics. Tics can be simple (e.g. blinking, eye rolling, squinting, nasal twitching, head nodding) or complex (e.g. touching, hopping, squatting). Simple vocal/phonic tics include sniffing, throat clearing, gulping, snorting and coughing. Complex vocalisations include barking, making animal noises and uttering strings of words. Tics characteristically wax and wane, are usually preceded by premonitory sensations (PMS), diminish during goal-directed behaviour and increase with emotional excitement and fatigue. Other important and characteristic features include echolalia (copying what others say), echopraxia (copying what others do), palipraxia (repeating an

action) and palilalia (repeating the last word or part of sentence said by the individual (Robertson, 2012, 2013)).

Echo-phenomena have long been understood to be part of GTS, first described by Gilles de la Tourette himself in 1885. In a recent review it was re-emphasised that echo-phenomena are healthy phenomena in child development between 28 and 36 months: beyond those ages, one should investigate for neuropsychiatric pathology. It was also concluded that echo-phenomena are a feature of GTS, with patients echoing healthy movements and tics, but echoes are mainly part of the tic repertoire (Ganos et al., 2012).

Coprolalia has been widely misunderstood as a pathognomonic feature. It is the inappropriate, involuntary out-of-context swearing, often disguised by the patient and without offensive intent. It occurs in approximately 20–30% of GTS clinic populations (e.g. Robertson et al., 1988) and often begins about 5 years after tics onset. The USA Tourette Syndrome Association suggests that as few as 10–15% of all people with GTS have coprolalia. Copropraxia (the inappropriate making of obscene gestures) occurs in about 6–18% of clinic patients (Robertson et al., 1988; Freeman et al., 2009). Freeman et al. (2009) showed that coprophenomena are associated with tic severity, repetitive behaviours, the amount of co-morbidities and the number of anti-tic medications. The peak of tic severity is around 10–12 years and symptoms reduce (but do not disappear) with age (Robertson, 2012, 2013; Pappert et al., 2003). However, the prognosis of GTS is better than originally thought with regards to tic symptomatology (for review see Robertson, 2008a,b). The co-morbidity and thus phenotype often changes with age (Rizzo et al., 2012) and the psychopathology (e.g. depression) worsens with age (e.g. Robertson, 2006).

8. The co-morbidities, integral symptoms and co-existent psychopathologies of GTS

Robertson (2012, 2013) suggested that GTS be viewed as “Pure GTS” (tics only), “GTS plus” (+echo- and copro-phenomena) and “Full Blown GTS” (full house of co-morbidities and co-existent psychopathologies) and there also exist symptoms which are “integral” to GTS such as self-injurious behaviours (SIB) and non-obscene socially inappropriate behaviours (NOSI).

The co-morbid disorders associated with GTS include attention deficit hyperactivity disorder (ADHD), obsessive-compulsive behaviours/disorder (OCB/OCD) and autistic spectrum disorders (ASD) (Eapen and Crncec, 2009). That there is a genetic relationship between GTS and OCB/D is almost certain whilst ADHD and GTS seem to be genetically related in a sub-group (see reviews Robertson, 2012, 2013). Studies have shown that GTS and ASD have clinical similarities, share many symptoms and recently evidence, from phenomenological, epidemiological and pathogenetic perspectives show that GTS and ASD overlap (Eapen et al., 2013; State, 2010). It has been further suggested that shared molecular pathways affect the development of both disorders: examples include disruption of NRXN1, NLGN4X and CNTNAP2 genes (Clarke et al., 2012; Eapen, 2012). Therefore it seems likely that the above disorders are co-morbid with GTS, as they have neuro-developmental similarities, clinical similarities and in some cases are probably genetically related.

There are also numerous other psychopathologies which are associated with GTS and are suggested to *co-exist* as there are to date no aetiological similarities. What is important to understand is that very few (around 10% only) GTS individuals have “pure tics” only. A recent study observed that 13% of GTS patients had no co-morbid disorders or co-existent psychopathologies whilst the remainder had other psychiatric co-morbid diagnoses (ADHD, OCD/B, ASD) or co-existent psychopathologies (e.g. depression, anxiety, phobias, conduct disorder (CD), oppositional defiant

disorder (ODD), learning difficulties, substance abuse and, in adults, personality disorder) (Robertson et al., 2014). It is often the co-morbid or co-existent disorders/psychopathology which results in lowered quality of life, impairment and social difficulties (Robertson, 2012, 2013).

In summary, co-morbidity and/or psychopathology are common, include a wide variety of disorders and it seems that they (the GTS phenotype) change over time and with increasing age. It appears that uncomplicated GTS (“GTS-only”) have the best prognosis. Some of these co-morbidities are common and integral (e.g. OCD/OCB, ADHD, ASD), while other co-existent psychopathologies (e.g. depression) are common but multifactorial in origin, and yet others such as personality disorder in adults and bipolar disorder may be due to co-morbid conditions (e.g. ADHD, OCD) rather than the GTS per se.

It was suggested (Robertson, 2008b) that for GTS there should be a nomenclature change, along the lines of both phenotypes and aetiologies. A nomenclature change has occurred in many disorders once thought to be unitary or homogeneous and which are now realised to be heterogeneous and divided into types, such as diabetes and another movement disorder, Parkinson’s Disease (PD). Diabetes has a clear type 1 and type 2. Much the same has happened with respect to PD as it once thought to be a single entity that now clearly looks to have a variety of causes.

In fact, many now refer to it as *Parkinson’s Diseases*. It is now recognised that there is more than one type of PD, and one simple classification may be into Type 1 and Type 2. Type 1 could be seen as the presumed idiopathic PD type (late onset, no clear evidence of a genetic aetiology) and Type 2 which is familial PD (early onset, genetic, often Mendelian inheritance, heterogeneous and currently classified by the gene involved (PARK 1–12). The majority of PD cases are sporadic and of unknown origin, but several genes have been identified that, when mutated, give rise to the rare, familial forms of the disease. It may therefore be argued that there are more than just two types, while further studies have indicated that the genetic basis may, however, be even more complex. Thus, as with GTS, the many phenotypes and aetiologies (both genetic and non-genetic) of PD are more complex than was once thought. It was thus suggested that GTS, as has occurred with many disorders including diabetes and PD, in the future, after research, will be divided into a variety of sub-types. These will be designated according to the replicated factors from the factor analytic studies, and are highly likely to also make clinical sense. The only factor which has been replicated, several times in fact, is that of a “pure simple motor and phonic tic factor”, and which all specialist clinicians treating patients with GTS are able to identify clinically (Robertson, 2008b).

It was therefore suggested (Robertson, 2008b) that Type 1 GTS (GTS) should be patients with simple motor and phonic/vocal tics only. Other subtypes may follow, and it is likely that these will mirror the clusters and factors defined by the studies: for example GTS + OCD, or GTS + ADHD, or GTS + OCD + ADHD. In future studies it is hoped that these clinically determined factors and subtypes may have different aetiological bases such as genetic, immunological and pre- or peri-natal bases, as suggested by Robertson and Eapen (2013).

It is suggested that what constitutes the GTS phenotype(s) would become clearer only when the GTS gene(s) are identified and other aetiological mechanisms are fully understood. It will also be clearer as to what psychopathologies as well as tic phenomena are important in the various phenotypes, and whether or not these are related to the various aetiologies. An example in this context would be the dementias (particularly Pick’s disease) and how their nosology has changed over the years via an iterative process. Only then will the true prevalence be able to be investigated and understood (Robertson, 2008b).

This suggested future nomenclature for GTS could allow more precise clinical and/or aetiological descriptions of GTS. It is acknowledged that these suggestions are perhaps slightly premature based on the available information. For example, in contrast to PD, there are no clearly delineated genetic factors or age distinct differences in GTS. Hence, a definition based solely on phenomenology and comorbidity may be ultimately misleading. Nevertheless in the future, when the aetiologies and phenomenological subtypes may be more established, a sub-type nomenclature may be appropriate (Robertson, 2008b).

9. Critical comments and appraisal DSM-5 and its predecessors, taking current knowledge into account

The DSM APA criteria have changed over time and the main areas are as follows:

- (i) We would first like to challenge all the DSM criteria for naming GTS a “Disorder”. The same applies for ICD classification as well, in that ICD-10 mentions that “the term “disorder” is used throughout the classification, so as to avoid even greater problems inherent in the use of terms such as “disease” and “illness”. In both classificatory systems the use of the term “Disorder” is not an exact term, but rather it is used to imply the existence of a clinically recognisable set of symptoms or behaviour associated in most cases with distress and with interference with personal functions. Social deviance or conflict alone, without personal dysfunction, should not be included in mental disorder as defined here. In this regard, whilst GTS is clearly characterised by a clinically recognisable set of symptoms namely tics, for many individuals with GTS, this does not cause distress or dysfunction and hence the term “syndrome” may be preferable over “disorder”. Similarly, the use of the word “illness” in criteria a) (“...tics have been present at some time during the illness”) is also problematic in that the term illness assumes a subjective distressing state, and illness philosophically also suggests suffering whereas “disease” implies abnormal functioning and often associated with a clear aetiology and pathology [e.g. an infection (TB, streptococcus) as well as observable or testable condition (e.g. diabetes, high cholesterol, high blood pressure)].
- (ii) The DSM criteria over time have changed the upper age limit (under which age GTS must begin/start) suggesting 15, 21 and 18 years, which to us seems somewhat arbitrary and without any absolute scientific basis. The main characteristic of GTS is that it has its onset during childhood or adolescence and it reduces in severity rather than increase with age (e.g. Pappert et al., 2003; Robertson, 2012).
- (iii) In DSM-IV, the following major change was added “the disturbance causes marked distress or significant impairment in social, occupational, or other important areas of functioning”, but met with much opposition (e.g. Freeman et al., 1995; Comings, 1995; Kurlan et al., 1997). Many clinicians and experts (particularly working in genetic studies with large families) would see multiply affected pedigrees (e.g. Robertson and Gourdie, 1990), where very few individuals even realised they even had tics and were certainly not distressed or impaired by them. In addition many of us know, for example, friends or colleagues who certainly have GTS, but are not distressed or impaired and in fact are functioning perfectly well. Similarly, some members of the family who are mildly affected may be unaware that they have GTS despite having a severely affected family member.

- (iv) In DSM-IV and DSM-IV-R it was also stated that “during the (one year) there was never a tic-free period of more than 3 consecutive months”; there seemed to be no evidence base for this and thus, yet another arbitrary criterion had been introduced.
- (v) DSM-IV as we have said came under criticism leading to changes in DSM-IV-TR and two of the three above criteria were eliminated. Many researchers, in the intervening periods, used DSM-111-R or other standard (e.g. WHO, ICD-10 (1992/3) criteria or those of the “Tourette Syndrome Classification Study Group” (1993). The criterion “during the (one year) there was never a tic-free period of more than 3 consecutive months” remained in DSM-IV-R but has been removed from DSM-5.
- (vi) We would suggest a change in nomenclature from “vocal” tics to “phonic” or indeed “vocal/phonic” tics. There are undoubtedly some “true” vocal tics (e.g. words, humming, phrases) but sniffing, snorting, etc. are nasal in origin and clicks usually emanate from the tongue and palate (after being joined).
- (vii) Recognition of tic as a rapid, sudden, recurrent, non-rhythmic motor movement or vocalisation that is distinct from the stereotypies. Tics tend to be quick movements that last for only brief periods while stereotypies are often slow, unhurried and more coordinated. Further, tics vary in severity and site over time, whereas stereotypies often tend to remain unchanged in site and pattern (Eapen et al., 2013).
- (viii) In DSM 5, the term *transient* tic disorder has been replaced by *provisional* tic disorder for tics that have been present for less than a year.
- (ix) In the usual clinical practice, and as included in The National Hospital Interview Schedule (Robertson and Eapen, 1996), the presence of tics is determined “By examination” or “By history” which is similar to the Tourette Syndrome Study Group Criteria viz.: “(vi) motor and/or vocal must be witnessed by a reliable examiner directly at some point in the course of the disorder or be recorded by videotape or cinematography (for definite TS, A-1) or (for tics not witnessed by a reliable examiner), tics must be witnessed by a reliable family member or close friend, and description of tics as demonstrated must be accepted by reliable examiner (for TS by history, A-2)”.
- (x) National, legal, billing (financial) and other influences on the DSM criteria and ICD criteria and vice versa.

In the authors opinion, there is considerable variability in the use of the two main diagnostic and classificatory systems across different parts of the world. It appears that internationally the major discrepancies are between (i) clinical work and documentation and (ii) research criteria and (iii) the requirements for the reimbursement sources (e.g. from insurance companies), which influence documentation. For example some countries use the ICD for clinical purposes and DSM for research (e.g. Hungary), some use ICD for all purposes (e.g. India), some use DSM for all purposes as detailed below (personal communication). Specifically, it appears that in the USA the dependence on recognised diagnostic codes for reimbursement influences the apparent medical care provided in both private practice and public psychiatry. Insurance companies and federal insurance plans have explicitly validated some DSM diagnoses for services. DSM criteria have become linked (albeit it at an uncomfortable level) to billing. For example 293.9 is a “Mental Condition due to Diabetes” or a major or minor neuro-cognitive deficit due to diabetes may be billed (paid for), whereas mild neurocognitive impairment would not be reimbursed. In some cases in psychotherapy, adjustment disorders have been over-used as a gentler and less specific diagnosis to reduce stigma of seeking

treatment. In times gone past, insurance did not pay for Axis II diagnoses (e.g. Personality Disorders) for inpatient care. Medicare only recognised Axis I diagnoses for reimbursement. So, for example, Borderline Personality Disorder (BPD) was not recognised for reimbursement, whereas BPD plus a mood disorder was reimbursed, for the treatment of the mood disorder. In this manner, the accuracy of psychiatric disorder prevalence data has been distorted by selective (rather than comprehensive) reporting of psychiatric conditions.

In addition, many insurance companies linked approval for psychotropic medications to uses or diagnoses for which there was Food and Drug Administration (FDA) approval. From this documentation requirement, the habit developed to list only the primary diagnosis that corresponded to a justification for a particular medication. For example, depression, anxiety, or ADHD might be the diagnosis of/on record, while the underlying reason and co-morbid condition of GTS might not be recorded, other than in clinical notes. These policies continue to affect prescribing practices, both by increasing paperwork justification of rationale for off-label uses of medications, and influencing slips of diagnostic accuracy (in terms of adherence to written diagnostic criteria).

Approximately 20 years ago GTS was not billable (i.e. paid for by insurance companies), whereas GTS + ADHD was billable: so at that time, it may have been “prudent” to give a combined diagnosis. However, apart from clonidine/guanfacine there were no other medications which were used for GTS and ADHD independently (see FDA requirements above).

In addition the insurance companies wished that the Food and Drug Administration (FDA) approved medication matched the diagnosis. In this regard, what was the “primary” disorder (e.g. GTS) was different from that which was billed for (e.g. depression, anxiety ADHD).

We understand that reimbursement for psychiatric services will be linked to DSM-5 by 2015, at the same time that all medical diagnostic billing in the US will be required to use ICD-10 (until now, the US has continued to use ICD-9 diagnostic codes and criteria). Some practices, institutions, and universities are converting to using DSM-5 criteria now in preparation for the federal government required deadlines. In Canada we understand that billing is single-payer and not by insurance companies and at present ICD-9 codes (3 digits only) are used, without any specifiers for all medical and surgical disorders except in psychiatry where DSM-criteria is used for clinical diagnosis, medico-legal, and insurance purposes.

Research usage may vary on the other hand from using ICD-9 3-digit or full-digit codes, or DSM depending upon the funding granting agency or the anticipated publication requirements.

In the author’s experience in the UK, by far the majority use ICD-10 for clinical and medico-legal purposes. In the UK there is the NHS as well as the private or independent sector. In the NHS there is a complex system of contra-funding and payment. There are the Commissioners, the Primary care Trusts (PCTs) and local Hospitals (NHS Trusts). In all NHS patients’ electronic patient records, an ICD code *must* be entered. Moreover, each NHS Trust/hospital has to report the proportion of patients who have recorded ICD diagnosis/es to the Commissioners: this is one of the key “Performance Indicators” that they have to deliver data on – in order to obtain funding each year. In clinical letters to general practitioners (GPs) some also use multi-axial coding. While it is not necessary to use any diagnostic coding for physical health or co-morbidity, most clinicians use ICD for hospital records, billing and insurance purposes, whilst using DSM for research. In this regard, ICD “hyperkinetic disorder” may be used for clinical purposes while the term “ADHD” may be used for research, in order to fit in with the bulk of published data and to publish in American or international journals.

In most European countries, ICD is used for clinical, insurance, billing and medico-legal purposes, whilst DSM is used for research (personal communication).

In Australia, as per the author's experience, ICD is used for medical records in the hospital and it is the official classificatory system for coding purposes in clinical psychiatry. However psychiatrists and paediatricians use DSM for research purposes and also for the purpose of prescribing medications under the Pharmaceutical Benefits Scheme (PBS) to avail the medicines on subsidised rates. Billing in private practice is usually not based on the diagnosis or the classificatory system but rather on the nature (complexity) of the problem, the practice setting, new assessment or follow up, time spent on consultation, etc.

In most Asian countries, ICD-10 criteria is used in all spheres, both clinical and research. Hence the changes in DSM 5 in the case of GTS will not be having any significant impact in terms of clinical practice. However the changes would be relevant for research purposes as DSM 5 is used in the context of research in some Asian countries for ease of comparison with other international studies.

10. Summary of changes in DSM-5 and conclusions

The main changes in DSM-5 are summarised below. GTS, referred to as Tourette's disorder in DSM5 is included in the chapter on "neurodevelopmental disorders" under the section of "Motor Disorders", alongside developmental coordination disorder, stereotypic movement disorder, persistent (chronic) motor or vocal tic disorder, provisional tic disorder, other specified tic disorder, and unspecified tic disorder.

We agree with the inclusion of GTS as a "Neurodevelopmental Disorder" where it is alongside/after (i) Intellectual Disabilities (ii) Communication Disorders (iii) Autistic Spectrum Disorder (ASD) (iv) Attention Deficit/Hyperactivity Disorder (ADHD) as all these disorders have a neurobiological origin and share the onset during the developmental period. In this regard, it is noteworthy that there have been some concern that TS was "designated" a "Mental Disorder" (by being included in DSM), rather than a "neurological disorder", which according to some, may have been preferable, for a variety of reasons.

Some of the specific changes in DSM-5 are highlighted here. The word *stereotyped* has been removed from the definition of tics and the rationale for this comes from the need to distinguish tics from the stereotypies commonly encountered in ASD and related conditions. Further, the requirement that 'there is no tic-free period of 3 months or more' has been removed. The term *transient tic disorder* has been replaced with *Provisional tic disorder* because *transient* can only be defined retrospectively and hence not very useful to the clinician when the tics have been present for less than a year. The term *provisional* accounts for those initially presenting with tics which may eventually last for more than one year and thereby satisfy the duration criteria for chronic tic disorder or Tourette Disorder subsequently. The term *Persistent (Chronic) Motor or Vocal Tic Disorder* is used when motor or vocal tics (but not both) have been present for more than one year duration. Thus DSM-5 has also added a specifier chronic motor or vocal tic disorder to distinguish between vocal and motor tics that are chronic. The specific reference to "stimulant" use as a cause has been removed as there is no evidence base to support this but the criteria that the symptoms are not due to taking medicine or other drugs is retained. A new category of "Other specified" and "Unspecified" has been added to refer to tic disorders that result in significant impairment to the individual yet do not meet the full criteria for other tic disorders. Examples of these include tics with onset in adulthood, or tics triggered by other medical conditions (Eapen et al., 2002).

We would suggest adding additional features which would help improve the diagnostic confidence such as echo-phenomena, copro-phenomena, palilalia, etc. and the presence of premonitory urges, tic suppressability, suggestibility, waxing and waning (as documented in the Diagnostic Confidence Index [Robertson et al., 1999]). The second would be to include the spectrum of behaviours that co-occur with tics under the "Tic Spectrum disorder" such as OCD with tics, ADHD with tics and ASD with tics, etc. (Eapen and Crncec, 2014). It remains to be seen as to how enthusiastically clinicians will take up the new criteria and how this will influence both clinical practice and research over time.

Conflict of interest

MMR has conflicts of interest through her links with the Tourette Syndrome Association International Genetics Consortium (TSAIGC), Tourette Action UK & European Society for the Study of Tourette Syndrome (ESSTS). VE reports no conflict of interest.

Acknowledgements

We would like to thank all our colleagues who participated in our survey by answering questions and/or participated in fruitful discussions during the preparation of this manuscript.

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