

1-1-2020

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Black, Kevin J, "Case Report: DSM-5 misses an edge case in tic disorders nosology." F1000Research. 9, 505 (2020).

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CASE REPORT

Case Report: DSM–5 misses an edge case in tic disorders nosology [version 1; peer review: 2 approved]

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V1 **First published:** 03 Jun 2020, 9:505
<https://doi.org/10.12688/f1000research.23991.1>
Latest published: 03 Jun 2020, 9:505
<https://doi.org/10.12688/f1000research.23991.1>

Abstract

A boy with multiple phonic tics, one lifetime motor tic, and no impairment or marked distress does not meet criteria for any DSM–5 tic disorder diagnosis. The next version of the Diagnostic and Statistical Manual should adjust the criteria for Tourette's Disorder and/or for "other specified tic disorder" and "unspecified tic disorder."

Keywords

Tourette syndrome, tic disorders, nosology, DSM-5, case report



This article is included in the **Tics** collection.

Open Peer Review

Reviewer Status

	Invited Reviewers	
	1	2
version 1 03 Jun 2020	 report	 report

- 1 **Andrea Cavanna** , University of Birmingham, Birmingham, UK
- 2 **Carol Mathews** , University of Florida, Gainesville, USA

Any reports and responses or comments on the article can be found at the end of the article.

Corresponding author: Kevin J. Black (kevin@wustl.edu)

Author roles: **Black KJ:** Conceptualization, Funding Acquisition, Investigation, Writing – Original Draft Preparation, Writing – Review & Editing

Competing interests: Research support: NIH and Emalex Biosciences. Speakers bureaus within the past year: Acadia Pharmaceuticals and Teva Pharmaceutical Industries Ltd.

Grant information: This work was supported in part by the U.S. National Institutes of Health (NIH) [R01MH104030, UL1RR024992, UL1TR000448].

The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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How to cite this article: Black KJ. **Case Report: DSM–5 misses an edge case in tic disorders nosology [version 1; peer review: 2 approved]** F1000Research 2020, 9:505 <https://doi.org/10.12688/f1000research.23991.1>

First published: 03 Jun 2020, 9:505 <https://doi.org/10.12688/f1000research.23991.1>

Introduction

Over time, tic disorder nosology has changed¹. The current research criteria changed modestly with the adoption of DSM–5². I describe a case of a child with motor and vocal tics that demonstrates a gap in DSM–5’s diagnostic criteria for tic disorders³.

Case report

At age 9, a right-handed, non-Hispanic white boy and his mother participated in a thorough research assessment as part of the New Tics study⁴. The study was approved by the Washington University in St. Louis Human Research Protection Office, IRB ID #201109157, and his mother gave informed consent. This visit included questionnaires, K-SADS parent and child interviews, history of illness, neurological exam, 30 minutes of observation of the child alone via video, and YGTSS rating. His mother was a reliable informant, an elementary school teacher well informed about tics, and his father was a physician. His mother dated his tic onset to 8 months ago, at age 8. The child had seasonal allergies but the phonic tics were present when he had no allergy symptoms, and the tics did not respond to cetirizine. He was taking extended-release mixed amphetamine salts (40 mg daily) for ADHD with good response. K-SADS diagnoses were specific phobia, past social anxiety disorder, past nocturnal enuresis, predominantly inattentive ADHD since age 6, and provisional tic disorder.

Neurological exam was normal except for a medium-loud snort occurring once during the exam. He had simple phonic tics (sniff or snort, cough, clear throat), one motor tic (biting lower lip softly, seen during video observation) and no complex tics. He bit his nails sometimes since early childhood, but this was not counted as a tic given the timing and its high prevalence among young children⁵. He also had one probable simple motor stereotypy (rarely shook his hands up and down near his chest before sports or social events since age 5 or younger; he said “I like doing that” and said it didn’t feel like his tics; seen only once in over an hour of observation). YGTSS scores were: motor tics 5, phonic tics 12, impairment 10.

He returned at 12 months after tic onset. The stimulant continued at the same dose, now without an antihistamine. The same tics continued within the past week, though not every day, but no tics were observed at the visit. YGTSS scores were: motor tics 4, phonic tics 6, impairment 0. By 24 months after tic onset, he was taking no medications. The lip biting had disappeared but the phonic tics continued. He reported that “they’re kind of annoying and I would like them to go away,” but he did not have marked distress and the tics did not affect self-esteem, family life, friendships or school functioning. No tics were observed during thorough history and a neurological examination, but sniffing, coughing and forceful nasal exhalations were observed by video when he was alone in the room. No motor tics were observed. YGTSS scores were: motor tics 0, phonic tics 9, impairment 0. Diagnostic Confidence Index score was 35⁶.

Discussion

This boy has a fairly typical history for mild Tourette syndrome, except that he has only one motor tic. (Other clinicians may choose to count the nail biting or hand shaking as tics, but for the present discussion the main point is that some children will have a presentation with vocal tics and one motor tic.) The DSM–5 criteria for Tourette’s Disorder require “multiple” motor tics². The criteria for Persistent (Chronic) Vocal Tic Disorder exclude patients who have experienced both motor and vocal tics. At the follow-up visits, the duration of ticcing excludes Provisional Tic Disorder, and the history and examination provided no evidence for causation by a substance or non-psychiatric illness. The residual categories, Other Specified Tic Disorder and Unspecified Tic Disorder, require “clinically significant distress or impairment in social, occupational, or other important areas of functioning.” This last criterion is shared with most DSM–5 disorders, but since DSM–IV–TR it has been omitted for Tourette’s Disorder⁷. Thus this boy does not meet DSM–5 criteria for any tic disorder. Woods and Thomsen addressed the situation in which a patient has vocal tics and exactly one motor tic, and concluded that “the requirement that multiple motor tics exist seems arbitrary and unnecessarily exclusive”⁸. A DSM–5 work group discussed the nosological issues in detail⁷. They retained the “multiple” motor tic requirement for Tourette’s Disorder, and did not propose reinstating the impairment or distress criterion in the “not otherwise specified” diagnosis. Roessner and colleagues provided critical feedback on the proposed criteria for tic disorders, but also assumed the impairment or distress criterion would be absent for all tic disorders⁹. This change appears to have been inadvertent.

Conclusions

There is no clinical import for this child, as his symptoms bother him only slightly. But this case demonstrates that the current DSM–5 criteria inadvertently provide no diagnosis in this case, which may occasionally affect research on tic disorders. The exclusion of one motor tic from both Tourette’s Disorder and Persistent Vocal Tic Disorder leaves a gap. The residual diagnostic categories no longer cover this gap since DSM–5 requires the “impairment or distress” criterion for them, though that requirement may have been accidental. I propose that future revisions omit it for all tic disorders. I also agree with Woods and Thomsen’s opinion that one motor tic and multiple phonic tics is best described as Tourette syndrome.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Consent

Written informed consent for publication of their clinical details was obtained from the parent of the patient.

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Current Peer Review Status:  

Version 1

Reviewer Report 17 June 2020

<https://doi.org/10.5256/f1000research.26466.r64376>

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Carol Mathews 

Department of Psychiatry, Genetics Institute, University of Florida, Gainesville, FL, USA

This case report discusses a 9 year old boy who presents with persistent, multiple vocal tics, and one motor tic. Based on history and physical exam, he clearly meets criteria for multiple chronic tics, but has no demonstrated impairment. I would argue that he has mild distress, as evidenced by the fact that he finds them annoying, but from a practical perspective, they do not functionally impact his life. The author argues that this child's tics would not fit into any of the current DSM-5 tic disorder classifications, and most specifically, that he would not fit into either of the two that are closest to his presentation--Tourette's Disorder, because he does not have multiple motor tics, or persistent vocal tic disorder, because he does have one motor tic. This highlights a gap in the current diagnostic criteria (even leaving out the question of whether impairment should be necessary for a diagnosis, which is also addressed). Genetic and emerging clinical data suggest that chronic tics, whether primarily motor,vocal, or a mix of both, emerge from the same underlying genetic causes, and thus are likely to be manifestations of the same disorder. The argument is made, appropriately, for a revisiting of the tic disorder diagnoses in the DSM, to account for this gap, and perhaps also to incorporate the current genetic information when determining the disorder definitions for the next update.

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Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Tourette syndrome genetics, OCD genetics

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response (F1000Research Advisory Board Member) 17 Jun 2020

Kevin J Black, Washington University in St. Louis School of Medicine, St. Louis, USA

I appreciate Dr. Mathews's thoughtful input, including the 2 recent references, and agree with her comments about the likely nosological unity of the various DSM-5 chronic tic disorders including TS.

Competing Interests: No competing interests were disclosed.

Reviewer Report 11 June 2020

<https://doi.org/10.5256/f1000research.26466.r64373>

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Andrea Cavanna 

Department of Neuropsychiatry, BSMHFT, University of Birmingham, Birmingham, UK

Kevin Black's case report highlights a possible hole in the current classification scheme for tic disorders. The author describes the clinical presentation of a 9-year-old boy who has a fairly typical history for mild Tourette syndrome, except that he has only one motor tic. Since he does not have an impairment or marked distress, he does not meet current diagnostic criteria for any DSM-5 tic disorder diagnosis. This interesting report is useful, as it hints at two possible solutions, not mutually exclusive. Firstly, it would be appropriate to consider relaxing the current criteria for the categories "Other Specified Tic Disorder" and "Unspecified Tic Disorder", by reviewing the persisting criterion of "clinically significant distress or impairment in social, occupational, or other important areas of functioning". Secondly, on a more general level, this case report calls into question the rationale for the distinction between motor and vocal/phonic tics. It is hoped that future classification systems will be increasingly more informed by advances in research that would allow them to "carve nature at its joints".

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Behavioural neurology and neuropsychiatry

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response (F1000Research Advisory Board Member) 11 Jun 2020

Kevin J Black, Washington University in St. Louis School of Medicine, St. Louis, USA

I thank Dr. Cavanna for the review. I agree with his comment about the artificial and historical nature of the distinction between motor and vocal tics.

Competing Interests: No competing interests were disclosed.

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