



## BRIEF REPORT

# REVISED Functional tic-like presentations differ strikingly from Provisional Tic Disorder [version 2; peer review: 2 approved]

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## Abstract

**Background:** Recent years have seen a dramatic increase in new “tic” cases in teens and young adults. These individuals often present with fulminant onset of symptoms not commonly seen in Tourette syndrome (TS) and are often diagnosed with Functional Neurological Symptom Disorder (FND-tic). However, some authors have questioned whether this illness truly differs from typical Provisional Tic Disorder (PTD) and TS. Previous studies have compared FND-tic, usually a few months after symptom onset, to patients with TS, usually years after symptom onset. We sought to test whether the presenting symptoms of FND-tic differ substantially from those in patients at a similar duration of symptoms who are later diagnosed with TS.

**Methods:** This comparative study examines clinical features summarized from published reports of FND-tic with novel data from a longitudinal study of PTD. This study came from a referral center for TS and tic disorders and included 89 children with tics whose first tic occurred a median of 3.6 months earlier, nearly all of whom were diagnosed with a chronic tic disorder at follow-up. Specifically, we examine clinical features identified in a recent literature review as supporting a diagnosis of FND-tic, including symptom characteristics, course, severity and comorbidity.

**Results:** Several clinical features dramatically distinguish the patients diagnosed with FND-tic from those diagnosed with typical PTD. For example, coprophenomena are reported at or shortly after symptom onset in over half of FND-tic patients, whereas even several months after onset, coprophenomena had occurred in only 1 of 89 children with PTD. Six clinical features each have a positive predictive value over 90% for FND-tic diagnosis if prior probability is 50%.

**Conclusions:** These new data provide strong evidence supporting the diagnostic validity of FND-tic as distinct from TS.

## Open Peer Review

Approval Status

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2. **Tammy Hedderly** , King's College London, Guy's & St Thomas' NHS Foundation Trust, London, UK

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

## Keywords

Tic Disorders/classification, Provisional Tic Disorder, Functional Neurological Symptom Disorder, Conversion Disorder, Diagnosis, Differential, Tourette Syndrome



This article is included in the [Tics](#) collection.

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**REVISED Amendments from Version 1**

In this revision, we address the thoughtful comments of the reviewers. Specifically:

- \* In the discussion, we expand on the limitations of this study, including likely differences between groups in ascertainment bias.
- \* We add statistical tests to Table 1 and add sensitivity and specificity as new columns in Table 2.
- \* We supply new, individual participant data for the PTD sample as supplementary files added to the same dataset referenced in version 1 (doi 10.17605/OSF.IO/XZ328).
- \* We add context for the test statistics including the reliance of PPV on prior probability.
- \* We discuss the applicability of our results to the situation in which a child presents with both typical-appearing tics and apparent functional tic-like symptoms.
- \* We note that features other than those studied here are likely important to diagnosing FND-tic.

**Any further responses from the reviewers can be found at the end of the article**

## Introduction

Recent years have seen a dramatic increase in new “tic” cases in teens and young adults.<sup>1,2</sup> These individuals often present with fulminant onset of symptoms not commonly seen in Tourette syndrome (TS), but often similar to those found in videos tagged as “Tourette” on social media.<sup>3</sup> For instance, echopraxia and coprolalia occur in more than half of these individuals at symptom onset.<sup>3</sup> The nature and characteristics of these symptoms, and the onset age and course of illness have led experienced clinicians to differentiate these cases from TS and to diagnose instead Functional Neurological Symptom Disorder (FND). FND with tic-like symptoms (hereinafter “FND-tic”) has been reported previously, but prior to 2019 was considered rare, occurring in < 2% of tic or tic-like cases at five major referral centers from three continents.<sup>4</sup>

Some experts have expressed skepticism as to whether a new diagnosis (FND) is needed for these patients, positing that the previous understanding of TS may have been too narrow.<sup>5</sup> Perhaps, for instance, echopraxia is present early in the course of TS much more often than has been recognized. We concur with these authors that clearly differentiating a new diagnosis from existing diagnoses is a key component of diagnostic validity.<sup>6</sup> Substantial published data describe typical clinical features of chronic tic disorders, but almost no prospective data have been published on symptoms in the first year after tic onset.<sup>7,8</sup> This evidence gap is crucial because most FND-tic patients at clinical centers have had symptoms for only a few months (mean 0.4 years in one study<sup>4</sup>). Here, we directly address this concern by providing for the first time substantial data on relevant clinical features during the first few months after symptom onset in children ultimately diagnosed with TS.

## Methods

The New Tics study is a prospective, longitudinal study that enrolled 89 children ages 5–10 years whose first tic occurred in the past 9 months (median 3.6 months).<sup>9</sup> Children are assessed using multiple informants (child, parent, trained interviewer, and observation by an experienced clinician for more than an hour, including by video while the child is alone). The diagnosis in this situation is Provisional Tic Disorder (PTD), and nearly all these children (77 of 79) were diagnosed with TS (70) or a persistent tic disorder (7) when they returned at the one-year anniversary of their first tic.<sup>10</sup> Here we report the prevalence and timing from the New Tics sample of various features that occur commonly in FND-tic patients.

The feature list was drawn from a recent review of FND-tic, a narrative review that included all primary data publications on FND-tic known to its authors as of August, 2022.<sup>3</sup> The comparison data for FND-tic patients comes from 26 published reports, with pertinent data in 17 reports<sup>4,11–26</sup> describing a total of 336 patients (data file available as *Underlying data*<sup>27</sup>). For quantitative variables, the weighted mean is provided (weighted by *N* in each report), along with the median and range of the summary values reported in each relevant publication.

Most of these features thought to suggest FND-tic were recorded prospectively in the New Tics study, including age, sex, premonitory urges, tic suppression, coprophenomena, and family history. However, since the New Tics sample was enrolled almost entirely before the recent FND-tic upsurge, some of these clinical features were recorded indirectly. For instance, to match “severe symptoms at onset,” defined differently in various reports on FND-tic, we conservatively chose from the New Tics sample all patients with emergency department visits or disability prior to the screening visit, or

a high score on the Yale Global Tic Severity Scale's<sup>28</sup> impairment item at screening. Details on other such choices are given in footnotes to [Table 1](#).

### Data analysis

Fisher's exact test was used to find the probability of differences in frequency of features between the two populations (fisher\_exact from SciPy (RRID:SCR\_008058) 1.9.1).<sup>29</sup>

**Table 1. Comparison of various clinical features in FND-tic and in typical PTD.**<sup>a</sup> FND, Functional Neurological Symptom Disorder; FND-tic, Functional Neurological Symptom Disorder with tic-like symptoms; PTD, Provisional Tic Disorder; NP, number of publications; OCD, obsessive compulsive disorder; ADHD, attention deficit hyperactivity disorder; YGTSS, Yale Global Tic Severity Scale.

Feature	FND NP <sup>b</sup>	FND numerator/denominator (percentage)	FND median percentage (range)	New Tics <sup>c</sup> number (percentage)	p <sup>d</sup>
Sex (% female)	17	251/336 (75)	72 (20–100)	25 (28)	.000
Typical tic disorder diagnosis prior to current episode onset	11	33/150 (22)	15 (0–100)	20/124 (16) <sup>e</sup>	.185
Sudden, abrupt onset	10 <sup>f</sup>	136/142 (96)	100 (77–100)	15/75 (20) <sup>g</sup> or 46/62 (74) <sup>h</sup>	.000
Symptoms in extremities before face and neck	7	43/100 (43)	40 (15–100)	10 (11)	.000
Coprophomina at onset	8	68/115 (59)	54 (0–77)	1 (1)	.000
Tics involving the body or limbs without a history of tics involving the eyes, face, and head	7	29/84 (35)	18 (0–77)	10 (11)	.000
Premonitory urges present	9	56/118 (48)	60 (0–100)	85 (96)	.000
Severe symptoms at onset	6	82/172 (48)	77 (30–100)	3–5 (3–6)	.000
Extreme "attacks" of tic-like behavior	4	44/68 (65)	82 (36–100)	0–2 (0–2)	.000
Inability to suppress	10	74/120 (62)	70 (0–100)	20 (22)	.000
Tic-like phenomena are constant in severity over time rather than waxing and waning	5	50/75 (67)	68 (15–100)	51 (58)	.164
Movements or vocalizations that are dramatically worse in the presence of others <i>versus</i> when alone	3	15/32 (47)	50 (11–100)	0 (0) <sup>i</sup>	.000
Symptoms that dramatically and persistently disrupt the person's intended actions or communications	3	34/52 (65)	39 (36–89)	2 (2)	.000
Family history of tics	9	17/131 (13)	0 (0–60)	30 (34)	.000
Family history of OCD	1	1/22 (5)	5	14 (16)	.152
Family history of ADHD	1	6/22 (27)	27	25 (28)	.584
ADHD diagnosis before/at presentation	9	69/216 (32)	22 (0–48)	39 (43)	.050
OCD diagnosis before/at presentation	8	11/127 (9)	6 (0–23)	27 (30)	.000
Anxiety disorder prior to/at presentation	8	77/132 (58)	53 (11–100)	27 (30)	.000

**Table 1.** *Continued*

Feature	FND NP	FND weighted mean	FND median (range)	New Tics mean
Age of onset	15	22.3	16.5 (7.5–53.6)	7.6
Age at presentation	5	20.5	18.8 (11.2–36.3)	7.9
YGTSS <sup>1</sup> Total Tic Score (0-50)	2	32.7	32.4 (31.5–33.3)	16.9
YGTSS Impairment (0-50)	2	30.2	31.2 (28.6–33.8)	7.6
YGTSS Global Severity Score (0-100)	3	62.8	62.6 (61.9–65.3)	24.3

<sup>a</sup>Specified clinical features in patients with tic-like symptoms (“FND”) from the articles reviewed in Malaty *et al.* (2022),<sup>3</sup> compared to participants with typical Provisional Tic Disorder from the New Tics study (“New Tics”).

<sup>b</sup>NP = number of publications from the table in Malaty *et al.* (2022)<sup>3</sup> that contributed data to the statistics in this row.

<sup>c</sup>N = 89 except where numerator and denominator are provided.

<sup>d</sup>Fisher’s exact test; “.000” means < .0005; *p* values bolded if *p* < .05/20 = .0025.

<sup>e</sup>N = 124, the number of participants who came for a screening visit after reporting recent onset of tics during initial telephone contact. Some were found during careful screening to have a prior episode of transient tics.

<sup>f</sup>Excludes the two reports that defined the sample by sudden onset.<sup>4,21</sup>

<sup>g</sup>The investigator assigned the most likely symptom onset date within a date range (possible onset dates) of less than 7 days.

<sup>h</sup>From parents’ answers as to whether the onset of the first tic was sudden or gradual.

<sup>i</sup>Investigator noted tics during the history and exam (approximately 45 minutes), but no tics when observing the child alone in the room via video camera (approximately 40 minutes).

<sup>j</sup>Yale Global Tic Severity Scale.

**Ethical considerations**

The New Tics study was approved by the Washington University Human Research Protection Office (IRB, protocol numbers 201109157 and 201707059), all participants assented to participation, and a parent or other legal guardian provided written documentation of informed consent.

**Results**

Stark differences in presentation distinguish the FND-tic patients from typical PTD (Table 1). For example, coprophomina are reported at or shortly after symptom onset in 59% of FND-tic patients. By contrast, coprophomina had occurred in only 1 of 89 children with PTD at an average of 3.6 months after tic onset. Similarly, the TS International Database Consortium found that only 2% of TS patients in tertiary centers retrospectively reported coprophomina at symptom onset, and only 20% ever manifested coprophomina by an average of 5 years after tic onset.<sup>30</sup> Movements or vocalizations that were dramatically worse in the presence of others *versus* when alone occurred in 47% of FND-tic patients, but in none of the New Tics PTD sample. Symptoms dramatically and persistently disrupted intended actions in 65% of FND-tic patients, but in only 2% of PTD. The prevalence of prolonged tic attacks was 65% in FND-tic, but 0–2% in PTD. Other features that differed substantially include lack of premonitory urges (53% *vs.* 4%) and severe symptoms at onset (48% *vs.* 3–6%). Table 1 provides details on these comparisons and includes statistics on a dozen more clinical features of FND-tic that differ from the New Tics PTD sample.

**Table 2. Diagnostic utility of the binary features in Table 1.**<sup>a</sup> PPV, positive predictive value; NPV, negative predictive value; OCD, obsessive compulsive disorder; ADHD, attention deficit hyperactivity disorder; TS, Tourette syndrome; FND, Functional Neurological Symptom Disorder.

Feature	Sensitivity	Specificity	PPV (NPV)	prior 2%	prior 50%
Movements or vocalizations that are dramatically worse in the presence of others <i>versus</i> when alone	47%	100%	PPV =	<b>100%</b>	<b>100%</b>
Coprophomina at onset	59%	99%	PPV =	52%	<b>98%</b>
Coprolalia at presentation	49%	99%	PPV =	47%	<b>98%</b>
Symptoms that dramatically and persistently disrupt the person’s intended actions or communications	65%	98%	PPV =	38%	<b>97%</b>
Extreme “attacks” of tic-like behavior	65%	98%	PPV =	38%	<b>97%</b>
Premonitory urges present	47%	4%	<b>NPV =</b>	19%	<b>92%</b>
Severe symptoms at onset (defined variously in different studies)	48%	94%	PPV =	15%	<b>89%</b>

**Table 2.** *Continued*

Feature	Sensitivity	Specificity	PPV (NPV)	prior 2%	prior 50%
Sudden, abrupt onset (NewTics: onset confidence window < 7 days)	96%	80%	PPV =	9%	<b>83%</b>
Symptoms in extremities before face and neck	43%	89%	PPV =	7%	<b>79%</b>
Tics involving the body or limbs without a history of tics involving the eyes, face, and head	35%	89%	PPV =	6%	<b>75%</b>
Inability to suppress	62%	78%	PPV =	5%	<b>73%</b>
Female	75%	72%	PPV =	5%	<b>73%</b>
Anxiety disorder prior to/at presentation	58%	70%	PPV =	4%	66%
Family history of tics	13%	66%	<b>NPV =</b>	3%	57%
OCD diagnosis before/at presentation	9%	70%	<b>NPV =</b>	3%	57%
Typical tic disorder diagnosis prior to current episode onset	22%	84%	PPV =	3%	58%
Sudden, abrupt onset (NewTics: per parent tic survey)	96%	26%	PPV =	3%	56%
ADHD diagnosis before/at presentation	32%	57%	<b>NPV =</b>	2%	54%
Tic-like phenomena are constant in severity over time rather than waxing and waning	67%	42%	PPV =	2%	53%
Family history of OCD	5%	84%	<b>NPV =</b>	2%	53%
Family history of ADHD	27%	72%	<b>NPV =</b>	2%	50%

<sup>a</sup>PPV of a non-TS diagnosis for the binary features in Table 1, assuming a prior probability for FND of 2% (typical pre-pandemic prevalence at a referral center<sup>4</sup>) or 50% (representing clinical equipoise about a given patient’s diagnosis before considering this feature). NPV is shown for features more common in typical TS, equivalent to PPV for the absence of the given feature. Note: Values for PPV and NPV are bolded if ≥67%.

Table 2 illustrates the clinical value of these comparisons, viewing each clinical feature as a “test” for FND-tic, with its associated sensitivity and specificity. We also provide the positive predictive value (PPV) for each feature thought to indicate FND-tic. The PPV represents how confident one can be of a FND-tic diagnosis given the presence of the listed feature. The PPV depends on the prior probability of the FND-tic diagnosis, or its likelihood before one knows whether a given patient has that feature. We provide PPV for each feature based on two prior probabilities, 2% and 50%. The first, 2%, represents the approximate rate before the pandemic of functional tic-like symptoms at clinical referral centers.<sup>4</sup> Clinical equipoise about a given participant’s diagnosis is represented by a prior probability of 50%, for instance if one knows that a referring clinician is ambivalent about whether the patient has PTD or FND-tic, but one has not yet read the chart nor seen the patient. Features thought to be less common in FND-tic are listed with a negative predictive value (NPV), equivalent to PPV for the absence of the given feature.

In a patient with recent onset of tics, coprophenomena at onset, or any one of the other features named above, raises the probability of a non-TS diagnosis from 50% (as when the clinician is ambivalent about the diagnosis prior to considering this feature) to over 90% (Table 2). Other features differ significantly but are less useful diagnostically. For instance, obsessive compulsive disorder (OCD) is more than three times less common in FND-tic than in PTD ( $p < .0001$ ), but its absence only raises the probability of FND-tic from 50% to 57%.

**Discussion**

We demonstrate conclusively that patients with functional tic-like symptoms differ notably from typical tic patients at the same stage of the disorder, namely in the first few months after symptom onset. Previous reports have compared FND-tic to TS,<sup>3,31</sup> but not to a large PTD sample. We also provide for the first time quantitative estimates of the diagnostic significance of individual clinical features previously suggested to indicate FND-tic. This approach addresses a current debate<sup>5,32</sup> by showing that some features are significantly more common in patients diagnosed with FND-tic, yet individually do not substantially raise the likelihood of an FND-tic diagnosis.

The primary concern with the validity of these conclusions arises from the different potential sources of ascertainment bias in the two groups. Fortunately, these differences may not be as problematic as one might suppose. The FND-tic group is older, and if one’s information were limited to this study alone, one might posit that the natural history of tic disorder included different early symptoms at different ages of tic onset. However, the literature includes decades of previous clinical information on typical tic disorders. Retrospective studies of TS and a prospective study of PTD in siblings of TS

probands all found peak tic onset before age 10 years old; adult onset of tics is quite uncommon.<sup>7,8,33</sup> The PTD cases were volunteers willing to participate in a rather intensive research study, and were ascertained by a variety of methods (advertising and referrals from clinicians being the most common) rather than by clinical care-seeking. However, at the screening visit for the study, over 60% of them had sought clinical care for the tics or were planning to. The two FND-tic reports from Calgary, Alberta (child and young adult) were from a prospective registry of all patients seen for tics at their center, which is the only specialty center in their region. FND-tic cases were defined by “rapid onset of complex tic-like behaviors, with escalation to peak severity within hours to days.” In other words, these reports are selected only for being seen at a specialty center for tic-like phenomena. Yet their clinical features are similar to those of the FND-tic literature overall (or even more different from the PTD group). For instance, 95% of the Calgary FND-tic child patients were of female sex, only 25% had ADHD, and only 5% had OCD. We can conclude that potential bias from selecting more interesting, severe or classic cases for published case series does not substantially alter our results. Perhaps most importantly, there are no larger published sources for data about FND-tic nor about PTD. Nevertheless, ideally the results presented here should be confirmed in a new, independent sample.

We note that 22% of the patients reported in the FND-tic papers in fact had a prior history of typical tics, and many of them exhibited both tics and functional tic-like symptoms at the time they presented due to the latter. We have structured our results around whether FND-tic can be diagnosed, not whether PTD can also be diagnosed. In other words, when a patient presents with a typical history for TS but also new symptoms, the features above would allow more confidence in diagnosing FND-tic in addition to TS.

Other features in addition to those studied here may also be important for diagnosis. For instance, exposure to tic-like symptoms on social media was a common feature discussed in the papers discussing FND-tic in the past few years.<sup>3</sup> Unfortunately, we have no prospectively collected data from the PTD group on exposure to others with similar symptoms.

The data presented here do not prove the etiology of the tic-like symptoms diagnosed in the cited reports; hence the limited claim that these symptoms represent a different illness than PTD/TS. However, the marked difference in presentation these data demonstrate is an important argument adduced in the cited reports to support the diagnosis of functional neurological symptom disorder. Diagnosing FND-tic is important, since to the extent of our current knowledge, its prognosis and optimal treatment differ from those of TS.<sup>3</sup>

In conclusion, these new clinical data about the first few months after tic onset prior to diagnosis of TS provide strong evidence supporting the diagnostic validity of functional tic-like symptoms as distinct from PTD and TS.

## Data availability

### Underlying data

Information on and individual subject data from ‘The New Tics Study: A Novel Approach to Pathophysiology and Cause of Tic Disorders’ can be found at [NIH RePORTER](#) (Project Number 1R01MH104030-01A1) and at the [NIMH Data Archive](#).

Open Science Framework: Supplemental materials for publication: Functional tic-like presentations differ strikingly from Provisional Tic Disorder. <https://doi.org/10.17605/OSF.IO/R5FXN>.<sup>27</sup>

This project contains the following underlying data:

- FNS\_vs\_TS.csv (summary table from the FND-tic publications cited by Malaty et al.<sup>3</sup>)
- FND-tic\_not\_PTD.py (python script used to summarize data for Tables 1 and 2)
- individual\_participant\_data.csv (individual participant data from the New Tics group)
- earlier\_typical\_tic\_disorder.csv (individual participant data for the “previous episode of typical tics” item, from the larger set of all children screened for the New Tics group based on parental report of recent tic onset; see the file earlier\_typical\_tic\_disorder\_legend.txt for details.)
- earlier\_typical\_tic\_disorder\_legend.txt (definitions and legend for entries in the earlier\_typical\_tic\_disorder\_legend.csv file)

Data are available under the terms of the [Creative Commons Attribution 4.0 International license](https://creativecommons.org/licenses/by/4.0/) (CC-BY 4.0).

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An earlier version of this article can be found on Open Science Framework (doi: <https://doi.org/10.31219/osf.io/3u6bk>).

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## Version 2

Reviewer Report 15 May 2023

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**Michael Bloch** 

Department of Psychiatry, Yale School of Medicine, New Haven, Connecticut, USA

The authors have responded adequately to my critiques. I appreciate this important contribution to the study of tic disorders.

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Meta-analysis, psychopharmacology, tic disorders, OCD

**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.**

Reviewer Report 18 April 2023

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**Tammy Hedderly** 

Paediatric Neurology Department, Evelina London Children's Hospital, King's College London, Guy's & St Thomas' NHS Foundation Trust, London, England, UK

Thank you to the authors for their response to all my queries and for the clarification. I would like to approve this article as a helpful addition to this field.

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Paediatric Neurology , Movement Disorders and Tourettes

**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.**

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Version 1

Reviewer Report 31 March 2023

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**Tammy Hedderly** 

Paediatric Neurology Department, Evelina London Children's Hospital, King's College London, Guy's & St Thomas' NHS Foundation Trust, London, England, UK

The authors, all highly experienced 'Tourettologists', have approached this topic in an interesting and novel manner to try and provide some evidence for the ability to differentiate functional tic like behaviours from those seen in provisional tic disorders, usually related to Tourettes at follow up. This is a complex area, with some researchers proposing some shared mechanisms and overlapping neurobiology in the presentations. This paper uses some usefully collected detailed (although some retrospective) clinical data of children with PTD within a few months of presentation to compare characteristics demonstrating some useful distinguishing features. The presentation of the statistics could benefit from a little more expansion and explanation, especially for the non-expert, as it seems an important aspect of this paper. For example, table 2 is not that clearly explained and could do with more detail.

There are some limitations, but these have been clearly highlighted by the authors, such as the difference in ages of the two groups. I therefore would support this paper as it represents a useful contribution to the ongoing exploration of the functional tic-like phenomenon. I do wonder whether the dichotomy of subjects is a problem and would welcome wider discussions by the authors, on the situations seen commonly in the clinic in which young people, demonstrate both Tourette-related tics and functional tic-like behaviours together. This paper raised a question in my mind, as to whether the authors could have included distinguishing factors for the concept of provisional or transient functional disorder, especially when appearing in the context of exposure to social media or influencers.

Thank you for an interesting contribution.

**Is the work clearly and accurately presented and does it cite the current literature?**

Yes

**Is the study design appropriate and is the work technically sound?**

Yes

**Are sufficient details of methods and analysis provided to allow replication by others?**

Yes

**If applicable, is the statistical analysis and its interpretation appropriate?**

Partly

**Are all the source data underlying the results available to ensure full reproducibility?**

Yes

**Are the conclusions drawn adequately supported by the results?**

Partly

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Paediatric Neurology , Movement Disorders and Tourettes

**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 ) 06 Apr 2023

**Kevin J Black**

We thank Dr. Hedderly for her thoughtful and expert critique. Below we respond to each comment in turn.

1. *The presentation of the statistics could benefit from a little more expansion and explanation, especially for the non-expert, as it seems an important aspect of this paper. For example, table 2 is not that clearly explained and could do with more detail.*

Thank you. We agree. The new text in 'Results' reads as follows:

"Table 2 illustrates the clinical value of these comparisons, viewing each clinical feature as a "test" for FND-tic, with its associated sensitivity and specificity. We also provide the positive predictive value (PPV) for each feature thought to indicate FND-tic. The PPV represents how confident one can be of a FND-tic diagnosis given the presence of the listed feature. The PPV depends on the prior probability of the FND-tic diagnosis, or its likelihood before one knows whether a given patient has that feature. We provide PPV for each feature based on two prior probabilities, 2% and 50%. The first, 2%, represents the approximate rate before the pandemic of functional tic-like symptoms at clinical referral centers. 4 Clinical equipoise about a given participant's diagnosis is represented by a prior probability of 50%, for instance if one knows that a referring clinician is ambivalent about whether the patient has PTD or FND-tic, but one has not yet read the chart nor seen the patient. Features thought to be less common in FND-tic are listed with a negative predictive value (NPV), equivalent to PPV for the absence of the given feature."

2. *I do wonder whether the dichotomy of subjects is a problem and would welcome wider discussions by the authors, on the situations seen commonly in the clinic in which young people, demonstrate both Tourette-related tics and functional tic-like behaviours together.*

We agree the discussion did not address this issue previously. The discussion now includes this additional paragraph:

"We note that 22% of the patients reported in the FND-tic papers in fact had a prior history of typical tics, and many of them exhibited both tics and functional tic-like symptoms at the time they presented due to the latter. We have structured our results around whether FND-tic can be diagnosed, not whether PTD can also be diagnosed. In other words, when a patient presents with a typical history for TS but also new symptoms, the features above would allow more confidence in diagnosing FND-tic in addition to TS."

3. *This paper raised a question in my mind, as to whether the authors could have included distinguishing factors for the concept of provisional or transient functional disorder, especially when appearing in the context of exposure to social media or influencers.*

The reviewer raises a very interesting question that appears to include two concepts: diagnostic utility of exposure to social media and nosology.

a) The exposure to social media or other examples that the new symptoms appear to recapitulate is an important diagnostic clue. We and others have discussed that issue elsewhere (e.g. [Frey et al. \(2022\)](#) and [Malaty et al. \(2022\)](#)). Unfortunately, we have no prospectively collected data on this question from the PTD group. Anecdotally, I can report that although over a third of our New Tics study participants had a parent or sibling with tics, only once or twice was there any suggestion that the child's first tic may have appeared to be prompted by the family member's similar tic. We have added to the discussion the following paragraph:

"Other features in addition to those studied here may also be important for diagnosis. For instance, exposure to tic-like symptoms on social media was a common feature discussed in the papers discussing FND-tic in the past few years. <sup>3</sup> Unfortunately, we have no prospectively collected data from the PTD group on exposure to others with similar symptoms."

b) Traditionally, functional neurological symptoms have not been diagnosed differently based on duration of the defining symptoms. The closest analogy I can think of is Briquet's syndrome (and its dramatically modified descendants such as DSM-IV somatization disorder), which required not only atypical, medically unexplained symptoms starting before midlife but also numerous symptoms over time, affecting a variety of organ systems. Personally I would not favor creating a new diagnosis to represent "presumed functional symptoms in the first year since onset".

**Competing Interests:** No competing interests were disclosed.

Reviewer Report 25 January 2023

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**Michael Bloch** 

Department of Psychiatry, Yale School of Medicine, New Haven, Connecticut, USA

This manuscript compares the clinical characteristics of the sample of patients with Functional Neurological Disorder (FND) described in the literature, with a well-characterized sample of Provisional Tic Disorder. Strengths of the manuscript include the clinical importance and novelty of the data as well as the expertise of the authors and the well-written manuscript. Overall, I think this is a manuscript worthy of indexing, although I have several critiques that could potentially improve the manuscript with revision.

1. I do not believe the primary limitation of the manuscript is really acknowledged in the current draft. Mainly that the FND cases are drawn from the existing literature (where there is likely a large degree of reporting bias) versus a Provisional Tic Disorder sample that is much more general. Basically, there is quite a strong possibility that there are other characteristics associated with group difference other than diagnosis.
2. Please consider providing statistical tests in table 1 as well as sensitivity, specificity in table 2.
3. I think in the conclusion the authors should discuss the importance of testing the potentially meaningful differentiators in an independent dataset.
4. It would also be very worthwhile in further studies to use individual participant data to determine the PPV of multiple predictors to engage in a diagnostic distinction.
5. If possible, a histogram exploring age and gender differences between the two groups would be helpful.

**Is the work clearly and accurately presented and does it cite the current literature?**

Yes

**Is the study design appropriate and is the work technically sound?**

Yes

**Are sufficient details of methods and analysis provided to allow replication by others?**

Yes

**If applicable, is the statistical analysis and its interpretation appropriate?**

Yes

**Are all the source data underlying the results available to ensure full reproducibility?**

Yes

**Are the conclusions drawn adequately supported by the results?**

Partly

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Meta-analysis, psychopharmacology, tic disorders, OCD

**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, however I have significant reservations, as outlined above.**

Author Response ( F1000Research Advisory Board Member ) 06 Apr 2023

**Kevin J Black**

We thank Dr. Bloch for his thoughtful and expert critique. Below we respond to each comment in turn.

*1. I do not believe the primary limitation of the manuscript is really acknowledged in the current draft. Mainly that the FND cases are drawn from the existing literature (where there is likely a large degree of reporting bias) versus a Provisional Tic Disorder sample that is much more general. Basically, there is quite a strong possibility that there are other characteristics associated with group difference other than diagnosis.*

We agree and have edited the ms. to reflect this point. However, the disparity is smaller than one might think. Here is our new text in the discussion:

"The primary concern with the validity of these conclusions arises from the different potential sources of ascertainment bias in the two groups. Fortunately, these differences may not be as problematic as one might suppose. The FND-tic group is older, and if one's information were limited to this study alone, one might posit that the natural history of tic disorder included different early symptoms at different ages of tic onset. However, the literature includes decades of previous clinical information on typical tic disorders. Retrospective studies of TS and a prospective study of PTD in siblings of TS probands all found peak tic onset before age 10 years old; adult onset of tics is quite uncommon. 7, 8, 33 The PTD cases were volunteers willing to participate in a rather intensive research study, and were ascertained by a variety of methods (advertising and referrals from clinicians being the most common) rather than by clinical care-seeking. However, at the screening visit for the study, over 60% of them had sought clinical care for the tics or were planning to. The two FND-tic reports from Calgary, Alberta (child and young adult) were from a prospective registry of all patients seen for tics at their center, which is the only specialty center in their region. FND-tic cases were defined by "rapid onset of complex tic-like behaviors, with escalation to peak severity within hours to days." In other words, these reports are selected only for being seen at a specialty center for tic-like phenomena. Yet their clinical features are similar to those of the FND-tic literature overall (or even more

different from the PTD group). For instance, 95% of the Calgary FND-tic child patients were of female sex, only 25% had ADHD, and only 5% had OCD. We can conclude that potential bias from selecting more interesting, severe or classic cases for published case series does not substantially alter our results. Perhaps most importantly, there are no larger published sources for data about FND-tic nor about PTD. Nevertheless, ideally the results presented here should be confirmed in a new, independent sample."

2. *Please consider providing statistical tests in table 1 as well as sensitivity, specificity in table 2.*

Done.

3. *I think in the conclusion the authors should discuss the importance of testing the potentially meaningful differentiators in an independent dataset.*

We agree. The discussion now includes: "ideally the results presented here should be confirmed in a new, independent sample."

4. *It would also be very worthwhile in further studies to use individual participant data to determine the PPV of multiple predictors to engage in a diagnostic distinction.*

That's an excellent idea. We have supplied individual participant data for the New Tics group as a supplementary file.

5. *If possible, a histogram exploring age and gender differences between the two groups would be helpful.*

We do not have individual participant data for the FND-tic group (which came from 17 different publications), so we can't do this.

**Competing Interests:** No competing interests were disclosed.



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