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► **To cite this version:**

Antigony Rigas, Tina Mainka, Tamara Pringsheim, Alexander Münchau, Irene Malaty, et al.. Distinguishing functional from primary tics: a study of expert video assessments. *Journal of Neurology, Neurosurgery and Psychiatry*, 2023, 94 (9), pp.751-756. 10.1136/jnnp-2022-330822 . hal-04523247

HAL Id: hal-04523247

<https://hal.sorbonne-universite.fr/hal-04523247>

Submitted on 27 Mar 2024






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Original research

Distinguishing functional from primary tics: a study of expert video assessments

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► Additional supplemental material is published online only. To view, please visit the journal online (<http://dx.doi.org/10.1136/jnnp-2022-330822>).

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Received 23 November 2022
Accepted 30 March 2023
Published Online First 11 May 2023

ABSTRACT

Background Reliably applied criteria to differentiate functional from primary tics are lacking. In the absence of biological markers, the development of new diagnostic criteria to assist clinicians is predicated on expert judgement and consensus. This study examines the level of diagnostic agreement of experts in tic disorders using video footage and clinical descriptions.

Methods Using a two-part survey, eight experts in the diagnosis and management of tics were first asked to study 24 case videos of adults with primary tics, functional tics or both and to select a corresponding diagnosis. In the second part of the survey, additional clinical information was provided, and the diagnosis was then reconsidered. Inter-rater agreement was measured using Fleiss' kappa. In both study parts, the factors which influenced diagnostic decision-making and overall diagnostic confidence were reviewed.

Results Based on phenomenology alone, the diagnostic agreement among the expert raters was only fair for the pooled diagnoses ($\kappa=0.21$) as well as specifically for functional ($\kappa=0.26$) and primary tics ($\kappa=0.24$). Additional clinical information increased overall diagnostic agreement to moderate ($\kappa=0.51$) for both functional ($\kappa=0.6$) and primary tics ($\kappa=0.57$). The main factors informing diagnosis were tic semiology, age at tic onset, presence of premonitory urges, tic suppressibility, the temporal latency between tic onset and peak severity, precipitants and tic triggers and changes in the overall phenotypic presentation.

Conclusions This study confirmed that in the absence of clinical information, the diagnostic distinction between primary and functional tics is often difficult, even for expert clinicians.

INTRODUCTION

Tic disorders are among the most common hyperkinetic movement disorders in childhood and may also affect adults with a direct impact on their health and quality of life. The most prevalent tic aetiologies are primary tic disorders, such as Tourette syndrome (TS), but there is a wide range of differential diagnoses, including functional neurological disorder. Historically, the issue of functional tics has been at the centre of a prolonged and heated debate both in neurology and psychiatry with particular focus on the diagnostic classifiers of such cases and their clinical and pathophysiological distinction from primary tics.¹ However, perhaps owing to the

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ The distinction of primary from functional tics is often difficult. The relatively recent increase in prevalence and recognition of functional tics and the differences in treatment approaches between these aetiologies necessitate clinical diagnostic consensus. However, the validity of existing classifiers remains uncertain and there is little agreement as to the exact phenomenological boundaries between primary and functional tics.

WHAT THIS STUDY ADDS

⇒ This study demonstrates that it is not possible to distinguish primary from functional tics reliably based on phenomenology alone and that, even when key diagnostic points from the clinical history are provided, differences in expert opinion occur. Useful classifiers include age at tic onset, presence of premonitory urges, tic suppressibility, temporal evolution of symptoms, changes in phenotypic presentation and presence of contextual factors and triggers related to tics.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ The study highlights the imperative need to develop more accurate phenomenological definitions of tics, including novel diagnostic criteria for the different aetiologies and to identify reliable biomarkers that may allow disentangling between primary and functional tics. Until then, clinicians are advised to retain diagnostic humility when approaching challenging clinical areas such as this one.

lack of more advanced neuroscientific and therapeutic tools at the time, this faded from focus for a longer period. In recent years, there has been a renewed interest in this discourse, specifically triggered by the growing incidence of cases with atypical characteristics from those observed in primary tic disorders.^{2–6} Moreover, during the COVID-19 pandemic, there has been an even greater increase of such cases,^{7–11} many of which have attracted the attention of millions in social media, further fueling the old discussion as to how to distinguish functional from primary tics.



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To cite: Rigas A, Mainka T, Pringsheim T, et al. *J Neurol Neurosurg Psychiatry* 2023;**94**:751–756.

Movement disorders

To date, several phenomenological classifiers have been proposed to discern primary tic phenomena from functional tics.^{2–6 9 10 12} These diagnostic aids have been developed by observing the typical patterns of tics documented in people with primary tic disorders and contrasting these to movements and behaviours that grossly differ. However, in the absence of an established diagnostic standard or biomarker for either aetiology, the validity of such classifiers remains uncertain. Moreover, there is little consensus as to the exact phenomenological boundaries between primary and functional tics. Importantly, it remains unclear how to diagnostically approach cases where primary tics may coexist with functional tics, even though the combination is not uncommon. According to a recent international survey of members of the Movement Disorders Society, the second most common differential diagnosis to primary tics was functional tics, and the coexistence of the two diagnoses was reported to be as high as 25%.¹³

To accelerate progress towards the definition of phenomenological classifiers and aid the diagnostic distinction between primary and functional tics, experts with longstanding clinical and research experience in tic disorders and members of the Tic Disorders and Tourette Syndrome Study Group of the International Movement Disorder Society were invited to participate in this case-based study. Experts were given 24 tic disorder cases previously diagnosed as either primary tics, functional tics or both as part of a two-step survey. In the first part of the study, expert clinicians were only shown the videos of each case with key phenomenological features and were requested to provide a diagnosis based on phenomenology alone. In the second part, additional information from clinical history and examination was provided to assess whether this led to diagnostic reclassification. The goal of the study was to explore the level of diagnostic agreement of experts in tic disorders on the basis of phenomenology alone and to determine the key factors that increase diagnostic consensus and confidence in discerning primary and functional tics, or their coexistence.

METHODS

A case-based, two-part survey was created using the REDCap software¹⁴ and presented to eight movement disorder experts with longstanding clinical and research experience in tic disorders (AM, TP, IM, YW, AEC, AJL, DM, AEL). The first part was comprised of videos of 24 adults with tic disorders (14 men; mean age 26.7±10.6 years) who were seen at the tic disorders and TS clinics of the senior author (CG). Cases were selected based on their phenomenology and clinical history, as well as their given diagnosis in clinic, which was either that of a primary tic disorder, a functional tic disorder or a primary tic disorder overlaid with functional tics. All videos were captured upon obtaining written consent as part of clinical consultation, either with and/or without the clinician in the room. Each case video was edited for 2.5 min to demonstrate the predominant phenotypic presentation. Following the presentation of each video in REDCap, questions to raters focused on the clinical characteristics, including the predominant phenomenology (eg, simple or complex tics) and the variability between the different behaviours (ie, whether a certain phenomenon occurred repetitively or whether each phenomenon differed from the previous one) in each case. Each of the experts was then requested to provide a diagnosis among the four following categories: (1) primary tic disorder/TS; (2) primary tic disorder/TS overlaid with tic-like behaviours, most likely of functional and/or other aetiologies (functional overlay); (3) tic-like behaviours non-consistent with

the diagnosis of a primary tic disorder, most likely of functional and/or other aetiologies (functional tics); (4) non-tic disorder. The fourth diagnostic category was added to allow diagnosing phenomena that could, for some experts, semiologically strongly depart from the rubric of tics (as discussed in Kurvits *et al*).¹⁵ The level of confidence for each provided diagnosis was captured on a scale from 0 to 100, as were the key phenomenological factors that led to each diagnosis. These included: the semiology of observed behaviours (ie, type of tics), their severity and body distribution as well as their variability. A fifth category of 'other factors' captured additional information that led to diagnosis.

In the second part of the survey, experts were given additional standardised historical information on each case. This information included age at onset of abnormal behaviours, clinical progression, the presence of sensory phenomena preceding the repetitive behaviours, their amenability to voluntary suppression, their impact on quality of life, the presence of additional medical diagnoses and the intake and effects of any medication. Based on this information, experts were then queried whether they would like to keep or reconsider their original diagnosis (from part 1), their current level of confidence (on a scale of 0–100, where 100 indicates absolute confidence) in the final diagnosis and the top factors that informed their decision. These now included age at presentation, age of onset of repetitive behaviours, presence of precipitants/contextual factors, type of first tic, changes in clinical presentation, time course from onset to maximum severity, presence of premonitory urges, ability to voluntarily suppress repetitive behaviours, presence of additional diagnoses as well as additional findings on clinical and additional investigations, other than those demonstrated in video. A further category captured 'other factors' (presented as free text) that informed final diagnosis. The full survey is found in the supplement (online supplemental file 1).

The level of diagnostic agreement between expert raters for parts 1 and 2 (ie, phenomenology alone; and phenomenology with additional clinical information) was computed using Fleiss' kappa. Kappa values between 0 and 0.20 were determined as slight agreement, 0.21–0.40 as fair, 0.41–0.60 as moderate, 0.61–0.80 as substantial and 0.81–1 as almost perfect.¹⁶ The SPSS software V.29 was used for all statistics. In two instances, specific data (tic phenomenology for case 5; top factors that lead to final diagnosis for case 13) could not be obtained due to a technical error in RedCap programming, and analyses were conducted without these specific datapoints. Kruskal-Wallis analysis was used to measure the effect of 'diagnosis' on the variance of the 0–100 scale-based judgement of diagnostic confidence and variability. Confidence between primary and final diagnosis was compared using the Wilcoxon test. Statistical significance was set at the $p < 0.05$ threshold.

The data that support the findings of this study are available in the supplement. The complete data set of all responses is available on reasonable request to the corresponding author.

RESULTS

The clinical characteristics of the 24 video cases are presented in online supplemental table 2.

From 184 possible responses for each of the phenomena (data from 23/24 video cases), simple motor tics were detected 148 times by the eight raters, followed by complex motor tics in 109 instances. Simple phonic tics and complex phonic tics were observed 96 and 32 times, respectively. Tic-like behaviours uncommon for primary tics were noted 41 times. Coprophenomena were detected 37 times, whereas echophenomena and

Table 1 Diagnostic confidence for all 24 cases together (all diagnoses) and split for each diagnosis at parts 1 (video-based diagnosis) and 2 (with additional information provided).

| | | All diagnoses | Primary tics/Tourette syndrome | Primary and functional tics | Functional tics | Non-tic disorder |
|-----------------------|--------|---------------|--------------------------------|-----------------------------|-------------------|------------------|
| Diagnostic confidence | Part 1 | 77.5 (64–90) | 85 (70–91) | 60 (50–72) | 76.5 (66.5–87.25) | 61 (49.5–63) |
| | Part 2 | 92.5 (80–100) | 100 (89.5–100) | 80 (70–90) | 92.5 (80–99) | 90* |

Diagnostic confidence is provided as median and interquartile range (Q1–Q3).
*Diagnosis given only once by only one expert.

paliphenomena 1 and 5 times only. Experts noted stereotypies eight times, and the phenomenological category of ‘other non-repetitive behaviours’ was selected 10 times. online supplemental table 3 provides a breakdown of detection frequency per case.

Based on the video-documented phenomenology of the 24 cases, the overall diagnostic agreement among the expert raters was fair ($\kappa=0.21$; 95% CI 0.152 to 0.26, $p<0.001$; see online supplemental table 4 for experts’ diagnoses per case). Examined per diagnosis, inter-rater agreement was fair for functional and primary tics ($\kappa=0.26$; 95% CI 0.181 to 0.332, $p<0.001$ and $\kappa=0.24$; 95% CI 0.166 to 0.317, $p<0.001$, respectively), but slight for primary tics coexisting with functional tics ($\kappa=0.08$; 95% CI .007 to .158, $p=0.032$), as also for the diagnosis of a non-tic disorder ($\kappa=0.03$, 95% CI -0.044 to 0.108, $p=0.408$). The median diagnostic confidence was 77.5 (IQR 64 to 90) and was highest for the diagnosis of primary tics/TS (median: 85; IQR 70 to 91), followed by functional tics (median: 76.5, IQR 66.5 to 87.25, $H(3)=37.757$, $p<0.001$) (see table 1).

The top factor driving the diagnostic distinction between primary and functional tics based on video evaluation alone was semiology (list of frequencies of reported factors provided in table 2).

Tic variability was significantly higher for cases diagnosed as functional tics (median: 75.5; IQR 59.5 to 85), and for those with overlap of primary and functional tics (median 66; IQR 50 to 71) compared with the diagnosis of primary tics (median: 20; IQR 10 to 30) and a non-tic disorder (median: 26; IQR 3.5 to 44.5, $H(3)=83.804$, $p<0.001$). Experts reported for 178 times (93% of all expressed judgements, that is, 24 cases rated by eight independent raters) that additional information was needed to increase diagnostic confidence. Most common queries included information about the age of tic onset, the presence of precipitants and contextual factors associated with tic onset and the temporal course of clinical symptom evolution.

When provided with additional information, the overall diagnostic agreement of experts increased to moderate ($\kappa=0.51$; 95% CI 0.456 to 0.565, $p<0.001$; see online supplemental table 4 for experts’ diagnoses per case). Inter-rater agreement was moderate for the diagnoses of functional ($\kappa=0.6$; 95% CI 0.528 to 0.679, $p<0.001$) and primary tics ($\kappa=0.57$; 95% CI 0.492 to 0.644, $p<0.001$) and fair for primary tics coexisting with functional tics ($\kappa=0.33$, 95% CI 0.254 to 0.405, $p<0.001$). The diagnosis of a non-tic disorder was applied at this stage in only one case and by one expert only ($\kappa=-0.005$; 95% CI -0.081 to

0.07, $p=0.892$) but was not further specified. Confidence in the final diagnosis was significantly higher than based on phenomenology alone (median: 92.5; IQR 80 to 100; $Z=-9.223$, $p<0.001$). Diagnostic confidence was highest for the diagnosis of a primary tic disorder (median: 100; IQR 89.5 to 100), followed by the diagnosis of functional tics (median: 92.5; IQR 80 to 99, $H(3)=43.374$, $p<0.001$) (also see table 1). The top clinical information factors most relevant for the diagnosis of primary tics were knowledge on the presence of premonitory urges and tic suppressibility as well as the age at onset of tics. The top factors that allowed the raters to reach a final diagnosis of functional tics were the type of precipitants that were reported to be associated with the manifestation of tics, the time course from onset to maximum tic severity and the age at onset of tics. The most common factor informing the diagnosis of coexisting primary and functional tics was an overall change in phenotypic presentation. Table 3 provides a list of frequencies of top-rated factors for each diagnosis.

DISCUSSION

This study assessed the level of agreement between tic experts in diagnosing 24 cases with tics as either primary, functional or both. A two-step approach was used, in which each expert was expected to make a diagnosis based on patient videos alone, and then again after receiving details of the clinical history. The findings emphasise that it is difficult to distinguish primary tics from functional tics based on observation of the movement disorder alone and that clues from the clinical history are needed. These include the age at tic onset, the temporal evolution of symptoms, changes in overall phenotypic presentation and the presence of contextual factors and triggers related to tic behaviours.

The main goal of this study was to explore whether experts would agree on the diagnostic distinction between primary and functional tics, or their coexistence, on the basis of clinical observation (video-presented cases) alone. This topic has been at the centre of a long-standing debate in tic disorders, specifically in relation to whether phenomenological classifiers are sufficient to inform diagnostic consensus^{1 6 17 18} and has been fuelled recently by a marked increase in the number of people diagnosed with functional movement disorders that seem to have been associated with exposure to social media platforms.^{7–11} Some experts have argued that distinguishing between primary and functional tics is straightforward and can be achieved by simply observing

Table 2 Top factors leading to the diagnosis (part 1, video-based only)

| | Primary tics/Tourette Syndrome | Primary and functional tics | Functional tics | Non-tic disorder |
|-------------------|--------------------------------|-----------------------------|-----------------|------------------|
| Semiology | 98.1% (104/106) | 92.6% (25/27) | 98.1% (53/54) | 100% (5/5) |
| Severity | 11.3% (12/106) | 14.8% (4/27) | 13% (7/54) | 100% (5/5) |
| Body Distribution | 63.2% (67/106) | 51.9% (14/27) | 55.6% (30/54) | 60% (3/5) |
| Variability | 30.2% (32/106) | 70.4% (19/27) | 64.8% (35/54) | 20% (1/5) |
| Other | 18.9% (20/106) | 37% (10/27) | 16.7% (9/54) | 100% (5/5) |

Table 3 Top factors leading to the diagnosis (part 2, additional clinical history)

| | Primary tics/Tourette syndrome | Primary and functional tics | Functional tics | Non-tic disorder* |
|---|--------------------------------|-----------------------------|-----------------|-------------------|
| Age at presentation | 32.2% (29/90) | 39.5% (15/38) | 41.8% (23/55) | 100% (1/1) |
| Age at onset of repetitive behaviours | 61.1% (55/90) | 63.2% (24/38) | 61.8% (34/55) | – |
| Precipitants/contextual factors | 11.1% (10/90) | 50% (19/38) | 63.6% (35/55) | – |
| First tic | 36.7% (33/90) | 26.3% (10/38) | 16.4% (9/55) | – |
| Changes in clinical presentation | 12.2% (11/90) | 71.1% (27/38) | 21.8% (12/55) | – |
| Time course from onset to maximum severity | 32.2% (29/90) | 34.2% (13/38) | 61.8% (34/55) | – |
| Presence of premonitory urge | 73.3% (66/90) | 36.8% (14/38) | 3.6% (2/55) | 100% (1/1) |
| Ability to voluntarily suppress repetitive behaviours | 57.8% (52/90) | 23.7% (9/38) | 10.9% (6/55) | 100% (1/1) |
| Additional diagnoses | 32.2% (29/90) | 36.8% (14/38) | 20% (11/55) | 100% (1/1) |
| Additional findings of clinical and paraclinical investigation, other than those demonstrated in the video, where available | 1.1% (1/90) | 2.6% (1/38) | 1.8% (1/55) | – |
| Other findings | 8.9% (8/90) | 7.9% (3/38) | 7.3% (4/55) | – |

Data related to the selection of top factors is missing for one case, due to a technical error.
*Diagnosis given once by a single expert.

the abnormal movements over a period of time,^{11 17} while others have questioned this belief.^{6 18} The low level of diagnostic agreement based on case videos in this study provides support for the latter view. However, even though diagnostic agreement was low, the experts did feel that tic semiology was the most important factor informing their overall decision-making and highlighted the importance of the cranio-caudal distribution of primary tics, and tic variability for functional tics. The experts were in agreement that additional information was required, specifically related to the onset, the presence of associated contextual factors or precipitants, and the temporal course of symptoms.

When clinical information was added, overall diagnostic agreement improved from fair ($k=0.21$) to moderate ($k=0.51$), which was also paralleled by an increase in diagnostic confidence. Although the age of tic onset was judged in all categories as a helpful distinguishing criterion, the presence of premonitory urges and the amenability of tics to voluntary tic inhibition were felt to be the most helpful factors to diagnose primary tics, even though they are not part of the current definition used to describe tics.¹⁹ In contrast, the diagnosis of functional tics was informed by the presence of specific precipitants and contextual factors associated with tic manifestation (eg, following COVID-19 vaccination or an episode of “collapse” at work), as well as the time course between tic onset and maximum severity. Physical and psychological precipitants associated with the acute onset of functional tics have been reported in the recent marked increase in functional tics observed during the past 3 years, as also documented in other functional movement disorders.^{20–23} In primary tics, stressors may lead to exacerbations of tic severity but have not been linked to tic onset.^{24 25} The time course of tic manifestation was a further useful classifier, as many cases of functional tics develop acutely or subacutely and may reach a “full blown” clinical picture within a matter of hours, days, or few weeks.²³

Despite the improvement in diagnostic reliability with access to the clinical data, it is noteworthy that the maximum overall level of agreement was still only moderate. Although there was agreement for several cases, diagnostic difficulties for overlapping phenomena, mostly for the diagnosis of a primary tic disorder overlaid with functional tics occurred. Indeed, diagnostic agreement here only improved from slight ($k=0.08$) to fair ($k=0.33$). In the absence of clear categorical criteria to distinguish primary and functional tics, which may also present with similar semiology, this diagnosis remains challenging, even though many movement disorder clinicians with experience in diagnosis and

managing patients with tics believe that this combination is quite common.¹³ Of note, changes in phenotypic presentation were deemed as the most useful factor in the clinical history to inform this diagnostic possibility. A related challenging diagnostic judgement relates to whether all the observed behaviours fall under the tic rubric. Our experts identified ‘tic-like behaviours uncommon for primary tics’ 41 times, and two experts selected the diagnosis of a non-tic disorder based on video-evaluation alone five times in total. Importantly, the diagnosis of a non-tic disorder was retained by one expert even after the additional clinical information provided. Overall, our results highlight the existing difficulties, even for experts, to reliably apply the operational definition of tics, as it appears that the term tic is used to indicate a diversity of repetitive behaviours (also see²⁶).

Diagnostic disagreement between expert clinicians may have been the result of differing criteria used to establish a functional tic diagnosis, and differences in practical experience with functional tic patients based on referral biases and practice volumes. At the time of performing this analysis, diagnostic clues for the diagnosis of functional tic disorder had been discussed in several publications^{6 9} but specific criteria were not formally established, leaving the clinicians involved in this project to mainly rely on their own clinical intuition to make this diagnosis. Furthermore, clinical volumes with patients with functional tics varied between expert clinicians, which likely influenced individual expertise and confidence in making a functional tic diagnosis.

Our study could not assess the accuracy of our experts’ final diagnostic judgement, due to the lack of diagnostic standards or biomarkers. Very recently, after our data collection was complete, a single-centre study developed a set of diagnostic criteria that yielded encouraging discriminatory capacity to differentiate between the diagnoses of ‘functional tic disorder’ and ‘primary tic disorder’.¹² Although differentiation by these criteria required the presence of at least 2 of 7 different phenomenological characteristics potentially detectable through direct observation alone, it also required additional clinical information that included type of onset, comorbidity, and even sex at birth and family history. Even more recently, consensus-based diagnostic criteria for the clinical diagnosis of functional tic like behaviours have been published by an international group with expertise in tic disorders.²⁷ A ‘clinically definite’ diagnosis requires the presence of three major criterion which allow clinicians to differentiate functional tic like behaviours from tics—age of symptom onset (age 12 and older), rapid onset and evolution of symptoms (over hours to days) and the presence of four of

nine phenomenological features. While broad clinical applicability and usefulness of these criteria will need to be verified by other authors, their formulation aligns with our findings that phenomenology alone is insufficient to differentiate between functional and primary tic disorders.

The 24 cases that we selected to measure agreement among experts were designed to test clinicians' ability to distinguish primary tic disorder, functional tic disorder or an overlap between the two. The 'real-world' diagnoses that these patients had received were not factored in the analyses, because it may have confounded the interpretation. A potential limitation of our study is the duration of the edited videos. Although we presented 2.5 min videos for each case, it could be argued that a longer observation would have allowed for greater agreement, for example, through a more detailed representation of how certain behaviours cluster in time. However, the edits were selected to depict all relevant clinical signs that each patient exhibited during their clinical presentation, and, therefore, accurately reflect the phenotype observed in clinic. Finally, although we selected several top factors as distilled from the existing literature for experts to choose from for each of the two study parts (phenomenology vs clinical information), it is possible that other informative factors were omitted. However, no other top factor was consistently brought up in the 'others' category of both top factors lists.

This study indicates that it is not possible to distinguish primary tics from functional tics with any level of confidence from short video clips alone and that even when key diagnostic points from the clinical history are also provided differences in expert opinion occur. This highlights the imperative need to develop more accurate phenomenological definitions of tics, including novel diagnostic criteria for the different etiologies^{12 27} as well as to identify reliable biomarkers that may allow disentangling between primary and functional tics. In the current study, the contrast between the fair to moderate achieved agreement at both steps and the individual high to very high confidence in diagnosis suggests that clinicians should retain diagnostic humility when approaching challenging clinical areas such as this one.

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Funding This research was supported by a VolkswagenStiftung (Freigeist) grant (AZ. 94 268) held by Christos Ganos.

Competing interests None declared.

Patient consent for publication Not applicable.

Ethics approval This study involves human participants and was approved by Charité University Medicine Berlin local ethics committee (EA2/152/22). Participants gave informed consent to participate in the study before taking part.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement Data are available upon reasonable request. All data relevant to the study are included in the article or uploaded as supplementary information. Data beyond the ones included in the article or uploaded as supplementary information will be available upon reasonable request.

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Supplement 1. Expert questionnaire.

This questionnaire was presented to each expert for each of the 24 cases.

Part 1

Video.

1. What do you see?

- simple motor tics (list top three if applicable)
- simple phonic tics (list top three if applicable)
- complex motor tics (list top three if applicable)
- complex phonic/vocal tics (list top three if applicable)
- echophenomena (list top three if applicable)
- paliphenomena (list top three if applicable)
- coprophenomena (list top three if applicable)
- stereotypies (list top three if applicable)
- tic-like behaviours uncommon to classic presentations of primary tics (e.g. allo-aggressive behaviours, stimulus triggered behaviours)
- other non-repetitive behaviours

2. What is the predominant phenomenon from the above? (more than one may apply; e.g. simple and complex motor tics)

3. Please rate the severity of the observed behaviour

- 0 = absent tics
- 1 = minimal: could be normal
- 2 = mild: limited to a single muscle group/ single words or sounds, separated by at least one breath or 4 sec
- 3 = moderate: limited to a single body part/words or sounds repeated 2 or 3 times in series or single obscenities separated by at least 1 breath or 4 sec
- 4 = severe: involve more than one body part or complex/words or sounds repeated four or more times in series or obscenities repeated at least 2-3 times in series

4. Please note the affected body parts

- Eyes
- Nose
- Mouth
- Neck
- Shoulders
- Arms
- Hands
- Trunk
- Pelvis
- Legs
- Feet

5. Is there a rostrocaudal tic distribution?

- Yes
- No

6. Please provide a percentage from 0-100 with regard to the variability of the observed behaviors (0 signifies no variability in observed behaviors (e.g., repetitive blinking only), 100 signifies maximum variability of observed behaviors (every observed behavior is different from the previous one))

7. Based on the above observations, which diagnosis do you feel applies best?

- Primary tic disorder/Tourette Syndrome
- Primary tic disorder/Tourette Syndrome overlaid with additional tic-like behaviors, most likely of functional and/or other etiologies (functional overlay)
- Tic-like behaviors non-consistent with the diagnosis of a primary tic disorder, most likely of functional and/or other etiologies (functional tics)
- Non-tic disorder

8. How confident are you about this video-based diagnosis? (0 – 100%)

9. Do you feel more information is needed to increase your diagnostic confidence?

- Yes
- No

10. Kindly note the specific information you would need in this case

11. Please list the top factors which helped you reach the diagnosis in this specific case (You may either select from the following list or write free text)

- Semiology of observed behaviours
- Severity
- Body distribution
- Variability
- Free text

Part 2

Patient history. The following clinical information is about the patient from the previous video.

1. Based on the additional information you received about this case would you like to change your diagnosis?

- Yes
- No

2. Based on the above observations, which diagnosis do you feel applies best?

- Primary tic disorder/Tourette Syndrome
- Primary tic disorder/Tourette Syndrome overlaid with additional tic-like behaviors, most likely of functional and/or other etiologies (functional overlay)
- Tic-like behaviors non-consistent with the diagnosis of a primary tic disorder, most likely of functional and/or other etiologies (functional tics)
- Non-tic disorder (please specify which non-tic disorder)

3. Which top factors from the provided clinical history above led to your final diagnosis? (Please select or write free text)

- Age at presentation
- Age of onset of repetitive behaviours
- Precipitants/Contextual factors
- First tic

- Changes in clinical presentation
- Time course from onset to maximum severity
- Presence of premonitory urge
- Ability to voluntarily suppress repetitive behaviors
- Additional diagnoses
- Additional findings of clinical and paraclinical investigations, other than those demonstrated in video, where available
- Free text

4. Based on the documented semiology and the information from the patient's history how confident are you about your final diagnosis? (0-100%)

Supplementary table 2. Individual patient data.

| Case | Age | Sex | Predominant Tics and Tic-like behaviors | Comorbidities | Diagnosis |
|------|------------|-----|---|--|------------------------------------|
| 1 | Late teens | F | head jerks, eye blinks, arm jerks, clicking sounds | Depression, anxiety disorder and panic attacks | Functional tics |
| 2 | Early 20s | M | eyebrow elevation, grimacing, head-to-side, shoulder elevation | - | Primary tics/ Tourette Syndrome |
| 3 | Mid 40s | M | lip pulling, isolated and bilateral platysma contractions | OCD, anxiety disorder | Primary tics/ Tourette Syndrome |
| 4 | Mid 20s | M | head jerking, eye closing, complex tics with patterned movements (turning to the side, downward head movement, eye closing, vocalizing "banana"), coprolalic behaviors | ADHD, OCD | Primary and functional tics |
| 5 | Early 20s | F | arm raising, using arm to slap herself, swinging leg movements, sudden full body movements either to the front or back, startling movements, going to the floor to hug the examiner | - | Functional tics |
| 6 | Early 20s | F | eye blinking, eye rolling, grimacing, opening mouth widely, backward head jerks, shoulder jerks, head to side, hitting left fist on her chest, whistling | Previous diagnosis of foot dystonia, chronic myalgia, Bell's palsy in the past | Primary and functional tics |
| 7 | Early 40s | M | blinking, eye rolling, mouth movements, head jerking, simple phonic tics (nose clearing, grunting), thumb twitching | ADHD, OCD | Primary tics/ Tourette Syndrome |
| 8 | Late teens | F | jerky head movements, head retraction, protecting ears with hands while making sounds and closing eyes, clapping with hands, tongue protrusion | Depression | Functional tics |
| 9 | Early 20s | M | eye blinking, eyebrow elevation, repetitive sudden and brisk head turning to either side (mostly left), clapping with hands, shouting phonation, bouts of episodes of sequences of aforementioned movements | Depression, possible borderline personality disorder | Primary and functional tics |

| | | | | | |
|----|------------|---|--|---|---------------------------------------|
| 10 | Mid 20s | F | head jerking, facial grimacing (racing eyebrows, opening mouth, and making plopping sound), head turning and shoulder elevation, or head turning, plopping sound and hand toward head | Depression, anxiety disorder, post-traumatic stress disorder, anorexia nervosa | Functional tics |
| 11 | Mid 20s | F | eyebrow movements (isolated and bilateral) forehead wrinkling, mouth pulling, nose movements, truncal jerks whilst on chair | Depression, anxiety disorder | Primary tics/ Tourette Syndrome |
| 12 | Mid 30s | M | Mouth grimacing and turning head to the side, tonic and dystonic platysma and lower face tensing, phonations ("ahhh"), repetitive deep in- and exhales | Depression, functional neurological disorder | Functional tics |
| 13 | Late teens | F | head/neck jerks to the side and back, brief arm movements (e.g., arm extension, hitting herself on the chest with fist), bending with body whilst sitting, repetitive shouting phonations (as if holding air and letting out) accompanied by head and body movements | Depression, OCD | Primary and functional tics |
| 14 | Mid 30s | F | eye blinking, rolling and oculogyric tics, mouth movements to side and side-to-side jaw movements, repetitive swallowing, head turning, platysma spasms | Insomnia, post-traumatic stress disorder, borderline personality disorder, depression, migraine, eating disorder | Primary tics/ Tourette Syndrome |
| 15 | Early 20s | M | eye blinking, sniffing, to-and-fro movements of scalp, ear tics, neck to side, neck turning | OCD | Primary tics/ Tourette Syndrome |
| 16 | Late teens | M | head jerking, facial expressions, touching the doctor/grabbing objects from doctor's hand, arm jerks, squeaking noises and eye closing, coprolalic behaviors, shouting "no" | ADHD | Functional tics |
| 17 | Late teens | M | head jerks, mouth opening, pulling a finger, clapping hands, grabbing objects, variable arm and hand movements, repetitive trunk flexion, diverse and continuous coprolalic content, many different inappropriate words and phrases | Oppositional defiant disorder, autism spectrum disorder, ADHD, alcohol- and other addictions, borderline personality disorder | Primary and functional tics |
| 18 | Mid 20s | M | eyebrow elevation, eye rolling, grabbing objects and stamping on the desk, jerks with hands and truncal bending, throwing items | ADHD | Functional tics |

| | | | | | |
|----|-----------|---|---|--|--------------------------------|
| | | | away | | |
| 19 | Early 20s | F | repetitive head turning and checking behavior to the left, facial grimacing/dystonic facial movements, extending left arm, repetitive "hmm" sounds, finger snapping | ADHD, seizures, alcohol addiction | Primary and functional tics |
| 20 | Late 50s | M | repetitive blinking, raising eyebrows, wide mouth opening, stuttering, repetitive noises either in isolation or combined with facial expression and/or arm movements, pulling a finger | Anxiety disorder, post-traumatic stress disorder, chronic laryngitis | Primary Tics/Tourette Syndrome |
| 21 | Early 40s | M | repetitive blinking, eyes to the side, head/neck jerks, left hand to nose, truncal and/or pelvic jerks, leg jerks | - | Primary and functional tics |
| 22 | Early 20s | M | eye rolling, eyebrow elevation, sniffing, grimacing, laryngeal movements, platysma movements, dystonic platysmal tensing, side to side movements of finger ligaments | - | Primary tics/Tourette Syndrome |
| 23 | Early 20s | M | eyebrow elevation and frowning, blinking, grimacing, pulling finger repetitively, head jerks, abrupt hand jerks, coprolalia and shouting words or phrases like "help", "now I have Covid" | Depression, cannabis addiction, ADHD | Primary and functional tics |
| 24 | Mid 20s | F | head turning and jerking, grimacing and clicking mouth sounds, arm movements synchronously to repetitive wording, coprolalic words/sentences together with arm movements and head jerks | OCD, functional seizures | Functional tics |

ADHD, Attention Deficit Hyperactivity Disorder, OCD, obsessive compulsive disorder

Supplementary table 3. Individual data on the frequency of tics and other repetitive behaviours rated by 8 experts.

*Case 5 omitted due to technical error in data sampling

| | Simple motor tics | Simple phonic tics | Complex motor tics | Complex phonic tics | Echo-phenomena | Pali-phenomena | Copro-phenomena | Stereotypies | Tic-like behavior uncommon of primary tics | Other non-repetitive behaviours |
|-----------|-------------------|--------------------|--------------------|---------------------|----------------|----------------|-----------------|--------------|--|---------------------------------|
| Case 1 | 6/8 | 1/8 | 5/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 | 2/8 |
| Case 2 | 5/8 | 4/8 | 7/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 | 0/8 |
| Case 3 | 7/8 | 1/8 | 2/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 |
| Case 4 | 5/8 | 5/8 | 6/8 | 5/8 | 0/8 | 0/8 | 7/8 | 0/8 | 4/8 | 0/8 |
| Case 6 | 6/8 | 6/8 | 5/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 | 1/8 | 0/8 |
| Case 7 | 7/8 | 8/8 | 4/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 | 1/8 | 0/8 |
| Case 8 | 6/8 | 7/8 | 7/8 | 0/8 | 1/8 | 0/8 | 0/8 | 0/8 | 3/8 | 0/8 |
| Case 9 | 7/8 | 7/8 | 7/8 | 0/8 | 0/8 | 0/8 | 2/8 | 0/8 | 3/8 | 0/8 |
| Case 10 | 7/8 | 7/8 | 4/8 | 0/8 | 0/8 | 2/8 | 0/8 | 0/8 | 1/8 | 1/8 |
| Case 11 | 6/8 | 2/8 | 2/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 | 0/8 | 1/8 |
| Case 12 | 6/8 | 4/8 | 4/8 | 1/8 | 0/8 | 0/8 | 0/8 | 0/8 | 2/8 | 0/8 |
| Case 13 | 6/8 | 7/8 | 7/8 | 0/8 | 0/8 | 0/8 | 1/8 | 0/8 | 2/8 | 0/8 |
| Case 14 | 7/8 | 1/8 | 1/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 1/8 |
| Case 15 | 8/8 | 1/8 | 1/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 |
| Case 16 | 7/8 | 6/8 | 7/8 | 7/8 | 0/8 | 1/8 | 6/8 | 0/8 | 4/8 | 0/8 |
| Case 17 | 6/8 | 6/8 | 5/8 | 6/8 | 0/8 | 0/8 | 6/8 | 1/8 | 6/8 | 1/8 |
| Case 18 | 7/8 | 0/8 | 7/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 4/8 | 0/8 |
| Case 19 | 5/8 | 7/8 | 7/8 | 0/8 | 0/8 | 0/8 | 0/8 | 2/8 | 1/8 | 0/8 |
| Case 20 | 7/8 | 6/8 | 3/8 | 2/8 | 0/8 | 0/8 | 1/8 | 2/8 | 0/8 | 1/8 |
| Case 21 | 8/8 | 0/8 | 4/8 | 0/8 | 0/8 | 0/8 | 1/8 | 0/8 | 0/8 | 0/8 |
| Case 22 | 8/8 | 0/8 | 2/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 0/8 | 2/8 |
| Case 23 | 8/8 | 4/8 | 6/8 | 6/8 | 0/8 | 2/8 | 7/8 | 0/8 | 3/8 | 0/8 |
| Case 24 | 3/8 | 6/8 | 6/8 | 5/8 | 0/8 | 0/8 | 6/8 | 0/8 | 4/8 | 0/8 |
| All cases | 148 (80.4%) | 96 (52.2%) | 109 (59.2%) | 32 (17.4%) | 1 (0.5%) | 5 (2.7%) | 37 (20.1%) | 8 (4.3%) | 41 (22.3%) | 10 (5.4%) |

Supplementary table 4. Individual data on the diagnoses given by the 8 experts after part 1 and part 2.

| | Diagnosis Part 1 | Diagnosis Part 2 |
|----------------|---|--|
| Case 1 | Primary Tics/ Tourette Syndrome 2/8 Primary and functional tics 2/8 Functional tics 4/8 | Primary and functional tics 1/8 Functional tics 7/8 |
| Case 2 | Primary Tics/ Tourette Syndrome 6/8 Functional tics 2/8 | Primary Tics/Tourette Syndrome 8/8 |
| Case 3 | Primary Tics/ Tourette Syndrome 5/8 Functional tics 1/8 Non-tic disorder 2/8 | Primary Tics/Tourette Syndrome 8/8 |
| Case 4 | Primary Tics/ Tourette Syndrome 1/8 Primary and functional tics 4/8 Functional tics 3/8 | Primary Tics/Tourette Syndrome 1/8 Primary and functional tics 7/8 |
| Case 5 | Functional tics 8/8 | Functional tics 8/8 |
| Case 6 | Primary Tics/ Tourette Syndrome 5/8 Primary and functional tics 2/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 2/8 Primary and functional tics 2/8 Functional tics 4/8 |
| Case 7 | Primary Tics/ Tourette Syndrome 6/8 Primary and functional tics 1/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 7/8 Primary and functional tics 1/8 |
| Case 8 | Primary Tics/ Tourette Syndrome 2/8 Primary and functional tics 1/8 Functional tics 5/8 | Functional tics 8/8 |
| Case 9 | Primary Tics/ Tourette Syndrome 3/8 Primary and functional tics 4/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 5/8 Primary and functional tics 3/8 |
| Case 10 | Primary Tics/ Tourette Syndrome 3/8 Primary and functional tics 1/8 Functional tics 4/8 | Primary Tics/Tourette Syndrome 2/8 Primary and functional tics 1/8 Functional tics 5/8 |
| Case 11 | Primary Tics/ Tourette Syndrome 5/8 Functional tics 2/8 Non-tic disorder 1/8 | Primary Tics/Tourette Syndrome 7/8 Primary and functional tics 1/8 |
| Case 12 | Primary Tics/ Tourette Syndrome 5/8 Primary and functional tics 2/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 3/8 Functional tics 5/8 |
| Case 13 | Primary Tics/ Tourette Syndrome 3/8 Primary and functional tics 1/8 Functional tics 4/8 | Primary Tics/Tourette Syndrome 3/8 Primary and functional tics 4/8 Functional tics 1/8 |
| Case 14 | Primary Tics/ Tourette Syndrome 7/8 Non-tic disorder 1/8 | Primary Tics/Tourette Syndrome 7/8 Non-tic disorder 1/8 |

| | | |
|----------------|---|--|
| Case 15 | Primary Tics/ Tourette Syndrome 8/8 | Primary Tics/Tourette Syndrome 8/8 |
| Case 16 | Primary Tics/ Tourette Syndrome 5/8 Primary and functional tics 2/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 2/8 Primary and functional tics 2/8 Functional 4/8 |
| Case 17 | Primary Tics/ Tourette Syndrome 1/8 Primary and functional tics 1/8 Functional tics 6/8 | Primary Tics/Tourette Syndrome 1/8 Primary and functional tics 5/8 Functional tics 2/8 |
| Case 18 | Primary Tics/ Tourette Syndrome 4/8 Primary and functional tics 2/8 Functional tics 2/8 | Functional tics 8/8 |
| Case 19 | Primary Tics/ Tourette Syndrome 6/8 Functional tics 2/8 | Primary Tics/Tourette Syndrome 5/8 Primary and functional tics 3/8 |
| Case 20 | Primary Tics/ Tourette Syndrome 7/8 Non-tic disorder 1/8 | Primary Tics/Tourette Syndrome 8/8 |
| Case 21 | Primary Tics/ Tourette Syndrome 8/8 | Primary Tics/Tourette Syndrome 7/8 Primary and functional tics 1/8 |
| Case 22 | Primary Tics/ Tourette Syndrome 8/8 | Primary Tics/Tourette Syndrome 8/8 |
| Case 23 | Primary Tics/ Tourette Syndrome 4/8 Primary and functional tics 3/8 Functional tics 1/8 | Primary Tics/Tourette Syndrome 1/8 Primary and functional tics 6/8 Functional tics 1/8 |
| Case 24 | Primary Tics/ Tourette Syndrome 2/8 Primary and functional tics 1/8 Functional tics 5/8 | Primary and functional tics 5/8 Functional tics 3/8 |