

Creating a Patient-Based Diagnostic Checklist for Functional Tics During the COVID-19 Pandemic

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Abstract

Background and Objectives

Since the onset of the COVID-19 pandemic, there has been a dramatic change in the presentation of patients with tics. The explosive presentation of atypical tics (TT) has been noted worldwide and thought to be the manifestation of a pandemic-associated functional neurologic disorder following social media exposure to tics. Nevertheless, despite the frequent diagnosis of functional tics (FT), there are no existing formal diagnostic criteria. The primary aim of this study was to create a patient-based diagnostic checklist for making the diagnosis of a functional tic disorder (FTD) during the COVID-19 pandemic.

Methods

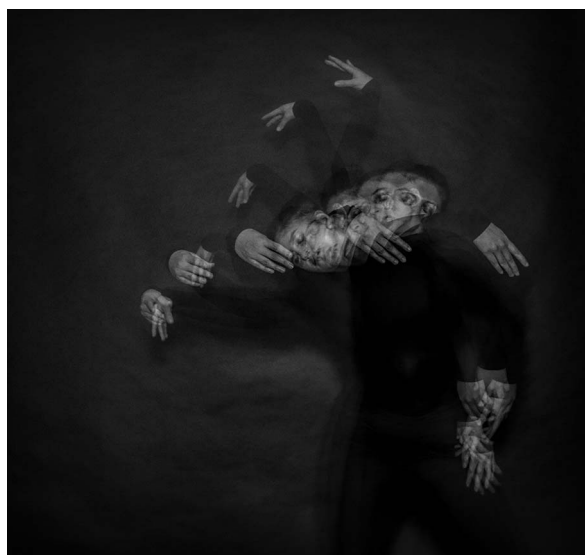
A retrospective chart review at a single institution during the pandemic was performed. Based on the available literature, diagnostic criteria were created for TT, FT, and patients with dramatically evolving symptoms (i.e., mixed with prior history of mild tics with later fulminant functional worsening). Patient demographics, comorbidities, and tic characteristics of these groups were then compared. Following initial assessments, new diagnostic criteria were established and statistically reanalyzed.

Results

One hundred ninety-eight patients underwent investigation. Significant differences in age, sex, psychological comorbidities, tic characteristics, and tic severity were found between patients with TT when compared with either of the 2 the functional groups. Only the presence of rostrocaudal progression and increased obsessive-compulsive behaviors were significantly different between patients with new-onset FT and those with functional worsening of a previous tic disorder. Results also showed that age at tic onset was not a contributing factor for group differentiation. Many patients with FT were not exposed to videos depicting tics on social media.

Discussion

This study confirms the presence of a distinct presentation of aTT during the pandemic period. It further establishes the validity of specific criteria useful in dividing patients with tics into 3 formal diagnostic criteria: (1) primary tic disorders (PTDs), (2) a strictly FTD, and (3) a mixed tic disorder consisting of patients with an initial history of a PTD and the later development of FT. Explicit diagnostic criteria should enable clinicians and researchers to make a definitive identification and assist patients and families become more knowledgeable and accepting of the diagnosis of FT.



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Tics, defined as brief, rapid, abrupt, recurrent, and non-rhythmic movements or vocalizations, are a common movement disorder in children. The COVID-19 pandemic has, however, led to a significant alteration in the clinical presentation of many patients with tics and tic-like behaviors. For example, recent studies have linked the sudden onset of atypical motor and phonic tic-like behaviors in teenaged girls to the negative psychological effects of the pandemic and social media exposure.¹⁻³ This increase in atypical tics (TT) has emphasized the need for a practical criteria to assist the treating physician in deciding whether the movement or vocalization is a typical tic or a functional tic-like behavior. An additional deficiency is the lack of information on the pandemic's effect on the presentation, quality of symptoms, and outcome of children presenting with more TT.

Prepandemic studies have shown that tics typically present between 4 and 8 years of age, and 93% are symptomatic before age 10 years. Motor tics usually precede the appearance of phonic tics and are often localized to the face, head, or neck. Tics have a waxing and waning pattern with an evolving course along with fluctuating frequency and intensity. Brief exacerbations can be provoked by stress, anxiety, excitement, anger, fatigue, or infection. In contrast, tic suppressibility frequently occurs with concentration, participating in emotionally pleasing activities, or while asleep.⁴ A premonitory urge/sensation occurs in up to 95% of people with tic disorders, with most becoming aware of its presence by age 10 years.⁵ Peak tic severity is greatest between 10 and 12 years of age with subsequent improvement thereafter in about 70% of affected individuals.^{6,7} Chronic tic disorders (CTDs) are more frequent in males, require the presence of tics for greater than 1 year, have an age at onset before age 18 years, a waxing and waning course, and the absence of either a substance-induced or general medical condition causation for the tics. In addition to tics, 86% of patients with CTD have at least 1 psychiatric comorbidity, with attention deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder/obsessive-compulsive behaviors (OCBs) being the most common.⁸ Etiologically, tic disorders are epigenetic, and a positive family history of tics is present in about one-half of patients. Therapeutically, beneficial tic-suppressing approaches include behavioral therapy and a variety of pharmacologic agents including alpha-adrenergic agonists, topiramate, and typical and atypical antipsychotics.⁹

Tics and tic-like behaviors are also seen as a manifestation of a functional (psychogenic) movement disorder. For example, pre-COVID, functional movement disorders resembling tics were observed in 4.9% of 184 patients.¹⁰ More recently, during the COVID-19 pandemic, there has been a dramatic case increase of aTT resembling functional tic-like behaviors in teenagers and young adults, especially females, in numerous countries throughout the world.^{2,11,12} This acute and fulminant rise has occurred in children and adults with a

history of tics as well as in patients without preexisting tics.^{12,13}

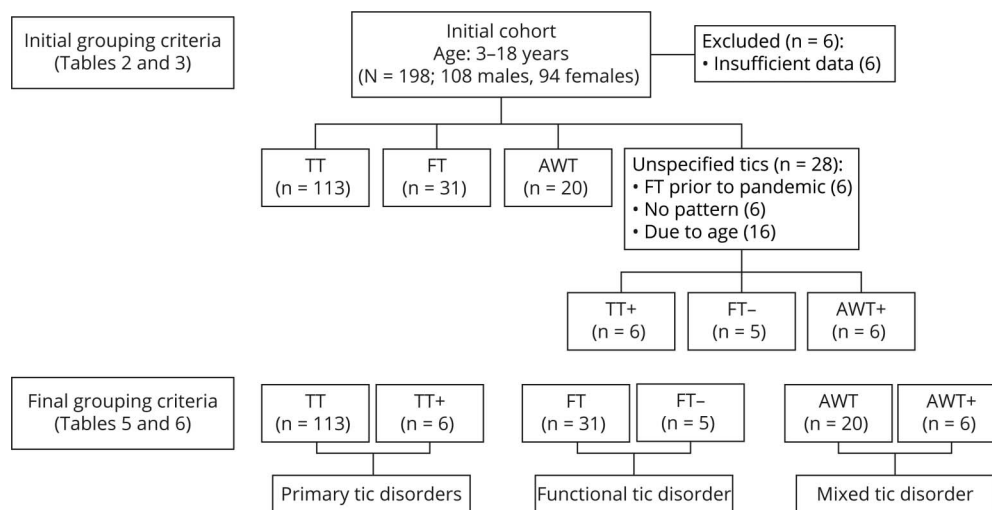
Despite neurologists frequently making a diagnosis of functional tics (FT), there are, to date, no formal diagnostic criteria. Prior investigators have suggested that several features can be helpful in differentiating FT from organic tics. These features include female preponderance, older age at presentation, abrupt onset, and lack of premonitory sensations.^{10,14,15} While recognizing that these and other criteria may be helpful, there remains a need for definitive and testable diagnostic criteria. Hence, recognizing the significant psychological effect of the COVID-19 pandemic, one goal of this study was to develop a diagnostic checklist to assist providers, researchers, and families in differentiating between organic and FT. A second major aim was to assess the effect of the COVID-19 pandemic on the presentation, comorbidities, and outcome of 198 consecutive children presenting with a chief complaint of tics.

Methods

A retrospective chart review of 198 consecutive children (age range 3–18 years) presenting with a chief complaint of tics was completed by 2 authors (S.P.T. and L.Q.). All patients were evaluated in person at a single institution (UNC Clinic for Tourette Syndrome and Tic Disorders) between May 1, 2020, and December 17, 2021. Data gathered at the time of initial evaluation included sex, age at onset of tics, tic characteristics, history of symptom worsening, presence of a premonitory urge and suppressibility, patient/family-reported presence of significant psychiatric comorbidities, family history of tics, and an initial Yale Global Tic Severity Score (YGTSS) and YGTSS-Total Tic Score (YGTSS-TTS). For patients who elected to start treatment for their tics, YGTSS and YGTSS-TTS were gathered at the follow-up visits. Study design is depicted in the Figure.

Initial Major Group Comparisons

Initial working criteria for differentiating patients into 3 categories were established by 2 authors (S.P.T. and H.S.S.) based on specific patient characteristics and specific tic characteristics from review of current literature (Table 1).^{14,15} The categories were as follows: (1) TT, (2) FT, and (3) acutely worsened tics (AWT, indicating patients with a prior history of TT and an acute, fulminant tic exacerbation following onset of the pandemic) with the criteria described in Table 2. Fulminant was defined as rapid progression in frequency and severity over less than 1 month. The analysis between these groups is presented in Table 3. A fourth unspecified tics group was created for patients not fulfilling the criteria for any of the aforementioned groups. Although coprolalia/coprophrenomenon and self-injurious tics can occur in patients with typical tic disorders, the combination of the 2 is less common. Hence, this combination was used as an exclusion criterion, labeling as unspecified, with subsequent revisions to be made as needed. Similarly, an age cutoff of



AWT = acutely worsened tics; AWT+ = typical tic onset <12 years with functional worsening ≥ 12 years; FT = functional tics; FT- = functional tic characteristics with tic onset <12 years; TT = typical tics; TT+ = typical tic characteristics with tic onset ≥12 years.

12 years was used in the initial working criteria as FT have previously been noted in older children; the potential for changing this age cutoff was planned for analysis in the next step. Patients whose complete tic history was unknown (for example, due to adoption) were excluded.

Absolute and relative frequencies were computed for each of the categorical variables with mean and SDs calculated for the quantitative variables (Table 3). Mean and SDs were calculated for continuous variables. The 3 groups were then compared in terms of the collected variables using the Kruskal-Wallis rank-sum test for continuous variables and Pearson χ^2 test for categorical variables. Multiple comparisons were performed using the Bonferroni correction. All statistical analyses were performed in R version 4.0.2 (R Core Team, 2020). Complete case analysis was considered, with $p < 0.05$ determining statistical significance.

Unspecified Group Comparisons

Following the initial analysis, 28 patients were noted to be in the unspecified category. Review of these patients revealed 3 distinct groups who otherwise would have met one of the

working criteria for TT, FT, or AWT except for the age cutoff criterion: (1) patients meeting all criteria for TT except with tic age at onset ≥12 years (TT+), (2) patients meeting all criteria for FT except tic age at onset ≤11 years (FT-), and (3) patients with onset of TT ≥12 years with later acute worsening (AWT+). A second analysis (Table 4) was undertaken to compare these secondary groups to the initial working criteria (TT vs TT+, FT vs FT-, and AWT vs AWT+).

Combined Comparisons (Major Group Plus Unspecified Group)

Following analysis of the unspecified group, reanalysis was conducted to compare the initial working criteria to the final group categories (TT and TT+ [primary tic disorders, PTDs], FT and FT- [functional tic disorder, FTD], AWT and AWT+ [mixed tic disorder, MTD]).

Standard Protocol Approvals, Registrations, and Patient Consents

The Institutional Review Board at the University of North Carolina Chapel Hill approved this study including a waiver of informed consent (IRB number 21-1907).

Table 1 Functional Tic Characteristics Defined in the Literature

1. Tic attacks (variable, very complex, and prolonged episodes of motor and vocal tic-like behavior)
2. Frequent coprolalia (obscene words or profanity) or coprophenomenon (obscene gestures)
3. Broad spectrum of word use (for example, atypical vocalizations such as beans, woowho, extended sentences, speaking in different accents, changing vocal pitch, and many others)
4. Blocking tics (including negative symptoms such as immobility of body parts, staring, and posturing)
5. Self-injurious tics or tics injurious to others
6. Presence of other functional neurologic symptoms (FNS; e.g., psychogenic nonepileptic seizures)

Table 2 Initial Working Criteria for Differentiating Between Typical Tics and Functional Tics

Diagnostic for typical tics (TT)
1. Must have age at onset <12 y
2. Must not be fulminant in onset
3. Must include at least 2 of the following patient characteristics:
1. Presence of a premonitory urge and/or suppressibility
2. Rostrocaudal progression
3. Presence of at least 1 co-occurring conditions (anxiety, ADHD, and OCB)
4. Positive family history
4. May only include one of the following:
1. Coprolalia or coprophenomenon
2. Self-injurious tics
5. Must not include:
1. Tic attacks
2. Blocking tics
3. Broad spectrum of word use or frequent coprolalia
4. Throwing
5. Presence of other FNS
6. Must not be associated with drug use of underlying medical condition
Diagnostic for functional tics (FT):
1. Must have age at onset >12 y
2. Must be sudden and fulminant in onset (rapid progression in frequency and severity over less than 1 month)
3. Must include at least one of the following patient characteristics:
1. Comorbid anxiety
2. Female sex
3. Lack of family history
4. Lack of premonitory urge and suppressibility
4. Must have at least 2 of the following:
1. Tic attacks
2. Blocking tics
3. Frequent coprolalia or coprophenomenon
4. Broad spectrum of word use or frequent coprolalia
5. Self-injurious tics or tics injurious to others
6. Throwing
7. Presence of other functional neurologic symptoms (FNS)
Diagnostic for acutely worsened tics (AWT)
1. Meets all criteria for TT including age at onset <12 y
2. Develops at least 2 of the following >12 y of age

Table 2 Initial Working Criteria for Differentiating Between Typical Tics and Functional Tics (*continued*)

1. Tic attacks
2. Blocking tics
3. Frequent coprolalia or coprophenomenon
4. Broad spectrum of word use or frequent coprolalia
5. Self-injurious tics or tics injurious to others
6. Throwing
7. Presence of other FNS
Diagnostic for unspecified tics (UT): does not meet any of the above criteria

Data Availability

Anonymized data not published within this article will be made available by request to the first author.

Results

Of 198 patients initially included, 6 patients were excluded due to lack of full tic history. One hundred thirteen patients met the initial criteria for TT, 31 patients met the criteria for FT, and 20 patients met the criteria for AWT. Twenty-eight patients were initially in the unspecified group, which was subsequently subdivided into 3 groups (TT+, n = 6; FT-, n = 5; and AWT+, n = 5). Following this subdivision, 6 patients still did not meet the diagnostic criteria. Five patients met all criteria for FT but were excluded as their FT started prior to onset of the pandemic.

Initial Major Group Comparisons

Data comparing TT, FT, and AWT are presented in Table 3. A significant difference was found between TT and FT as well as between TT and AWT with regard to sex (more frequently female with FT and AWT), family history (more frequent with TT and AWT), presence of other significant self-reported mood symptoms and disorders such as ADHD and OCB (more frequent with FT and AWT), and initial YGTSS and YGTSS-TTS (higher with FT and AWT). Rostrocaudal progression was noted to be significantly different between all groups (most commonly seen with TT and least commonly seen with FT). A significant difference was also found in the presence of a premonitory urge or suppressibility with higher frequency being seen in the AWT group when compared with TT and FT.

Additional differences in tic characteristics between the groups included coprophenomenon (less frequent with TT), broad or extended word use (less frequent with TT), self-injurious tics or tics injurious to others (less frequent with TT), throwing (completely absent with TT), and presence of other functional neurologic symptoms (FNS; absent with TT). Tic attacks and blocking tics were absent from the TT

Table 3 Analysis of the Working Criteria

Characteristic	TT N = 113	FT N = 31	AWT N = 20	p Value
Age at tic onset Mean (SD)	5 (2)	14 (2)	8 (3)	<0.001 ^a ◆♣
Sex				<0.001 ^a ◆
Male	73 (65%)	1 (3.2%)	5 (25%)	
Female	40 (35%)	30 (97%)	15 (75%)	
Family history	48 (43%)	6 (19%)	10 (50%)	0.035
Premonitory urge or suppressibility	76 (67%)	24 (77%)	19 (95%)	0.030
Social media exposure				0.001 ^a ◆
None	11 (92%)	4 (31%)	0 (0%)	
Yes, prior to tic onset	1 (8.3%)	8 (62%)	3 (60%)	
Yes, but only after tic onset	0 (0%)	1 (7.7%)	2 (40%)	
Unknown	101	18	15	
Tic attack	0 (0%)	21 (68%)	10 (50%)	<0.001 ^a ◆
Frequent coprophenomenon	2 (1.8%)	8 (26%)	6 (30%)	<0.001 ^a ◆
Broad/extended word use	2 (1.8%)	22 (71%)	14 (70%)	<0.001 ^a ◆
Blocking	0 (0%)	4 (13%)	6 (30%)	<0.001 ^a ◆
Self/other injurious	1 (0.9%)	19 (61%)	9 (45%)	<0.001 ^a ◆
Throwing	0 (0%)	6 (19%)	3 (15%)	<0.001 ^a ◆
Other FNS	0 (0%)	5 (16%)	3 (15%)	<0.001 ^a ◆
Rostrocaudal progression	90 (80%)	1 (3.2%)	7 (35%)	<0.001 ^a ◆♣
Anxiety	78 (69%)	28 (90%)	20 (100%)	0.001◆
ADHD	91 (81%)	21 (68%)	14 (70%)	0.2
OCB	64 (57%)	18 (58%)	19 (95%)	0.005◆♣
Initial YGTSS-TTS	14 (7)	30 (14)	29 (12)	<0.001 ^a ◆
Initial YGTSS-TOTAL	22 (15)	54 (25)	53 (27)	<0.001 ^a ◆
Follow-up YGTSS-TTS	12 (7)	28 (13)	33 (12)	<0.001 ^a ◆
No follow-up	95	18	13	
Follow-up YGTSS-TOTAL	17 (13)	53 (26)	63 (25)	<0.001 ^a ◆
No follow-up	96	18	13	
Response to treatment	12 (67%)	3 (23%)	0 (0%)	0.003◆
No follow-up	95	18	13	

Abbreviations: ADHD = attention deficit hyperactivity disorder; AWT = acutely worsened tics; FNS = functional neurologic symptoms; FT = functional tics; OCB = obsessive-compulsive behavior; TT = typical tics; YGTSS-TTS = Yale Global Tic Severity Score–Total Tic Score, response to treatment: $\geq 25\%$ improvement in YGTSS-TTS from initial to subsequent visit.

◆ Bonferroni-adjusted *p* value for comparison TT \times AWT < 0.05 .

♣ Bonferroni-adjusted *p* value for comparison FT \times AWT < 0.05 .

^a Bonferroni-adjusted *p* value for comparison TT \times FT < 0.05 .

group by definition. There were no patients who had both self-injurious tics and coprolalia that would otherwise have met the criteria for TT. A significant difference in response to pharmacologic treatment (defined as $\geq 25\%$ improvement

between initial and subsequent YGTSS-TTS) was found between TT and AWT with a strong tendency toward lack of improvement in the FT group compared with TT. No differences were found between FT and AWT in any categories except

Table 4 Comparison of the Working Criteria Without Regard to Age

Characteristic	TT N = 113	TT+ N = 6	p Value	FT N = 31	FT-N = 5	p Value	AWT N = 20	AWT+ N = 5	p Value
Age at tic onset	5 (2)	13 (1)	<0.001	14 (2)	11 (0)	0.027	8 (3)	12 (0)	0.002
Sex			>0.99			>0.99			>0.99
Male	73 (65%)	5 (83%)		1 (3.2%)	1 (20%)		5 (25%)	2 (40%)	
Female	40 (35%)	1 (17%)		30 (97%)	4 (80%)		15 (75%)	3 (60%)	
Family history	48 (43%)	1 (17%)	>0.99	6 (19%)	1 (20%)	>0.99	10 (50%)	2 (40%)	>0.99
Premonitory urge or suppressibility	76 (67%)	4 (67%)	>0.99	24 (77%)	2 (40%)	0.70	19 (95%)	5 (100%)	>0.99
Social media exposure			>0.99			>0.99			0.15
None	11 (92%)	1 (50%)		4 (31%)	0 (0%)		0 (0%)	1 (100%)	
Yes, prior to tic onset	1 (8.3%)	1 (50%)		8 (62%)	2 (67%)		3 (60%)	0 (0%)	
Yes, but only after tic onset	0 (0%)	0 (0%)		1 (7.7%)	1 (33%)		2 (40%)	0 (0%)	
Unknown	101	4		18	2		15	4	
Tic attack	0 (0%)	1 (17%)	0.12	21 (68%)	3 (60%)	>0.99	10 (50%)	1 (20%)	>0.99
Frequent coprophenomenon	2 (1.8%)	0 (0%)	>0.99	8 (26%)	0 (0%)	>0.99	6 (30%)	0 (0%)	>0.99
Broad/extended word use	2 (1.8%)	0 (0%)	>0.99	22 (71%)	4 (80%)	>0.99	14 (70%)	3 (60%)	>0.99
Blocking	0 (0%)	0 (0%)	>0.99	4 (13%)	0 (0%)	>0.99	6 (30%)	0 (0%)	>0.99
Self/other injurious	1 (0.9%)	0 (0%)	>0.99	19 (61%)	2 (40%)	>0.99	9 (45%)	2 (40%)	>0.99
Throwing	0 (0%)	0 (0%)		6 (19%)	0 (0%)	>0.99	3 (15%)	1 (20%)	>0.99
Other FNS	0 (0%)	0 (0%)		5 (16%)	1 (20%)	>0.99	3 (15%)	1 (20%)	>0.99
Rostrocaudal progression	90 (80%)	5 (83%)	>0.99	1 (3.2%)	0 (0%)	>0.99	7 (35%)	3 (60%)	>0.99
Anxiety	78 (69%)	5 (83%)	>0.99	28 (90%)	4 (80%)	>0.99	20 (100%)	5 (100%)	
ADHD	91 (81%)	5 (83%)	>0.99	21 (68%)	4 (80%)	>0.99	14 (70%)	3 (60%)	>0.99
OCB	64 (57%)	3 (50%)	>0.99	18 (58%)	2 (40%)	>0.99	19 (95%)	5 (100%)	>0.99
Initial YGTSS-TTS	14 (7)	14 (9)	>0.99	30 (14)	26 (12)	>0.99	29 (12)	21 (8)	0.57
Initial YGTSS-TOTAL	22 (15)	25 (16)	>0.99	54 (25)	42 (24)	0.93	53 (27)	43 (20)	>0.99
Follow-up YGTSS-TTS	12 (7)	16 (6)	>0.99	28 (13)	0 (NA)	0.16	33 (12)	18 (4)	0.37
No follow-up	95	4		18	4		13	3	
Follow-up YGTSS-TOTAL	17 (13)	31 (13)	0.47	53 (26)	0 (NA)	0.22	63 (25)	32 (11)	0.46
No follow-up	96	4		18	4		13	3	
Response to treatment	12 (67%)	0 (0%)	0.86	3 (23%)	1 (100%)	>0.99	0 (0%)	2 (100%)	0.13
No follow-up	95	4		18	4		13	3	

Abbreviations: ADHD = attention deficit hyperactivity disorder; AWT = acutely worsened tics; AWT+ = acutely worsened tics ≥ 12 y; FNS = functional neurologic symptoms; FT = functional tics; FT- = functional tics < 12 y; OCB = obsessive-compulsive behavior; TT = typical tics; TT+ = typical tics ≥ 12 y; YGTSS-TTS = Yale Global Tic Severity Score–Total Tic Score, response to treatment: $\geq 25\%$ improvement in YGTSS-TTS from initial to subsequent visit.

presence of rostrocaudal progression (more common with AWT) and presence of OCB (more common with AWT).

Unspecified Group Comparisons

During the data review, 28 patients were noted to be in the unspecified category with 3 distinct presentations noted who

would have otherwise met one of the working criteria except for the age at tic onset excluding them from a group. Analysis of these 3 groups in comparison to patients with TT, FT, and AWT is shown in Table 4. No statistically significant differences were noted between the individually compared groups (TT vs TT+; FT vs FT-; AWT vs AWT+).

Table 5 Proposed Criteria for Differentiating PTDs From FTDs

Diagnostic for primary tic disorder (PTD)
1. Must not be fulminant in onset (rapid progression in frequency and severity over less than one month)
2. Must include at least 2 of the following patient characteristics:
1. Rostrocaudal progression
2. Presence of at least 1 co-occurring conditions (anxiety, ADHD, and OCB)
3. Positive family history
3. May only include one of the following:
1. Coprolalia or coprophenomenon
2. Self-injurious tics or tics injurious to others
4. Must not include:
1. Tic attacks
2. Blocking tics
3. Broad spectrum of word use or frequent coprolalia
4. Throwing
5. Presence of other functional neurologic symptoms (FNS)
5. Must not be associated with drug use or underlying medical condition
Diagnostic for functional tic disorder (FTD)
1. Must be sudden and fulminant in onset
2. Must include at least one of the following patient characteristics:
1. Comorbid anxiety
2. Female sex
3. Lack of family history
3. Must have at least 2 of the following:
1. Tic attacks
2. Blocking tics
3. Frequent coprolalia or coprophenomenon
4. Broad spectrum of word use or frequent coprolalia
5. Self-injurious tics or tics injurious to others
6. Throwing
7. Presence of other FNS
Diagnostic for mixed tic disorder (MTD)
Meets all criteria for PTD with subsequent fulminant development of at least 2 of the following:
1. Tic attacks
2. Blocking tics
3. Frequent coprolalia or coprophenomenon
4. Broad spectrum of word use or frequent coprolalia

Table 5 Proposed Criteria for Differentiating PTDs From FTDs (continued)

5. Self-injurious tics or tics injurious to others
6. Throwing
7. Presence of other FNS

Combined (Major Group Plus Unspecified Group Comparisons)

A third comparison was undertaken to confirm that results obtained after combining the similar groups (TT combined with TT+ [PTD], FT combined with FT- [FTD], and AWT combined with AWT+ [MTD]). Comparison of the initial groups (TT, FT, and AWT) to the proposed diagnostic groups (PTD, FTD, and MTD; criteria in Table 5) is presented in Table 6 and shows no statistically significant differences. Six patients did not meet any criteria due to having only tic attacks or due to lack of adequate information to classify them into specific groups.

Discussion

Since the onset of the COVID-19 pandemic and international lockdowns in early 2020, clinicians worldwide have reported worsening of tics in patients previously diagnosed with PTDs including Tourette syndrome, chronic motor/vocal tic disorders, and provisional tic disorder. Parental reports from Italy demonstrated that 67% of children with PTD demonstrated worsening of their underlying tic disorder.¹ Forty-nine percent of adult patients were noted to have a little worsening of their tics or much worse tics following the onset of the pandemic.¹³ In addition, an explosion of tics and tic-like movements has been reported in teenagers who had never before experienced tics.^{11,12}

Teenagers with the fulminant onset of tics following the pandemic onset were primarily females with a history of anxiety and with onset of their tics after the typical age at tic onset. It was also noted that they had very complex tic-like movements and vocalizations. This presentation also mimicked the pattern of presentation reported in pre-COVID functional tic-like disorders. Additional characteristics noted in this functional group included a reduced family history of tics, a frequently absent premonitory urge, an inability to temporarily suppress their tics, a greater tic suggestibility (i.e., an exacerbation of symptoms when discussing or observing), and a greater frequency of coprophenomenon than observed in patients with CTDs.¹⁴⁻¹⁷ Although each of these clinical characteristics is helpful in diagnosing patients with FT, a formal diagnostic checklist for differentiating between typical and FT does not yet exist.

The initial phase in this study was the gathering of information on children presenting with tics to a single

Table 6 Comparison of the Proposed Criteria for Differentiating PTDs From Functional Tic Disorders

Characteristic	PTD N = 119	FTD N = 36	MTD N = 25	p Value
Age at tic onset	6 (3)	14 (2)	9 (3)	<0.001
Sex				<0.001
Male	78 (66%)	2 (5.6%)	7 (28%)	
Female	41 (34%)	34 (94%)	18 (72%)	
Family history	49 (42%)	7 (19%)	12 (48%)	0.031
Premonitory urge/suppressibility	80 (67%)	26 (72%)	24 (96%)	0.014
Social media exposure				0.003
None	12 (86%)	4 (25%)	1 (17%)	
Yes, prior to tic onset	2 (14%)	10 (62%)	3 (50%)	
Yes, but only after tic onset	0 (0%)	2 (12%)	2 (33%)	
Unknown	105	20	19	
Tic attack	0 (0%)	24 (67%)	11 (44%)	<0.001
Frequent coprophenomenon	2 (1.7%)	8 (22%)	6 (24%)	<0.001
Broad/extended word use	2 (1.7%)	26 (72%)	17 (68%)	<0.001
Blocking	0 (0%)	4 (11%)	6 (24%)	<0.001
Self/other injurious	1 (0.8%)	21 (58%)	11 (44%)	<0.001
Throwing	0 (0%)	6 (17%)	4 (16%)	<0.001
Other FNS	0 (0%)	6 (17%)	4 (16%)	<0.001
Rostrocaudal progression	95 (80%)	1 (2.8%)	10 (40%)	<0.001
Anxiety	83 (70%)	32 (89%)	25 (100%)	<0.001
ADHD	96 (81%)	25 (69%)	17 (68%)	0.2
OCB	67 (56%)	20 (56%)	24 (96%)	<0.001
Initial YGTSS-TTS	14 (7)	29 (13)	27 (12)	<0.001
Initial YGTSS-TOTAL	22 (15)	53 (25)	51 (26)	<0.001
Follow-up YGTSS-TTS	13 (7)	26 (14)	29 (12)	<0.001
No follow-up	99	22	16	
Follow-up YGTSS-TOTAL	18 (13)	49 (29)	56 (26)	<0.001
No follow-up	100	22	16	
Response to treatment	12 (60%)	4 (29%)	2 (22%)	0.076
No follow-up	99	22	16	

Abbreviations: ADHD = attention deficit hyperactivity disorder; FNS = functional neurologic symptoms; FTD = functional tic disorder; MTD = mixed tic disorder; OCB = obsessive-compulsive behavior; PTD = primary tic disorder; YGTSS-TTS = Yale Global Tic Severity Score–Total Tic Score, response to treatment: $\geq 25\%$ improvement in YGTSS-TTS from initial to subsequent visit.

institution's pediatric neurology clinic. Using criteria suggested in the literature,^{14,15} children were then divided into 4 categories (Table 2) including TT, FT, patients with initially TT and subsequent fulminant exacerbation (AWT), and unspecified tics for patients who failed to fulfill the other criteria. Data from this initial comparison (Table 3) identified both similarities and differences from prior reports.

Similar to prior reports, patients with FT were overwhelmingly female compared with TT (97% vs 35%). A greater female prevalence was also noted in 75% of children who developed FT after a period of TT. A family history of tics was present in 43% of patients with TT and 19% of FT. Family history positivity in the AWT group was similar to those with TT, likely reflecting both having a common early typical tic presentation.

Compiled movement and vocalization data (tic attacks, coprophobia, extended word use, injurious behaviors, and throwing objects) were significant differentiating features between groups. No patients having both self-injurious tics and coprophobia met the criteria for TT signifying that this criterion is useful in differentiating between TT and FT in most patients. Rostrocaudal progression was more prevalent in patients with TT when compared with FT and AWT. Furthermore, there was also a significant difference in the presence of rostrocaudal progression between AWT and FT likely attributable to patients with AWT having an initial history of TT. Prior publications have emphasized that teenagers with psychiatric disorders are more likely to develop functional neurologic disorders.¹⁸ In this report, the presence of self-reported anxiety was significantly elevated in children with FT and AWT compared with TT, and the incidence of obsessive-compulsive behaviors was more common in patients with AWT than either of the other groups, which likely indicates that patients with additional psychological comorbidities are at an increased risk for developing functional disorders.

Overall tic severity as judged by YGTSS and YGTSS-TTS was significantly elevated in the patients with FT when compared with patients with TT. Therapeutically, this study broadly confirmed suggestions that pharmacotherapy, using recommended tic-suppressing medications (alpha agonists, atypical antipsychotics, or topiramate), is not as beneficial in children within FT and AWT groups compared with TT.

Age has also traditionally been used to categorize individuals into a typical tic or functional tic diagnosis. More specifically, teenagers and adults more commonly have functional neurologic disorders compared with younger children.^{14,18} In this study, the age of 12 years was initially used as a cutoff point to define TT and FT/AWT. Nevertheless, 16 patients had an age at onset that resulted in them being placed in the unspecified category. Thus, given these results, a second analysis was performed without using age as a specifying diagnostic factor. New groups included (a) patients with TT, using the working criteria, but with age at onset ≥ 12 years (labeled as TT+, 6 patients); (b) patients solely functional (FT) but with tic onset less than age 12 years (FT-, 5 patients), and (c) patients who had their initial TT present after age 12 years and later developed AWT (AWT+, 5 patients). A statistical comparison between these new groups and the initial groups (TT vs TT+, FT vs FT-, and AWT vs AWT+) was performed and is shown in Table 4. No significant differences were found, which suggest that age at tic onset should not be required as a factor in making a diagnosis of FT.

The premonitory urge is a vaguely defined internal tension, pressure, itch, or feeling that occurs prior to an individual's tic.¹⁹ Prior investigations in typical CTDs suggest that about 90% of older children/adults and 37% of younger children have a premonitory urge or sensation just prior to their

motor or phonic tic.^{20,21} This premonitory urge has been described as having several roles including being the driving force for tics, a diagnostic clue for the diagnosis of tics, a predictor in childhood of a poorer health-related quality of life as an adult with TS, and an important component of behavioral therapy.^{22,23} In addition, many individuals with TT report an ability to briefly suppress their tics, which has previously been reported to be reduced in patients with FT.¹⁵ In this report, patients' responses to questions pertaining to suppressibility indicated that children in the FT and AWT groups did show ability to temporarily suppress their tics. In this study, in distinct contrast to prior publications, children in FT and AWT categories tended to have a greater, not reduced, presence of premonitory urges or suppressibility. Therefore, it is suggested that neither premonitory urge nor tic suppressibility should be a required factor to make a formal diagnosis of FT.

Recognizing that a second study goal was to develop useful diagnostic criteria for separating typical tic disorders and functional tic-like disorders, the combined group of TT and TT+ was relabeled as PTD, the combined group of FT and FT- as FTD, and the combined group of AWT and AWT+ as MTD. Proposed criteria listed for these disorders are listed in Table 5. After applying these new diagnostic criteria, statistical separation between PTD and FTD groups was at its maximum (Table 6). Only 6 of the 187 analyzed patients (3.2%) failed to meet the specific diagnostic criteria for PTD, FTD, or MTD.

Etiologically, the link of FTD to the COVID-19 pandemic is not via a personal infection but rather by the pandemic's significant negative effect on mental health and daily activities.^{11,24} In addition, exposure to social media is proposed as a promoting factor in susceptible patients. For example, patients with FT following onset of the pandemic have reported high rates (up to 100%) of exposure to videos of patients with tics on social media (typically TikTok or YouTube).^{12,25,26} This phenomenon has been attributed to as a new mass sociologic illness by several authors and has even made its way into mainstream discussion.^{2,3,27} Patients in our study were not routinely asked about exposure to online tic-containing videos until mid-August 2021. Despite the resulting reduced sample size, patients with FTD and MTD were significantly more likely to have seen videos of tics on social media than patients with PTD. Nevertheless, 8 patients with FTD (31% of those asked) reported never having seen videos of tics on social media, and 1 patient (8%) had seen these videos only after her tics developed. In addition, 2 of 5 patients with MTD reported only seeing these videos after onset of their functional worsening. In summary, while exposure to social media videos is a risk factor, recognizing that not all patients have seen these videos, the authors believe that the term TikTok tics is inappropriate as a catch-all term. A recommended term for patients with FT in the setting of the pandemic (with or without exposure to videos on social media) is COVID-19-related FTD. Using

this term describes the phenomenon more accurately and avoids any negative or pejorative connotations that the term TikTok tics may give to the patient or family.

Historically, it is likely that additional outbreaks of FT will occur. For example, an outbreak of FT occurred in Le Roy, New York, in 2011–2012. Authors have also proposed that the abnormal behaviors demonstrated by the witches in Salem, Massachusetts, in the 1690 s may have represented a functional neurologic disorder or FTD.^{28,29} The term COVID-19–related FTD would therefore be a specific term representing FT brought on in the setting of the COVID-19 pandemic. Terminology in the future for such outbreaks could be determined on a case-by-case basis or could simply be referred to as stress-related FTD. For the individual patient with FT in the absence of an outbreak, the diagnosis of FTD can suffice.

Limitations of this study include its retrospective nature, relatively short longitudinal follow-up period, and potential loss of patients during the follow-up. These factors, especially in patients enrolled toward the end of the recruitment period, may have limited the power to detect small clinical differences and responses to treatment. This study also used patient-reported symptoms of ADHD, anxiety, and obsessive-compulsive behaviors, rather than standardized confirmatory psychiatric questionnaires. Unfortunately, clinician-confirmed scales were not obtained at the initial visit. The retrospective nature of the study also prevented the authors from later gathering these rating scales. It is suggested that future studies determine whether self-reported symptoms are sufficient or whether clinician-confirmed diagnoses are required for the diagnosis of FTD/MTD. The article was also limited by an inability to compare data prior to onset of the pandemic as the clinic was not formed until May 1, 2020. The broad finding that many children may not have seen videos of tics and tic-like behaviors prior to onset of their tics could be limited by either poor recall or by children not being truthful during their initial visit. This may be especially true in those children whose FT are identical to the ones seen on such videos, but the number of such children is not easily quantifiable. In addition, many children have FT that have similar, but not identical, characteristics, which is much more difficult to attribute strictly to social media exposure. Finally, this study did not assess the relative benefits of different therapeutic agents or evaluate the potential beneficial effect of behavioral therapy.

Fully recognizing these limitations, this study provides a framework for the clinical diagnosis of patients with various tic presentations and allows clinicians to differentiate between patients with FT and those with organic tics. In addition, since the onset of the COVID-19 pandemic, it is one of the largest studies focusing on patients presenting with tics, and the only one that has directly compared both tic and tic-like patients presenting during the pandemic period (May 1, 2020, through December 17, 2021). The newly proposed

criteria also remove diagnostic uncertainty from nearly all patients in this cohort, recognizing that only 6 of 187 patients (3.2%) failed to fit into 1 of the 3 final categories. The authors are optimistic that additional studies in other centers will confirm the validity of the presented proposed criteria for PTD, FTD, and MTD.

The Diagnostic and Statistical Manual of Mental Health, Fifth Edition (DSM-5) has diagnostic criteria for multiple typical tic disorders. These include (1) provisional tic disorder (tics for less than 1 year), (2) chronic motor or vocal tic disorder (having either motor or vocal tics, but not both, for greater than 1 year), and (3) Tourette syndrome (requiring the presence of at least 2 motor tics and 1 vocal tic with persistence of tics for more than 1 year). Common to all of these previously described disorders is that the tics must not be caused by another medical condition or medication. The proposed diagnoses in this study are not designed to alter established diagnoses but rather to create criteria that can effectively differentiate between primary tics disorders and FTDs. Without a diagnostic checklist, multiple patients with FT were likely incorrectly diagnosed with a PTD. Formal diagnostic criteria can enhance the ability of a physician or other practitioner to make a diagnosis without preexisting ambiguity.

Therapeutically, several treatment modalities are available for patients with functional neurologic symptom disorder (FNSD). A recent review has summarized these treatments, which are largely focused on psychological and behavioral therapies rather than medical management. Psychoeducation, cognitive behavioral therapy, behavioral modifications, family-centered therapy, and physical therapy have all been evaluated and shown to be effective. Research has also suggested that multidisciplinary and inpatient rehabilitation may be more effective in treating FNSD.^{30,31} However, none of these treatment options have been formally assessed in patients with FT/FTD. The lack of response to antitonic medications for the treatment of FT has been previously suggested and is confirmed in this study.¹² At present, recommend treatments tend to focus on psychological and behavioral modifications similar to treatment of other FNS. Comprehensive behavioral intervention for tics has also been frequently recommended.^{11,12,32} Despite these recommendations, no studies to date have examined the effectiveness of various behavioral therapies in patients with FT.

The long-term outlook for patients with COVID-19–related FTD is unknown at this time. Nevertheless, based on the prior reported spontaneous resolution in other outbreaks of FT/neurologic symptoms, the authors are optimistic regarding the long-term improvement of tics in these patients. Anecdotally, the authors have had several patients with vast improvement, if not complete resolution, of their tics associated with life returning toward normal and the return to school.

In summary, while the proposed criteria for distinguishing between PTDs and functional/MTDs were developed

TAKE-HOME POINTS

- There has been an explosion of atypical tics and tic-like behaviors since the onset of the COVID-19 pandemic.
- These atypical tics have characteristics similar to those previously described as functional tic-like behavior.
- This is the first study to propose a diagnostic checklist for making a formal diagnosis of FT, which could be known as FTD.
- Given that many patients diagnosed with FT during the pandemic have never been exposed to videos of influencers with tics on social media, the term TikTok tics may be inappropriate to describe the complete phenomenon; a preferred term is COVID-19-related FTD.
- For similar outbreaks in the future, terminology could be determined on a case-by-case basis, or the term stress-related FTD could be used.

during the COVID-19 pandemic, they will likely be useful in the future for FTD associated with other contributing factors. Formal criteria are essential for making a correct diagnosis, implementing appropriate therapy, and investigating the underlying pathophysiology and mechanism of treatment response. We look forward to the results of future studies designed to investigate therapeutic approaches, long-term outcomes, the influence of coexisting conditions, and pathophysiologic mechanisms. In addition, and equally important to research and treatment, the proposed diagnostic criteria can also help patients and families become more comfortable with the diagnosis of FTD and assist patients in finding appropriate treatment for their condition.

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Luke Quehl, MD	Division of Child Neurology, Department of Neurology, University of North Carolina - Chapel Hill School of Medicine	Drafting/revision of the manuscript for content, including medical writing for content; Major role in the acquisition of data
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Feng-Chang Lin, PhD	Translational and Clinical Sciences Institute, University of North Carolina, Chapel Hill	Drafting/revision of the manuscript for content, including medical writing for content; Analysis or interpretation of data
Harvey S. Singer, MD	Johns Hopkins Medicine and the Kennedy Krieger Institute, Baltimore, MD	Drafting/revision of the manuscript for content, including medical writing for content; Study concept or design

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