

# Tourette Syndrome with Functional Overlay: A Case Series

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## Abstract

Functional tics have long been described as part of the much wider spectrum of functional neurological disorders with motor manifestations. Reports of functional tics have been relatively rare, until their recent increase on a global scale during the COVID-19 pandemic. Such reports have often been characterized by an acute or subacute onset of complex motor and vocal manifestations. Moreover, functional tics have predominantly been reported in adolescent females, whereas neurodevelopmental tics, such as those reported by patients with Tourette syndrome (TS), typically begin in younger boys. In addition to their marked severity and complexity, functional tics can merge into other types of functional neurological disorders. However, functional tics can also coexist with neurodevelopmental tics, as patients diagnosed with TS can present with a functional overlay (dual diagnosis). In the present study, we report the clinical characteristics of a case series of 10 patients diagnosed with both TS and functional tics during the COVID-19 pandemic. We also provide the first within-subject comparison between neurodevelopmental tics and functional tics, to assist clinicians in the differential diagnosis of patients with TS who developed a functional overlay during challenging times.

**Keywords:** Functional neurological disorder, functional tics, neurodevelopmental tics, tic disorders, tics, Tourette syndrome

## INTRODUCTION

Patients with functional tics typically present with complex motor and vocal tics that can phenomenologically resemble neurodevelopmental tics but have a different etiology, pathophysiology, and outcome.<sup>[1,2]</sup> Functional tics are part of the broader diagnostic category of functional neurological symptoms with motor manifestations and have been described as having a lower prevalence compared to other functional movement disorders, such as functional tremor and functional dystonia.<sup>[3]</sup> However, during the COVID-19 pandemic, there has been a surge in case reports of functional tics worldwide.<sup>[4]</sup> Both the index cases and the reported samples have often been female adolescents who experienced an acute or subacute onset of severe functional tics.<sup>[5]</sup> This contrasts with the gradual onset of tics in male children, which characterizes neurodevelopmental tics in patients with a diagnosis of Tourette syndrome (TS).<sup>[6,7]</sup> The pandemic itself and its effects on teenage lives have been cited among the many possible explanations for this phenomenon, alongside increased time spent on social media and exposure to influencers portraying themselves as having TS.<sup>[8]</sup>

In addition to their marked severity and complexity, functional tics can merge into other types of functional neurological disorders, including other functional movement disorders and nonepileptic attack disorders.<sup>[4]</sup> However, functional tics can also coexist with neurodevelopmental tics, as patients diagnosed with TS can present with a functional overlay (dual diagnosis).<sup>[4,9,10]</sup> Little is known about the clinical characteristics of patients with TS and functional overlay, as reports of functional tics in the scientific literature have been relatively scattered in time, until their recent increase on a global scale.<sup>[4,5]</sup>

In the present study, we report a case series of 10 consecutive patients diagnosed with both TS and functional tics during the COVID-19 pandemic. We also provide the first within-subject

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comparison between neurodevelopmental tics and functional tics, to assist clinicians in the differential diagnosis of patients with TS who developed a functional overlay during the challenging times of the global pandemic.

## MATERIALS AND METHODS

Between March 2020 and October 2022, we retrospectively reviewed the medical records of all consecutive patients attending the specialist TS Clinic, Department of Neuropsychiatry, National Centre for Mental Health, Birmingham (United Kingdom). Out of a clinical sample of 504 patients, we identified a group of patients ( $n = 10$ ) who fulfilled current diagnostic criteria for TS (multiple motor and vocal tics with onset in childhood or adolescence) and developed functional tics (functional overlay) during the COVID-19 pandemic. Patients with a limited understanding of English and patients with severe autism spectrum disorder/learning disability were excluded from our analysis. All patients provided informed consent to participate in the study, which was approved by the local section of the National Research Ethics Service.

Each patient underwent a comprehensive clinical assessment using the National Hospital Interview Schedule (NHIS) for TS.<sup>[11]</sup> The NHIS is a detailed semistructured interview schedule covering personal and family histories, as well as demographic details. The clinical variables systematically collected for this study included age at assessment, age at first tic onset, family history of tics, psychiatric comorbidities, and pharmacotherapy. Within each subject, both neurodevelopmental tics and functional tics were characterized according to their type, onset, course, and modulating factors.

The Diagnostic Confidence Index (DCI) was used to rate the clinician's confidence in diagnosing TS, according to the clinical characteristics of neurodevelopmental tics, including the presence of specific complex tics.<sup>[12]</sup> The DCI is a clinician-rated measure of the lifetime likelihood of TS diagnosis in patients presenting with tics. The DCI score is expressed as a percentage, with higher scores indicating higher confidence that the patient has TS.

Tic severity was measured using the Yale Global Tic Severity Scale (YGTSS), a clinician-rated scale assessing the severity of both motors, and vocal tics across five different domains: number, frequency, intensity, complexity, and interference.<sup>[13]</sup> Each domain is scored 0–5 and severity scores are separately calculated for motor and vocal tics by summing together the individual domain scores. Finally, the combined motor and vocal tic severity scores produce a total tic severity score. An additional measure of overall impairment is scored 0–50. These two scores are combined to produce the total YGTSS score, ranging from 0 to 100, with higher scores indicating increased tic severity.

Anonymized data were stored on Microsoft Excel 2019. The Statistical Package for the Social Sciences for Windows (SPSS Inc., Chicago, IL, USA, version 25) was used to perform all statistical analyses. This retrospective study was conducted

using descriptive statistics to illustrate the demographic and clinical characteristics of the participants. In addition, we used Fisher's exact test for dichotomous variables and the *t*-test for continuous variables to assess possible differences between neurodevelopmental tics and functional tics within the same subjects.

## RESULTS

Nine out of the 10 patients diagnosed with TS who developed an overlay of functional tics during the COVID-19 pandemic were female. The average age at assessment was 18 years (range 13–24 years). There was a significant difference in the age at the onset of neurodevelopmental tics (6 years, range 2–10 years) and functional tics (17 years, range 12–22 years) ( $P < 0.001$ ). The full demographic and clinical characteristics of the study sample are shown in Table 1.

In addition to functional tics, seven patients (70%) presented with other functional neurological symptoms, namely, nonepileptic attack disorder. Of these, three patients (30%) also had a functional weakness. At the time of the assessment, four patients (40%) had been prescribed pharmacotherapy: selective serotonin reuptake inhibitors ( $n = 2$ ), second-generation antidopaminergic agents ( $n = 1$ ), alpha-2 agonists ( $n = 1$ ), and beta-blockers ( $n = 1$ ).

Table 2 presents the within-subject comparison between the characteristics of neurodevelopmental tics and those of functional tics.

**Table 1: Demographic and clinical characteristics of the sample of patients with Tourette syndrome (neurodevelopmental tics) and functional overlay (functional tics) assessed at the specialist Tourette syndrome clinic during the COVID-19 pandemic ( $n=10$ )**

|                                     | <i>n</i> (%) |
|-------------------------------------|--------------|
| Age, mean (SD)                      | 18.4 (2.9)   |
| Female gender                       | 9 (90)       |
| Family history of tics              | 4 (40)       |
| OCD                                 | 3 (30)       |
| OCB                                 | 8 (80)       |
| ADHD                                | 3 (30)       |
| ASD                                 | 2 (20)       |
| Affective disorder                  | 5 (50)       |
| Anxiety disorder                    | 7 (70)       |
| Other FNS                           | 7 (70)       |
| Pharmacotherapy                     | 4 (40)       |
| DCI, mean (SD)                      | 76.7 (23.9)  |
| YGTSS tic severity score, mean (SD) | 25.4 (10.2)  |
| YGTSS impairment score, mean (SD)   | 17.0 (4.6)   |
| YGTSS total score, mean (SD)        | 42.4 (13.7)  |

OCD: Obsessive-compulsive disorder, OCB: Obsessive-compulsive behaviors, ADHD: Attention-deficit and hyperactivity disorder, ASD: Autism spectrum disorder, LD: Learning difficulty, FNS: Functional neurological symptoms, DCI: Diagnostic confidence index, YGTSS: Yale Global Tic Severity Scale, SD: Standard deviation

**Table 2: Characteristics of neurodevelopmental tics and functional tics of patients with Tourette syndrome and functional overlay assessed at the specialist Tourette syndrome clinic during the COVID-19 pandemic (n=10)**

| Clinical characteristics  | Neurodevelopmental tics, n (%) | Functionalities, n (%) | P <sup>a</sup> |
|---------------------------|--------------------------------|------------------------|----------------|
| Age at onset, mean (SD)   | 5.6 (2.3)                      | 16.9 (2.7)             | <0.001         |
| Simple motor tics         | 10 (100)                       | 6 (60)                 | 0.087          |
| Complex motor tics        | 6 (60)                         | 10 (100)               | 0.087          |
| Simple vocal tics         | 10 (100)                       | 6 (60)                 | 0.087          |
| Complex vocal tics        | 5 (50)                         | 8 (80)                 | 0.350          |
| Coprolalia                | 5 (50)                         | 8 (80)                 | 0.350          |
| Copropraxia               | 4 (40)                         | 5 (50)                 | 1              |
| Echolalia                 | 6 (60)                         | 6 (60)                 | 1              |
| Echopraxia                | 6 (60)                         | 6 (60)                 | 1              |
| Palilalia                 | 6 (60)                         | 6 (60)                 | 1              |
| Palipraxia                | 7 (70)                         | 7 (70)                 | 1              |
| Forced touching           | 6 (60)                         | 8 (80)                 | 0.629          |
| NOSIB                     | 2 (20)                         | 8 (80)                 | 0.023          |
| SIB                       | 3 (30)                         | 7 (70)                 | 0.179          |
| Rostrocaudal distribution | 10 (100)                       | 2 (20)                 | <0.001         |
| Acute/subacute onset      | 0                              | 8 (80)                 | <0.001         |
| Fluctuating course        | 9 (90)                         | 10 (100)               | 1              |
| Suppressibility           | 9 (90)                         | 7 (70)                 | 0.582          |
| Distractibility           | 9 (90)                         | 10 (100)               | 1              |

<sup>a</sup>Fisher's exact test for dichotomous variables and *t*-test for continuous variables. NOSIB: Nonobscene socially inappropriate behaviors, SIB: Self-injurious behaviors, SD: Standard deviation

Compared to neurodevelopmental tics, functional tics were significantly more likely to develop at an older age, acutely or subacutely, and without rostrocaudal distribution (all  $P < 0.001$ ). With regard to their clinical phenomenology, functional tics were significantly more likely to present with a range of nonobscene socially inappropriate behaviors ( $P < 0.001$ ). Finally, there was a trend for functional tics to be more likely to include fewer simple tics and more complex motor tics (all  $P = 0.087$ ).

## DISCUSSION

The aims of the present study were twofold. First, we reported the clinical characteristics of one of the largest samples of patients diagnosed with both TS and functional tics during the COVID-19 pandemic. Second, we provided the first within-subject comparison between neurodevelopmental tics and functional tics.

Apart from the gender distribution, the clinical characteristics of neurodevelopmental tics in our case series were representative of the wider population of patients with TS.<sup>[7]</sup> Specifically, the severity of neurodevelopmental tics according to YGTSS scores was in line with the results of the most recent large-scale examination and strategic revision of the YGTSS (mean YGTSS tic severity score: 25 vs. 24; mean YGTSS impairment score 17 vs. 23; mean YGTSS total score: 42 vs. 47).<sup>[13]</sup> The mean DCI score of our patients was 77, slightly higher than the average scores reported at specialist clinics. For example, the mean DCI score in the clinical sample of patients with TS who took part in the DCI validation study ( $n = 280$ ; mean age 19 years) was 61. This discrepancy

could be related to differences between the two samples in both ages range (13–24 years versus 3–70 years) and male:female ratio (1:9 vs. 4:1).<sup>[12]</sup>

Our findings were consistent with those reported during the recent surge in patients with functional tics during the COVID-19 pandemic. In addition to the striking preponderance of the female gender (90%) and the onset of functional tics during teenage years (mean age of 17 years), we found a psychiatric profile characterized by a higher prevalence of affective and anxiety disorders compared to other neurodevelopmental disorders.<sup>[14]</sup> Specifically, 70% of the patients with TS and functional overlay fulfilled current diagnostic criteria for an anxiety disorder and 50% for an affective disorder. The prevalence of comorbid attention-deficit and hyperactivity disorder and obsessive-compulsive disorder (both 30%) were broadly in line with clinical data from TS populations, taking into consideration that subthreshold tic-related obsessive-compulsive behaviors were reported by the vast majority of our patients (80%).<sup>[15]</sup>

Moreover, most of our patients (70%) presented with at least one different functional neurological disorder as part of their functional overlay. They all had a diagnosis of nonepileptic attack disorder, and almost half of them had also developed functional weakness. The clinical characteristics of comorbid nonepileptic attack disorder and functional weakness were overall comparable to those displayed by other patients diagnosed with functional neurological disorders at the same center and previously published.<sup>[16,17]</sup>

To the best of our knowledge, we reported the second-largest sample of patients with TS (neurodevelopmental tics) who

developed a functional overlay (functional tics) during the COVID-19 pandemic. Fremer *et al.*<sup>[18]</sup> recently reported data on 32 patients attending a specialist TS clinic in Germany who received a diagnosis of functional tics (“functional neurological disorder presenting with Tourette-like symptoms”) after exposure to relevant social media content between May 2019 and September 2021. Of these, 15 patients were identified as having a dual diagnosis of TS and functional tics. Fremer *et al.*<sup>[18]</sup> compared the characteristics of tics between their 15 patients with functional tics plus comorbid TS (“functional tics plus”) and 17 patients with functional tics but without TS (“functional tics only”). Patients in the “functional tics only” group reported significantly higher rates of abrupt onset and constant increase of symptoms, as well as a significantly lower rate of comorbid obsessive-compulsive behaviors, compared to the patients in the “functional tics plus” group.

We were able to partially replicate these findings by conducting a within-subject comparison between neurodevelopmental and functional tics in our smaller sample of patients with TS and functional overlay. Specifically, we found that functional tics were significantly more likely to develop acutely or subacutely, at an older age, and without the rostrocaudal distribution that characterizes neurodevelopmental tics. The clinical phenomenology of functional tics was also somewhat peculiar, as they were significantly more likely to include a range of nonobscene socially inappropriate behaviors (bizarre gestures and random words), compared to neurodevelopmental tics. A basic assessment of tic type (motor vs. vocal; complex vs. simple) could be less helpful in the differential diagnosis between neurodevelopmental and functional tics. For example, echophenomena, which have notoriously been associated with TS since its first description can be part of the repertoire of both neurodevelopmental and functional tics.<sup>[19]</sup> However, the trend for functional tics to be more likely to include fewer simple tics and more complex motor tics is consistent with findings from recent studies comparing the clinical characteristics of patients with TS and those of patients with functional tics only.<sup>[4,5,8]</sup>

This study has limitations. Although we recruited the second largest sample of TS with functional overlay developed during the COVID-19 pandemic, our sample size is still relatively small. Moreover, referral bias needs to be taken into account, as participants were recruited from a specialist clinic and may therefore not be representative of the wider community of patients with TS and functional neurological disorders. A further limitation of this study is intrinsic to its retrospective approach.

Specific criteria for the diagnosis of functional tics are currently being developed, partly in response to the rapid surge of case reports during the COVID-19 pandemic. Other areas for future research include the investigation of the etiological factors for functional tics and their complex interplay with the biological and neuropsychological substrates in patients with preexisting neurodevelopmental tics. Within the clinical continuum encompassing nonepileptic attack disorder and functional movement disorders (including functional weakness), it has been noted that etiological and mechanistic distinctions

might justify a separation between these conditions.<sup>[20]</sup> Further studies on larger samples are needed to better characterize the clinical phenotypes of TS associated with different functional neurological disorders.

In summary, our study provides novel data that can be helpful in refining the differential diagnosis of patients with TS who developed a functional overlay during the COVID-19 pandemic.<sup>[5]</sup> At the same time, our findings suggest new avenues for future research in this complex and potentially expanding patient population.

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### Conflicts of interest

There are no conflicts of interest.

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