TOURETTE'S SYNDROME (M YADEGAR AND F ESPIL, SECTION EDITORS)



Sudden Onset Tic and Tic-Like Presentations in Older Adolescents and Adults

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Abstract

Purpose of Review The past 3 years have seen an unprecedented increase in patients with functional tic-like behaviors (FTLB), a previously rare form of functional movement disorder (FMD) that can be mistaken for Tourette syndrome (TS). This article contrasts the patient characteristics, phenomenology, risk factors, and comorbidities of FTLB and TS to define criteria for differential diagnosis. Clinical issues, treatments, theoretical explanations, and future research questions are discussed.

Recent Findings FTLB predominately affect females, with a later onset of movements and vocalizations that are more complex, directional, severe, debilitating, and non-suppressible compared to TS. Psychosocial stressors from the pandemic, exposure to tic-content on social media, and comorbid anxiety and depression are etiological factors. Cognitive behavioral therapies appear to be effective treatment strategies.

Summary Creation of standardized clinical practice guidelines for diagnosis and treatment of this now common FMD are recommended. Etiological explanations fit coherently within a biopsychosocial model of pathology.

Keywords Functional tic-like behaviors · Functional movement disorders · Tourette syndrome · Tic disorders

Introduction

Between 2019 and 2022, there has been a sharp rise in the number of adolescents/young adults seeking urgent help for complex abnormal movements and vocalizations that resemble symptoms of primary tic disorders, which in many cases involved attendance to emergency departments or fast-track referrals to specialist clinics $[1, 2, 3^{\bullet\bullet}]$. In an effort to describe this new clinical presentation, an international

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group of clinicians and specialists agreed the term functional tic-like behaviors (FTLB) reflected that the behaviors tics and shared common features with other functional movement disorders (FMD) [3...]. Whereas FTLB may not be clearly differentiated from primary tics (e.g., those in Tourette syndrome) merely on the basis of phenomenology captured by visual inspection, they are recognized from a few phenomenological characteristics (predominant involvement of upper extremities or long, context-dependent verbal utterances, including coprolalic-like utterances), their sudden onset following by a rapid escalation of severity within 1 month, their later age at onset, and their different comorbidity profile, which includes also other functional neurologic symptoms [4••]. This striking overrepresentation of other functional symptoms corroborated the use of the "functional" attribute for these tic-like behaviors. An interesting link of this clinical phenomenon to both the initial phase of the SARS-CoV2 pandemic and an increased exposure to published videos representing tics and tic-like behaviors on social media channels has attracted the attention of neurologists, psychiatrists, pediatricians, and clinical psychologists. Due to unique patient characteristics apparent in the new cohort, we present an updated overview of FTLB

as these manifested in the last 3 years, their potential pathogenic mechanisms, and the treatment approaches adopted so far to decrease their impact on quality of life and related disability.

Phenomenology: Contrasting Functional Tic-Like Behaviors and Tourette Syndrome

As briefly presented in the "Introduction" section, while FTLB closely resemble popular media perceptions of Tourette syndrome (TS) phenomenology, there are distinctive differences in the demographic characteristics, onset, course, prominent behavioral symptoms, and potential risk factors that support a functional diagnosis. We will summarize these differences to aid this differential diagnosis and contextualize current and prospective avenues of research.

Demographic Differences

Sex ratios can vary by region but, on average, FTLB have a strong female predominance with 87% of patients reported as female across 7 case series [$3 \cdot \cdot , 4 \cdot \cdot , 5 \cdot \cdot , 6 \cdot , 7 \cdot \cdot , 8 \cdot , 9 \cdot , 10 \cdot \cdot]$. In contrast, TS has a male predominance with 76% of patients reported as male across 6 case series [$3 \cdot \cdot , 4 \cdot \cdot , 11 - 14$]. FTLB onset occurs typically later, with an average age of 14 years compared to 6 years in TS [$3 \cdot \cdot , 4 \cdot \cdot , 5 \cdot \cdot , 6 \cdot , 7 \cdot \cdot , 8 \cdot , 9 \cdot , 15 \cdot \cdot]$. Some patients with FTLB have a history of tics in earlier childhood and FTLB are not uncommon in patients with an established diagnosis of persistent tic disorder [$3 \cdot \cdot , 4 \cdot \cdot]$. Incident cases of FTLB during the SARS-COV-2 pandemic are younger than pre-pandemic FMD cohorts, suggesting this patient population may be unique [$4 \cdot \cdot , 16$].

Symptom Onset and Progression

TS typically begins as simple motor tics that follow a rostro-caudal distribution pattern, with tics initially primarily affecting the face, and evolving over time to include tics of the trunk and extremities [12, 13, 17]. Simple phonic tics usually emerge after motor tic onset [15••, 17]. TS patients experience typical fluctuations in tic severity [15••, 18••], usually with a peak in tic severity during pre-adolescence followed by a decrease in severity by late adolescence [12, 13, 17]. Tics diminish to milder presentations or remit by adulthood in many TS patients [12, 13].

Unlike TS, FTLB tend to have an explosive onset in adolescence or young adulthood as complex tic-like behaviors that often peak within hours or days [$6 \cdot$, $10 \cdot \bullet$]. Patients describe a worsening of symptoms during periods of high anxiety and stress, but without the classic waxing and waning pattern observed in TS [$7 \cdot \bullet$, $15 \cdot \bullet$]. Given their recent appearance, the long-term outcome of FTLB remains to be determined.

Phenomenology

Simple motor tics are rapid, repetitive, and purposeless movements that are usually limited to one muscle group [12, 13, 17]. Common simple motor tics observed in TS include eye blinking, eye-rolling, nose scrunching, and brief head jerks [12, 13]. Complex motor tics tend to be slower movements relative to simple motor tics, engage more muscle groups, and can involve movements that convey purpose [12, 13, 17]. Examples of complex motor tics include clapping, hand gestures, arm movements, jumping, or combinations of simple tics that occur simultaneously [13, 15••]. Simple phonic tics are brief meaningless sounds that commonly include throat clearing, sniffing, or noises like grunting [12, 13]. Complex phonic tics include words, phrases, echolalia (repeating after someone), palilalia (repeating after self), or speech atypicalities like yelling or speaking in novel accents [12, 19•].

Simple behaviors like blinking, facial grimacing, and shrugging occur in both TS and FTLB patients [3••, 13, 19•]. Patients with FTLB often also display rare manifestations of complex motor and phonic tics compared to TS patients. Copropraxia- (obscene gestures) and coprolalia-like (obscene words) behaviors, throwing objects, and behaviors potentially causing accidental self-harm are common in FTLB, whereas less than 20% of TS patients experience these types of symptoms [3••, 4••, 5••, 6•, 9•, 15••, 18••]. FTLB also disproportionately affect the limbs and trunk compared to TS [3••, 4••, 5••, 6•, 9•, 15••, 18••]. Tic attacks are lengthy paroxysms of non-suppressible motor and phonic tics lasting minutes to hours, reported in only 8% of TS patients, but in between 36 and 100% of FTLB patients across studies [4., 9., 20]. Pre-pandemic research has suggested that tic attacks in TS are psychologically maintained, with affected patients reporting higher levels of tic-focused attention, comorbid anxiety and panic disorders [20]. As we discuss below, anxiety and panic disorder are also common comorbidities of FTLB [4••, 9•, 15••, 18••]. FTLB can be socially reactive and directional with an extensive repertoire of variable behaviors that pose an additional facet of complexity than typically observed in TS $[4 \bullet \bullet, 6 \bullet, 8 \bullet, 9 \bullet, 18 \bullet \bullet]$.

Tics in TS are usually preceded by a premonitory urge, an uncomfortable sensory experience of tension that is temporarily relieved by performing a tic [12–14]. Data on premonitory urges associated with FTLB are somewhat conflicting, but studies suggest these sensations may be qualitatively and quantitatively different from those experienced in TS. Premonitory urges are reported by 90% of adolescents with TS and are often described as an itch, tingling, or pressure sensation [12–14]. In contrast, while some studies report lower incidence or even absence of premonitory urges before FTLB, others have reported that patients experience sensations that are physiologically similar to anxiety symptoms such as sweating, stomach discomfort, and palpitations $[4 \cdot \cdot, 8 \cdot, 9 \cdot, 21, 22]$. While there are clear differences between the disorders, TS and FTLB also share some common features. Both TS and FTLB are subject to suggestibility and distractibility $[4 \cdot \cdot,$ $7 \cdot \cdot, 23, 24 \cdot \cdot]$: both categories of behaviors can be triggered or aggravated by directing the focus of communication toward the behaviors and by intercurrent stressors, and both can be alleviated by re-orienting attention toward other attention-demanding tasks (particularly tics) or simply away from the behaviors (particularly FTLB).

Severity and impairment are other distinguishing factors. FTLB tend to have greater frequency, complexity, intensity, cause more interference with intended action, and are less suppressible than tics in TS [$4 \cdot \cdot \cdot , 9 \cdot , 15 \cdot \cdot$]. The level of impairment experienced by FTLB patients is concerningly high, with many patients reporting severe distress, tic-related absence from school and work, and social/relationship difficulties [$4 \cdot \cdot , 5 \cdot \cdot , 6 \cdot , 9 \cdot , 15 \cdot \cdot ,$ 18••]. Table 1 summarizes the demographic and phenomenological differences between FTLB and TS.

Risk Factors and Impact of the Pandemic

The most consistently reported predisposing factor associated with FTLB is a personal history of anxiety and mood disorders. Case series pre-dating the pandemic period showed a higher frequency of anxiety (27%) and depressive (45%) disorders compared to the general population and primary tic disorders [25-27]. This comorbidity was even more striking in patients who developed FTLB in the past 3 years, with anxiety disorders present in 50-75% of patients, and depression in 25–55% [4••, 7••, 15••]. In these case series, comorbidity was reported either based on the review of medical records, which were informed by routine diagnostic screening in outpatient clinics, or by ad hoc screening performed using validated rating scales. Although a higher frequency of suicidal behavior (encompassing both suicidal ideation-only and suicide attempts) has been reported [4...], more investigation is needed to clarify

Table 1 Clinical features of patients with functional tic-like behaviours and Tourette syndrome

Clinical features	Functional tic-like behaviours	Tourette syndrome
Demographics and progression		
Age of onset	10 to 25 years Mean = 14.25	5 to 7 years <i>Mean</i> =6.12
Sex-ratio	Female predominance	Male predominance
Symptom onset	Explosive; complex motor and phonic tics. Tics can affect any part of the body	Gradual; simple motor tics followed by simple phonic tics Rostro-caudal progression
Waxing/Waning progression	Absent	Present
Peak Severity	Within hours or days of onset	~5 years after onset
Tic Features	-	
Motor tics	Typically affect limbs and trunk Common tics include copropraxia, throwing objects, self-injuring behaviors. Tics are often directional and socially reactive	Typically affect the face and upper body Common tics include eye blinking, eye-rolling, head jerks, facial grimace, and shrugs. Can later progress to complex tics
Phonic tics	Complex words and phrases: Common tics include coprolalia, offensive remarks, phrases, bird and animal noises	Simple phonic tics Common tics include throat clearing, sniffing, grunting Complex tics can develop later Coprolalia rare
Tic-attacks	Common	Rare
Premonitory urge	Less common; anxiety like symptoms	Common; tingling, itching, pressure sensation
Suppressibility	Less adept	Yes
Distractibility	Yes	Yes
Impairment	High	Variable (Lower on average)
Severity	High	Variable (Lower on average)
Risk Factors		
Social media	Yes	Unknown
Psychiatric Comorbidities	Anxiety, Depression	ADHD, OCD, ASD, depression,

Clinical features from patients with functional tic-like behaviors that emerged since the onset of COVID-19 pandemic

whether self-harming behaviors are the expression of acting out on true suicidal plans or rather represent parasuicidal behaviors. Among concurrent neurodevelopmental disorders, one clinical series from London, England, reported a remarkably high frequency of clinically diagnosed (50% of 14 patients) and clinically suspected (57% of 34 patients) autism spectrum disorder, significantly higher than in primary tic disorders [7••]. Autistic comorbidity might have been under-detected in other clinical series and requires further investigation [4...]. Comorbid attention deficit/hyperactivity disorder (ADHD) or obsessive-compulsive disorder (OCD) were reported in 14% and 23%, respectively, of 22 patients from a case series from Sydney [4.., 5..]. Only about 15% of patients with FTLB seeking medical attention between 2019 and 2022 had a prior history of tic disorder $[4 \bullet \bullet, 7 \bullet \bullet, 15 \bullet \bullet]$. The frequency of a family history of tic disorder varied across case series from different geographical regions, with the highest estimate reported at around 30% [4••, 7••]. For this reason, a disease modeling mechanism based on personal or family-related experience of tics may be relevant only for a minority of these patients.

Additional predisposing factors could be represented by adverse childhood experiences or psychological trauma experienced during childhood or adolescence. However, the prevalence of these potential predisposing factors and their role in the pathogenesis of FTLB needs to be explored. Similarly, the actual frequency of pre-existing post-traumatic stress disorder in these patients warrants specific investigation.

Until the early 2000s, functional movement disorders (FMD) have consistently been reported at a frequency lower than 5% among all movement disorders diagnosed in both adult and pediatric movement disorders specialist clinics [28–30]. These disorders have always been in proportion more frequent in the emergency setting. An exemplary study from Italy that evaluated prospectively movement disorders diagnoses in emergency departments between 2013 and 2017 reported FMD as the second most common cause of referral to an emergency service (almost 20%) [31].

During the past 6 years, frequency estimates for FMD demonstrated a steady rise within tertiary movement disorders outpatient clinics, approximately doubling their prevalence. An interesting cross-sectional study from movement disorders specialists at Baylor College of Medicine, Houston, TX [32] compared the frequency of incident FMD in this large sub-specialty clinic between the March-to-October periods of 2018 and 2020. This frequency rose from about 5% to about 8%, a 60% increase of the incidence of FMD in both children (90%) and adults (51%). In this population, the most common FMD were tremor (53%), dystonia (31%), myoclonus (18%), a frequency distribution like the one observed in pre-pandemic times. Other typically less common FMD (tic- and stereotypy-like) were estimated at

9%. Twenty percent of patients manifested multiple FMD. A further rise of the frequency of FMD has been reported also among acute movement disorders clinics accessing emergency departments. At the Salpetriere Hospital in Paris, waves of hospitalization for FMD followed the SARS-CoV2 epidemic epochs and ICU admissions [33]. However, FTLB and tic attacks were not observed in this specific study, probably because it focused on adult emergency services.

The origin of this rise in incident cases of FTLB during the SARS-CoV2 pandemic is probably multifactorial. First, it needs to be acknowledged that the past decade has witnessed a background increase in awareness and ability to diagnose FMD in general, supported by a reduced latency to diagnose that has reached a median duration of 2 years [34]. Second, the SARS-CoV-2 pandemic has driven an escalation of psychosocial stressors and anxiety in youth and young adults, fostered by the repeated period of social deprivation, concerns about the risk of infection, and academic, financial, and professional repercussions of the economic impact of the pandemic on individuals and families [35-37]. Third and probably most important, the use of social media channels among adolescents and young adults has increased significantly during the pandemic, potentially fueling a further rise in anxiety and emotional dysregulation [9•, 18••, 37]. However, besides this "non-tic-specific" effect of increased social media use on emotional control, the greater time spent on these communication channels has increased the chances of exposure to a "tic-specific" content, characterized by videos posted by web personalities exposing their tics or tic-like behaviors as core characteristics of their "public persona" [38, 39]. Several clinical observational studies have examined this social media influence by reporting both a phenomenological similarity between patients' FTLB and the tics exhibited in these videos $[40 \bullet \bullet]$ and the high interrater agreement among pediatric neurologists on diagnosing the behaviors in these videos as functional [39]. It is therefore plausible that FTLB in patients have been driven by a modeling mechanism, based on the concept of nonconscious mimicry modulated by the desire to affiliate with specific social partners [41]. Reinforcement by peers, family, and even health professionals may even promote the persistence of these behaviors, catalyzed by the diffusion of specific stimuli through social media.

Theoretical Explanations

Social Media Induced Mass Psychogenic Illness

Müller-Vahl et al. [42••] theorize that a mass psychogenic illness spread through social media and triggered by pandemic anxiety or climate change anxiety may be responsible for the sudden surge in patients with FTLB. Through their own clinical observations and communication with other centers, they were able to confirm that an increase in FTLB patients with reported exposure to tic- and TS-related social media content was occurring globally [42••]. Notably, they report that these patients presented with strikingly similar behaviors and demographic characteristics paralleled by social media personalities popular in each region [42••].

While exposure to tic-related social media content appears to be an important contributing factor to FTLB for some patients, there are several observations that suggest the etiology is more complex than can be explained by a mass social media-induced psychogenic illness alone. The SARS-CoV-2 pandemic has been a universal stressor, affecting citizens in nearly every country on the planet [43]. At the same time, there has been a staggering increase in tic and tic-like behavior content on social media that has been viewed by tens of millions of individuals [40••]. If FTLB were spread by social media and triggered by anxiety, we might expect a surge in FTLB cases, well beyond the sizeable increase seen in neurology clinics today. This suggests that patients presenting with FTLB possess special characteristics that are not shared by most people viewing tic content.

From our own clinical observations, as indicated above, we have noted many patients share common factors that can include complex psychological symptom profiles, selfreported adverse life experiences, and family history of neurological and psychiatric disorders. It is also important to note that some patients do not report exposure to tic-related social media content and that many patients with FTLB have a prior history of organic tics.

Biological Explanations

Prior to the pandemic, FTLB were a relatively uncommon presentation of FMD resulting in few studies investigating biological differences in FTLB, specifically. There are however several biological studies and reviews that used mixed FMD presentations that included individuals with FTLB or functional "jerk" movements of the limbs that may indicate possible biological etiologies.

The bereitschaftspotential (BP) is a slow potential that is detected immediately prior to voluntary movement on EEG [44]. Van Der Salm et al. compared BP in patients with FMD, TS, myoclonus, and healthy controls [45]. The study asked participants to perform voluntary wrist movements in addition to recording data when muscle jerks occurred spontaneously. Healthy controls were also asked to feign jerking movements for comparison. BPs were completely absent in 59% of voluntary wrist movement trials in FMD patients, whereas myoclonus and healthy control participants displayed BPs 100% of the time before voluntary movements. TS patients displayed BP 100% of the time before voluntary movements of their dominant hand [45]. BP was present more often before spontaneous jerks in FMD patients than TS and myoclonus patients, and it was present 100% of time before feigned jerk movements in healthy controls [45]. These results are significant as they indicate neurological anomalies preceding volitional and spontaneous movements in FMDs that are distinct from feigning.

Behavioral data assessing drift rate, a measure of the accuracy and response rate of a participant's ability to detect if they have received either one or two stimuli, indicated impaired sensory processing and decision-making in FMD patients compared to healthy controls [46]. FMD patients asked to complete a decision task while viewing positively and negatively valenced stimuli exhibited increased limbic activity in all conditions whereas healthy controls only experienced enhanced activity when viewing negative stimuli [47]. These findings were proposed to indicate FMD patients experience higher baseline arousal which is consistent with studies finding FMD patients have elevated levels of salivary cortisol and lower basal vagal tone [47, 48].

Imaging studies have demonstrated aberrant connectivity and structural abnormalities in brain areas thought to be responsible for motor initiation and control, emotional and self-awareness, interoceptive processing, motor intention awareness, self-agency perception, and social processing of behavior in patients with FMD [47, 48, 49••, 50, 51••, 52, 53]. Altered functional connectivity between the limbic and motor regions in FMD patients appears consistent with the increase in tic-like behavior severity and frequency during times of stress reported by FTLB patients.

While these biological data offer some compelling potential neurological explanations for FTLB pathology, it is important to acknowledge potential limitations that may hinder generalizability to the current FTLB patient population. Studies that investigated mixed presentations of FMD often included patients experiencing positive motor symptoms (e.g., tics, jerks, tremors) and negative motor symptoms (e.g., paresis, paralysis, gait disturbance). We would presume that neurological activation patterns observed in positive and negative symptom presentations would be different, introducing uncertainty to how each presentation contributed to the above results. It is also important to note that FTLB patients represented only a small portion of the study samples. Additionally, the FTLB patients that emerged since the onset of the pandemic are demographically different, with a younger age of onset and an extreme female predominance compared to previous $[10 \bullet, 22, 54 \bullet]$. These demographic differences may suggest that these patient populations are distinct with different developmental and biological etiologies.

Future biological studies of the new FTLB cohort are needed. As many FTLB patients report vocalizations to be an extremely debilitating symptom, new investigations into brain regions controlling speech and language will be a new avenue of research. Collecting biological data from this patient population is not without its challenges. The frequency and severity of tic-like behavior movements experienced by many of the patients that emerged since the onset of the SARS-CoV-2 pandemic may preclude imaging data collection.

Diathesis-Stress Model

Pringsheim et al. have postulated that predisposing traits combined with environmental precipitating factors may have led to FTLB behaviors that are further reinforced in an adapted version of a diathesis-stress model. Predisposing factors include, but are not limited to, genetic and epigenetic factors, depressive and anxious traits and states, and early life events [3••]. Precipitating factors may include stressors related to the pandemic and increased social media exposure to tic-like behaviors. Behavioral reinforcers may include social rewards (potentially from social media and peer attention) and increased self-attention to behaviors [3••].

This model appears to better reflect the complexity in presentation and history that we have observed in our FTLB patients in clinic. As the SARS CoV-2 pandemic evolves through time, the stressors and our understanding of the precipitating factors are likely to change. With local health restrictions eased, we may begin to see a clearer picture of how predispositions and pandemic related precipitating factors interact.

Biopsychosocial Model

The merits of the mass sociogenic illness, biological, and diathesis-stress theoretical models of FTLB are cogent and evidence-supported. These models could co-exist harmoniously as components within a more general biopsychosocial model of pathology. There is increasing evidence that the relationships between the biological, social, and psychological factors in FMD are highly integrated and reciprocal [5••, 55]. An amalgamated model that incorporates the aforementioned theoretical explanations is illustrated in Fig. 1. This model provides a framework that can illustrate how different factors interact and may illustrate how interventions and treatments can be expected to influence the cycle of pathology. We may expect that therapies that target the factors that exist in the overlapping margins of biological, social, and psychological factors to be particularly effective. Preliminary data has suggested that CBT therapy for anxiety and depression, a therapy that focus on social cognition, reducing avoidance behaviors and coping skills has led to improved outcomes for FTLB patients [56••].

Treatment and Prognosis

To date, the treatment approach to FTLB starts with a clear and articulated diagnostic debriefing, in line with a therapeutic principle applicable to the whole spectrum of functional movement disorders [57]. Salient points of diagnostic debriefing are the validation of the core symptoms as genuine (i.e., not voluntarily performed or simulated), and the acknowledgement that FTLB should not be considered a "rare" clinical presentation. It is also important to name them correctly, using the attribute of "functional" and explaining its meaning, alongside providing useful educational resources (e.g., the www.neurosymptoms.org or https://fndhope.org websites). Naming and describing the phenomenology of FTLB may, however, not be sufficient. Providing a clear and concise mechanistic explanation, with reference to the existing knowledge to date, is also crucial to allow patients to understand the potential predisposing and precipitating factors, thus enhancing acceptance of the diagnosis, paving the way to compliance and responsiveness to active treatment, and promoting a good prognosis [58]. In addition, this type of psychoeducation would identify more comprehensively relevant treatment targets linked to the pathogenesis of FTLB, including stressors and comorbid psychiatric conditions.

Before introducing appropriate interventions, patients and families should be informed of the poor efficacy of established tic-suppressing medications, first and foremost antipsychotic drugs, in treating the vast majority of patients with FTLB. On the other hand, behavioral interventions should be prioritized, even though more information from different observations worldwide are warranted to clarify the most rewarding approaches. One of these approaches, reported from our Calgary experience [3••, 10••, 56••], is a specific adaptation of the Comprehensive Behavioral Intervention for Tics (CBIT) which focuses on function-based strategies to target precipitating and perpetuating factors. This approach entails mitigating potential triggers, especially social media exposures, initiating stress management interventions related to identifiable psychosocial stressors, minimizing social reactions or attention to symptom expression from families and peers, and managing avoidance and accommodation, including aiding patients and families to reinstate a "healthy" and pleasurable lifestyle. Importantly, the habit reversal therapy component of the CBIT-considered the core of this complex behavioral intervention when this is applied to tics-appears difficult to apply to this group of patients, primarily for the very high frequency and variability of tic-like behaviors, which makes the identification and implementation of competing motor responses highly challenging.

A similar experience has been presented by the Sydney group [4••], based on multidisciplinary targeting of

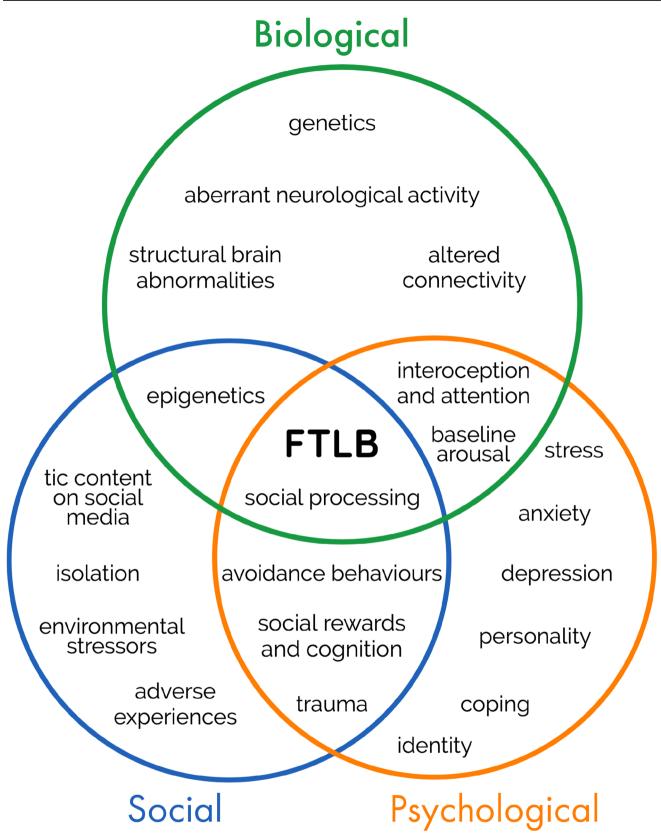


Fig. 1 A biopsychosocial model of functional tic-like behaviors (FTLB) pathology. This model incorporates the etiological data identified across numerous studies of FTLB [3••, 4••, 5••, 6•, 7••, 8•, 9•, 13, 14, 23, 41, 44–48, 49••, 51••, 54•, 55, 56••]. The biologi-

cal, social, and psychological components are highly integrated and reciprocal. Each element represents a potential, but not mandatory, factor contributing to the FTLB pathology accounting for the heterogeneity of patient characteristics and experiences biological, psychological, and social components, using a highly individualized approach. Their approach considers the re-introduction of healthy sleep, exercise and dietary routines, the recognition of triggers, stressors and signs of neurophysiological activation, and the intervention on thoughts, feelings, and maladaptive coping strategies. Pharmacotherapy is also included in this therapeutic plan, albeit only as an adjunct component, which may address, as needed, sleep quality (e.g., using melatonin), hyperarousal (e.g., using alpha-2-adrenergic agonists or beta-blockers), and anxiety/depression symptoms (e.g., antidepressants, or even antipsychotics if augmentation therapy is required in the case of self-injurious or aggressive behaviors).

In Calgary, our prospective 6-month follow-up observation showed that most patients with FTLB improve with time, with adolescents exhibiting a slightly more favorable evolution of symptoms [55]. Most of these patients were treated with cognitive-behavioral therapy for anxiety and depressive symptoms and/or antidepressants (in particular, serotonin selective reuptake inhibitors), confirming the value of treating actively predisposing psychiatric comorbidities in this population.

Next Steps in Research

The rapid escalation of incident cases of FTLB during the first year of the SARS-CoV2 pandemic has posed important challenges to patients, families, emergency, primary and secondary care clinicians, as well as to clinical researchers. These challenges can be grouped in three main areas: diagnosis, basic mechanisms, and treatment/prevention.

Despite the several demographic, phenomenological and course-related differences between FTLB and tics related to primary tic disorders, the lack of diagnostic criteria and classifications aiding this differentiation may have caused diagnostic delay and unnecessary treatment trials, including the use of antipsychotic medications. A first, important and urgent objective for current and future research is to achieve consensus on a set of diagnostic criteria for FTLB, prompted also by the importance of early diagnosis in the outcome of functional symptoms.

Functional motor symptoms are a profoundly diverse spectrum, which likely encompasses both shared and symptom-specific pathogenic mechanisms. A better understanding of the predisposing factors within psychiatric comorbidities and environmental exposures is necessary to clarify whether predisposing and precipitating factors in FTLB represent a consistent and recurring combination, or rather are differently represented in different subgroups of patients. Inevitably, the assessment of these factors appears crucial in guiding personalized treatment.

Clinicians have familiarized with this complex clinical presentation over the course of the last 3 years, but a standardized approach to personalized therapy is still under definition. Longitudinal studies synthesizing the experience of specialists around the world would highlight the most promising therapeutic approaches and evaluate their dissemination and implementation. Psychoeducation and multidisciplinary active interventions are emerging as key components of efficient management, and their operationalization is urgently needed. Avoidance of unnecessary, and potentially detrimental, therapies, e.g., antipsychotics prescribed as "ticsuppressing" interventions, is supported by recent observational data. Finally, the interaction between social factors (e.g., social media use, exposure to specific content related to tics and tic-like behaviors, previous adverse experiences) and adaptive and coping abilities needs to be investigated systematically to guide behavioral interventions and ideally prevent diffusion or recurrence of this often incapacitating clinical syndrome.

Declarations

Conflict of Interest The authors declare that they have no conflict of interest.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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