University of Southampton

Faculty of Environmental and Life Sciences

School of Psychology

Increasing Understanding of and Exploring Interoceptive Processes in Functional Tic-Like Behaviours in Comparison to Chronic Tic Disorders

Volume 1 of 1

by

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Thesis for the degree of Doctorate in Clinical Psychology

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Abstract

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Chapter 1 presents a systematic review exploring the characteristics of functional tic-like behaviours and how they compare to established features of chronic tic disorders. Overall, 33 studies were included in the review and were assessed for quality of research methods and reporting. Evidence from the studies highlighted differences between functional tic-like behaviours and chronic tic disorders such as a higher age of onset and female predominance. Studies revealed mixed findings for features such as the severity and types of tics that occur in functional tic-like behaviours in comparison to chronic tic disorders. The majority of studies included small sample sizes and were observational. It is proposed that future research should conduct some experimental studies to investigate differences between chronic tic disorders and functional tic-like behaviours and recruit larger samples.

Chapter 2 presents a quantitative study investigating differences in interoceptive processes in young people with chronic tic disorders, young people with functional tic-like behaviours, and young people with neither diagnosis nor history of tics. Relationships between interoceptive processes, attentional control, comorbid psychiatric symptoms, tic-specific features, and quality of life were also explored. The study recruited 53 participants (23 with chronic tic disorders, 7 with functional tic-like behaviours, and 23 controls). Participants completed self-report measures and two tasks measuring different domains of interoceptive accuracy. Results revealed interoceptive accuracy and interoceptive insight did not differ between the three groups, but young people with functional tic-like behaviours had reduced interoceptive beliefs compared to young people with chronic tic disorders and controls. Interoceptive beliefs and comorbid anxiety and depression predicted quality of life in young people with chronic tic disorders and young people with functional tic-like behaviours. The study has some limitations such as a small sample size and interoceptive measures may lack construct validity. Future research should seek to recruit larger samples and validate interoception measures.

Chapter 3 presents a more detailed overview of the clinical features of chronic tic disorders. The different diagnoses are reported including the types of tics associated with chronic tic disorders and current psychological interventions used to treat tics. This chapter also discusses difficulties in recruiting young people with functional tic-like behaviours to this quantitative study. Possible explanations for the small sample size are discussed and suggestions for future research are made.

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Research Thesis: Declaration of Authorship

Print name: Kayleigh Tuttle-Cull

Title of thesis: Increasing Understanding of and Exploring Interoceptive Processes in Functional Tic-Like Behaviours in Comparison to Chronic Tic Disorders

I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. None of this work has been published before submission

Signature: ..... Date: 17/09/2025

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And finally, a message to me. This thesis is proof that I can do it. I am good enough, I am smart enough, and I am resilient.

Definitions and Abbreviations

ACS-C..... Attentional Control Scale for Children

ANOVA ..... Analysis of Variance

ADHD ..... Attention Deficit Hyperactivity Disorder

ASD ..... Autism Spectrum Disorder

CGAS..... Children's Global Assessment Scale

CTDs..... Chronic Tic Disorders

CBT..... Cognitive Behavioural Therapy

CBIT..... Comprehensive Behavioural Intervention for Tics

CI ..... Confidence Intervals

DSM-V ..... Diagnostic Statistical Manual of Mental Disorders, Fifth Edition

ESSTS ..... European Society for the Study of Tourette Syndrome

FMD..... Functional Movement Disorder

FND ..... Functional Neurological Disorder

FNS ..... Functional Neurological Symptoms

FTLB ..... Functional Tic-Like Behaviours

FT ..... Functional Tics

FTD..... Functional Tic Disorder

GAD-7..... Generalised Anxiety Disorder-7

HCT ..... Heartbeat Counting Task

I-CBiT..... Integrated-Cognitive Behavioural Intervention for Functional Tics

IQ ..... Intelligence Quotient

IAS-C ..... Interoceptive Accuracy Scale for Children

MSMI-FTLB..... Mass Social Media-Induced Illness Functional Tic-Like Behaviours

MUS..... Medically Unexplained Symptoms

MTD ..... Mixed Tic Disorder

MOVES ..... Motor Tic, Obsessions and Compulsions, Vocal Tic Evaluation Survey

NHIS GTS ..... National Hospital Interview Schedule for the Assessment of Gilles de la Tourette Syndrome

NES ..... Non-Epileptic Seizures

NOSIB..... Non-obscene Socially Inappropriate Behaviour

OCB..... Obsessive Compulsive Behaviours

OCD ..... Obsessive Compulsive Disorder

PHQ-9..... Patient Health Questionnaire-9

Peds-QL..... Paediatric Quality of Life Inventory

PRISMA..... Preferred Reporting Items for Systematic Reviews and Meta-Analysis

PUTS..... Premonitory Urge for Tics Scale

PTD..... Primary Tic Disorder

PMD..... Psychogenic Movement Disorder

PMRT ..... Psychogenic Movements Resembling Tics

PT ..... Psychogenic Tics

RCADS..... Revised Child Anxiety and Depression Scale

TLB-SM ..... Tic-like Behaviours following Social Media Consumption

TTD ..... Transient Tic Disorder

TS ..... Tourette Syndrome

SSRIs ..... Selective Serotonin Reuptake Inhibitors

SNAP-IV ..... Swanson, Nolan, and Pelham Version IV Rating Scale

WASI-II..... Weschler Abbreviated Scale of Intelligence, Second Edition

YGTSS..... Yale Global Tic Severity Scale

Chapter 1 Systematic Review

Current Understanding of the Clinical Characteristics of Functional Tic-Like Behaviours: A Systematic Review

Journal Specification: The following chapter has been prepared for submission to the Journal of Neurology. The guidelines for authors are shown in Appendix A. However, font style and size has not been followed and instead the standard university template font (Aptos) has been selected for ease of readability for the examiner.

Word Count: 9298

(excluding abstract, keywords, figures, tables, captions and references)

1.1 Abstract

Background: The prevalence of functional tic-like behaviours has rapidly increased in recent years. Diagnostic criteria have been proposed. There is ongoing exploration around the phenomenology and how this disorder may differ from chronic tic disorders. This review aims to systematically examine the literature on functional tic-like behaviours and summarise the common features of this disorder.

Methods: The preferred reporting items for systemic review and meta-analysis guidelines were followed. The CINAHL Ultimate, MEDLINE, PsycInfo, and Web of Science databases were searched on 18 September 2024. Search terms included: Tourette, tic disorder, tics, functional tic, functional movement, sociogenic, psychogenic or conversion disorder. Reports were screened using a pre-established inclusion/exclusion criteria. Each study was quality assessed using the QualSyst. Findings were summarised via narrative synthesis.

Results: *k*=1007 reports were identified and screened. Overall, *k*=33 studies were included. Sample sizes of *k*=33 studies ranged from *n*=8 to *n*=294 participants. Mean age of functional tic-like behaviour onset ranged from 13 to 31 years old. 83.5% of participants were female while the typical ratio in tic disorders is 3-4:1 male to female. Participants had a varied repertoire of tics. Presence of premonitory urges, suppressibility, and severity was mixed when compared to chronic tic disorders.

Conclusions: Findings suggest age of onset is a good diagnostic indicator of functional tic-like behaviours and additional phenomenological features should be considered in conjunction to aid diagnosis. Results lack generalisability due to small sample sizes. Future research should explore differences between functional tic-like behaviours and chronic tic disorders in larger cohorts.

Keywords: functional tic-like behaviours, functional tics, tic disorder, phenomenology

1.2 Introduction

Functional movement disorders (FMD) are a subtype of functional neurological disorder (FND) and refer to altered motor function that is not explained by neurological conditions and lead to similar symptoms such as limb weakness, involuntary movements, or gait disturbances [1]. It is proposed that "functional" disorders can be understood in a similar manner as Freud's term "hysteria" to describe physical manifestations of psychological trauma which was later referred to as conversion disorder [2, 3]. Despite this terminology remaining in the Diagnostic Statistical Manual of Mental Disorders, 5th Edition (DSM-V), the requirement of a psychological stressor has been removed, highlighting the recent move towards understanding FND/FMD as an interplay of both neurobiological and psychosocial factors [4, 5]. Moreover, the DSM-V now relies upon a positive symptom criteria to diagnose FMD which involves conducting neurological examinations to identify inconsistencies in patterns of abnormal movements that would not be observed in neurological diseases [1, 4]. Features such as "sudden onset", "suggestibility", "symptom relief when distracted", and "increased severity with focused attention" have been proposed to be incongruent with established neurological conditions [1, 4].

In contrast, chronic tic disorders (CTD) including Tourette syndrome are neurological disorders which are diagnosed by the presence of motor and/or vocal tics lasting more than 12 months [6]. Tics are brief, non-rhythmic, sudden, and repetitive, and are known to wax and wane and regularly occur in bouts [6, 7]. Moreover, tics typically develop in early childhood with onset occurring between the ages of 3 to 8 [8, 9]. Males are more likely to be diagnosed with tics with most studies reporting a male to female ratio of 3-4:1 [8, 10]. Tics are highly suggestible and can be suppressed for short periods of time [11, 12]. In addition, tics are often preceded by a premonitory urge or uncomfortable pressure which dissipates following tic expression [13, 14]. Tics can be treated using pharmacological or psychological interventions [9] and comorbidities such as obsessive compulsive disorder (OCD) and attention deficit hyperactivity disorder (ADHD) are frequently observed in patients with CTDs [15].

Recently, researchers have become interested in exploring and furthering the understanding of functional tic-like behaviours (FTLB). These are classed as a subcategory of FMD and refer to an acute and sudden onset of tics, which are considered atypical of presentations of tics observed in CTDs [16]. Despite cases of FTLB being documented as early as the 19th century, they were primarily recorded as single case studies and appeared relatively rare in comparison to CTDs [16]. However, this may be due to the varying terminology used to describe FTLB over the recent years including psychogenic tics, pseudo tics, and functional tics,

which may have made assessing the prevalence difficult [17]. More recently, the terms sociogenic illness, mass social media-induced illness functional tic like behaviours (MSMI-FTLB), tic attacks, and TikTok tics have been used to describe a particular group of patients that develop FTLB after viewing social media content portraying tics [18, 19]. Despite the potentially different cases, these terms are theorised to be part of a similar functional phenomenon [18]. Throughout the 2010s, the number of patients with FTLB began to increase and multiple case series focused on describing the clinical characteristics of these patients [16]. Moreover, during the COVID-19 pandemic there was a sudden surge in the number of patients presenting with FTLB at emergency departments and specialist movement disorder clinics, with some services reporting almost double the number of referrals compared to pre-pandemic rates [20]. Researchers have suggested this increase may be due to the social restrictions imposed, leading to increased anxiety and depression [21]. Researchers suggest patients with FTLB may have higher rates of anxiety but lack awareness or the ability to manage their emotions resulting in increased susceptibility to somatise [21, 22]. This coupled with a possible pre-existing vulnerability for developing tics may lead to developing FTLB in response to high periods of stress [21, 22].

However, diagnosing FTLB proves challenging for clinicians as they can present as similar to CTDs, making it hard to differentiate between the two [21]. Existing criteria used to diagnose FMD such as suggestibility, distractibility, and varying frequency are redundant in identifying FTLB as these features are common amongst tics seen in CTDs [17]. Thus, it is evident a new criterion is required to diagnose FTLB and differentiate them from CTDs [21]. In 2023, experts in tics developed working diagnostic criteria for FTLB based on common features observed in hundreds of patients across multiple international sites [16]. To achieve a clinically definite diagnosis of FTLB, they proposed a rapid onset of tics at the age of 12 or older, and a particular phenomenology including: multiple complex movements predominantly of the arms and hands, multiple complex vocalisations (words and statements), tics varying in frequency, consistency, and intensity, new tics developing every few days and possibly increasing during examination, and mimicking popular social and cultural influences [16]. For a clinically probable diagnosis of FTLB, at least two of the major criteria above must be present and one minor criterion, which is either the presence of comorbid psychiatric disorders such as anxiety and depression, or additional functional neurological symptoms (FNS) or somatic disorders [16].

Despite these novel diagnostic criteria, clinicians report a lack of confidence in accurately diagnosing FTLB which can result in patients being misdiagnosed with CTDs [23, 24]. This can be problematic as recommended treatments for CTDs have been found to be ineffective in treating FTLB and, thus, a correct diagnosis is required to seek appropriate support and aid symptom reduction [21]. Diagnosis is further complicated by the functional overlay seen in FTLB and

CTDs, particularly as a number of patients with FTLB report a history of tics, and comorbidity between CTDs and FTLB is possible [24, 25]. Moreover, Anderson et al. [26] suggests studies diagnose patients with FTLB based on specific characteristics and then analyse these same characteristics and base further diagnoses upon these features. This may mean characteristics described in studies may not be valid features of FTLB and instead reflect biases within the diagnostic process [26]. Thus, the possible influence of circular reasoning further highlights the challenges in conceptualising FTLB and differentiating them from CTDs [26].

Therefore, the aim of this review is to systematically search the literature on FTLB, including associated terminology, in the hopes of providing a summary of the clinical features of FTLB in relation to the current diagnostic criteria. We aim to explore and summarise the types of tics present in FTLB including their expression, severity, intensity, and degree of impairment. We also aim to review sex and age differences in people presenting with FTLB alongside common comorbid psychiatric disorders and responses to treatment. This will hopefully provide useful insights into FTLB and allow researchers to compare the clinical features against the symptomology of CTDs, potentially having significant implications on interventions.

1.3 Methods

This systematic review followed the preferred reporting items for systematic reviews and meta-analysis (PRISMA) guidelines [27] and the protocol was registered on PROSPERO (CRD42024591263) on 17 September 2024. The protocol was updated twice following registration. This was to amend search terms and inclusion criteria. The quality assessment tool was also changed from the critical appraisal skills programme to the QualSyst [28] to suit the varying study designs included in the systematic review.

1.3.1 Search Strategy

The CINAHL Ultimate, MEDLINE, PsycInfo, and Web of Science databases were searched on 18 October 2024. An experienced librarian was consulted when developing search terms to help minimise the chances of relevant studies being missed during searches. Considering several different terminologies are used to describe FTLB within the literature, broad search terms were developed in the hopes of capturing all possible pseudonyms representing this phenomenon. Table 1 presents the search strategy used for each database. All databases were searched from the date of inception, with the earliest study retrieved published in 1946. No grey literature searches or citation chaining were conducted. The search was updated on 08 April 2025.

1.3.2 Eligibility Criteria

Since the systematic review aims were focused on identifying clinical features of FTLB, broad inclusion criteria were developed. Studies that investigated individuals with a confirmed diagnosis of FTLB were included in the review. Experimental and observational studies that included descriptive statistics or statistical analysis on individuals with FTLB were included. Studies that included a comparison group of participants with CTDs were also included. There were no restrictions on the age of participants or the date studies were published. This was to maximise the chances of relevant studies being included in the review. The search strategy also had no restrictions in the language reports were written in as the researchers aimed to translate any identified non-English reports.

Table 1 Database search strategies

Database (Host)	**Search Strategy**
CINAHL Ultimate (EBSCO)	(Tourette* or "tic disorder" or tics) and (functional N3 tic or functional N2 movement or sociogenic or psychogenic or "conversion disorder")
MEDLINE (EBSCO)	(Tourette* or "tic disorder" or tics) and (functional N3 tic or functional N2 movement or sociogenic or psychogenic or "conversion disorder")
PsycInfo (EBSCO)	(Tourette* or "tic disorder" or tics) and (functional N3 tic or functional N2 movement or sociogenic or psychogenic or "conversion disorder")
Web Of Science (Clarivate)	((ALL = (tourette*)) or ALL = ("tic disorder")) or ALL = (tics) and ((((TS = (functional NEAR/3 tic)) or TS = (functional NEAR/2 movement)) or ALL = (psychogenic)) or ALL = (sociogenic)) or ALL = ("conversion disorder")

1.3.3 Exclusion Criteria

Studies that did not investigate FTLB but researched CTDs, including those that referred to CTDs as “psychogenic tics” were excluded. Studies that described a singular psychogenic tic (e.g. psychogenic tic cough) were excluded as they do not fit the requirements of multiple tics seen in FTLB. Similarly, studies that investigated FMD more generally and provided no sub-group analysis on FTLB were excluded. Case series that provided no descriptive statistics and only described individual cases were excluded. Single case reports were also excluded.

Qualitative studies, position papers, systematic reviews, commentary papers, grey literature, and conference abstracts were also excluded. Studies that were not published in English were excluded only if they could not be translated.

1.3.4 Study Selection

Once database searches were complete, records were exported to EndNote 21 and duplicates were removed. The first author screened the titles and abstracts of each record and those that were irrelevant or clearly did not meet the inclusion criteria were excluded. A second rater independently screened 10% of randomly selected titles and abstracts and inter-rater reliability was 'fair' using Cohen's Kappa coefficient ($\kappa = .29$). Full texts were retrieved for all remaining reports and were screened by the first author using the inclusion/exclusion criteria to determine eligibility. The second rater independently screened 100% of full texts to check studies were correctly included and excluded and inter-rater reliability was 'substantial' ($\kappa = .75$) for this step. Disagreements were resolved via discussion or by consulting a third independent rater.

Reports that appeared identical to each other (e.g. same number of participants and demographics) were double-checked to identify whether they belonged to the same study (same patient sample) to prevent double-counting participants [29]. Reports of the same study were combined and the report that provided the most information on study design and participant selection was referred to as the "main" study, in which data extraction and quality assessment was undertaken. When reporting study findings, the "main" study and relevant report were both referenced to improve transparency.

1.3.5 Quality Assessing

All studies that met the inclusion criteria were quality assessed by the first author. The QualSyst tool (Online Resource 1) was selected as it was designed to assess the quality of diverse studies [28]. The QualSyst consists of 14 items assessing the appropriateness of study design and objectives, recruitment, sample size and description, use and definition of appropriate outcome measures, possible randomisation and blinding, attempts to control confounds, appropriate reporting of results including variance estimates, and conclusions. Each item received a score (0 = no, 1 = partial, 2 = yes) to indicate the degree to which the study met the specific criteria; some items could be marked as "n/a" if they were not applicable to the study design, and these were then subtracted from the total possible sum. An overall summary score was calculated by adding the scores of each item and dividing them by the total possible sum, producing a quality score between 0 and 1. Studies with a quality score below 0.55 were

considered low quality, scores between 0.55 and 0.75 were acceptable (medium), and 0.75 and above were considered high quality [28]. No studies were excluded based on the quality assessment score.

1.3.6 Data Extraction

The first author developed a data extraction form to help extract the following information from each study: (1) author, year, and country of publication, (2) study design (cross-sectional, longitudinal, experimental, case series), comparator and length of follow up (if applicable), (3) sample size and population, (4) age, sex, gender identity, ethnicity, and comorbid diagnoses of participants, (5) terminology used to describe FTLB, diagnostic criteria used, professional providing diagnosis, and when sample diagnosed, (6) age of FTLB onset and trajectory (acute vs gradual), (7) description of tics (types, triggers, severity, frequency, and impairment) and associated symptoms (premonitory urge and suppressibility), and (8) interventions used to treat FTLB (pharmacotherapy vs psychotherapy) and response to treatment. Data was extracted twice on two separate occasions by the first author to ensure it was correctly inputted and accurate. A second rater checked the final data extracted against each study and any disagreements were resolved via discussion.

1.3.7 Data Synthesis

A meta-analysis was considered unsuitable for this review because the studies included used a variety of designs and produced largely descriptive results, meaning data was heterogenous. Moreover, the review question itself was broad and did not seek to measure or evaluate study outcomes. It instead aimed to describe outcomes of studies, and therefore a narrative synthesis was deemed more appropriate. Data synthesis was grouped according to the outcomes the review was interested in, and sub-grouped based on whether the studies included direct comparisons to participants with CTDs.

1.4 Results

1.4.1 Overview Of Included Studies

Database searches identified *k* = 1007 records; *k* = 458 duplicates were removed, and the remaining *k* = 549 records were screened (Fig. 1). A total of *k* = 498 records were determined to be irrelevant to the study and were excluded. Following this, the full texts of the remaining *k* = 51 reports were screened and *k* = 14 were eliminated for not meeting the inclusion criteria. Moreover, *k* = 1 report was excluded as the authors were unable to translate it to English. The

first author identified an additional report whilst writing the literature review that was not found during database searching but was relevant to the search terms. The full text was screened and included resulting in a total of *k* = 37 reports (*k* = 33 studies) being included in this systematic review.

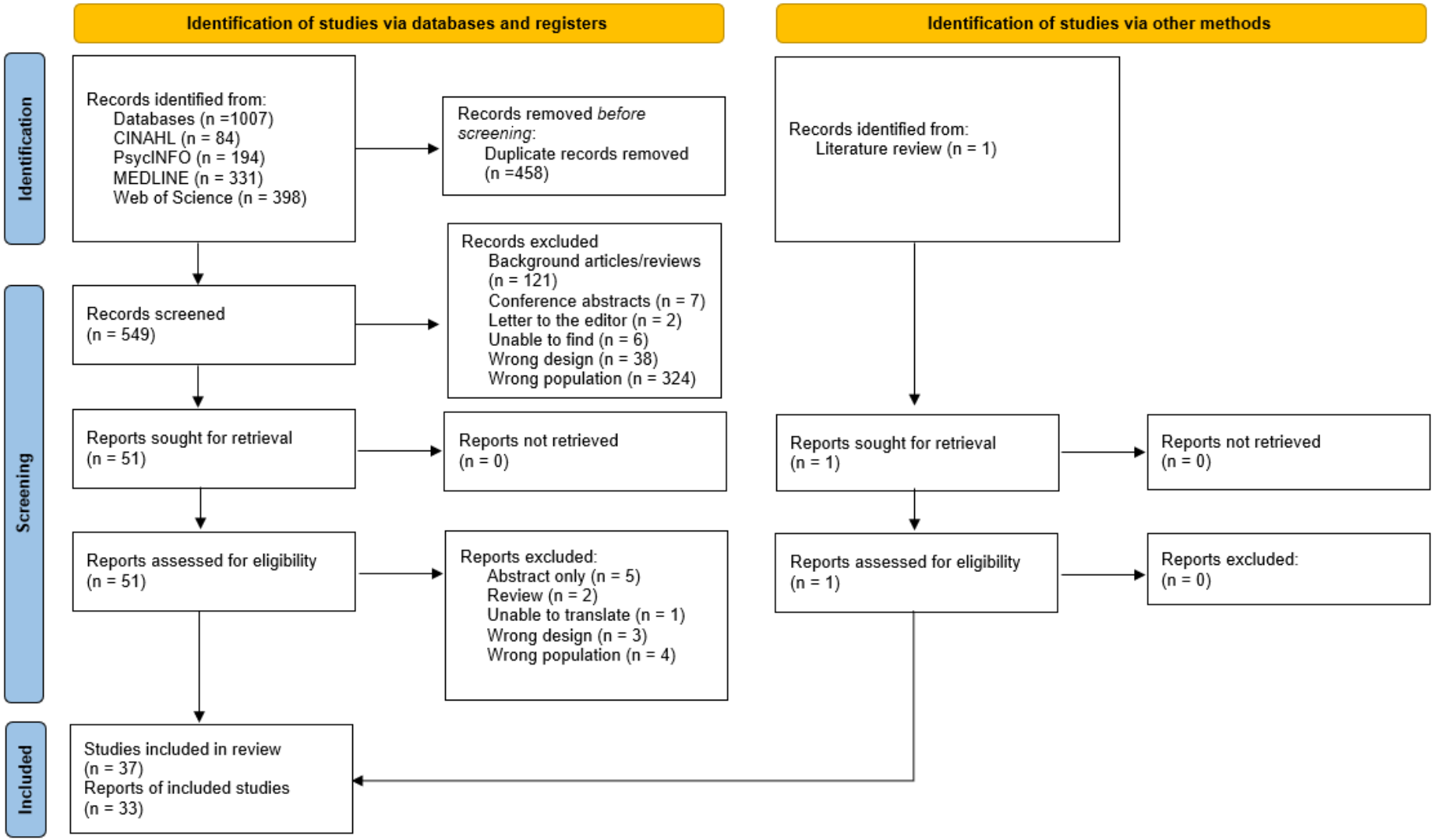


Fig. 1 PRISMA flow diagram

1.4.2 Study Characteristics

Overall, *N* = 1452 participants were included in *k* = 33 studies published between 2014 and 2025, and characteristics of each study are provided in Table 2. Sample sizes varied from *n* = 8 to *n* = 294, and the majority of studies recruited both child and adult (≥18yrs) samples (*k* = 17) [17, 25, 30-48] followed by children only (*k* = 13) [49-61] and adults only (*k* = 3) [62-64]. A cross-sectional design was used in *k* = 17 studies [30-35, 40-43, 45, 46, 48, 49, 53, 56, 61-64], *k* = 8 were of a longitudinal design [36, 37, 39-41, 47, 52, 54, 57, 59, 60], *k* = 7 were case series [17, 25, 38, 44, 50, 55, 58], and *k* = 1 was an experimental design [51]. Of the *k* = 33 studies, *k* = 16 compared participants with FTLB to participants with CTDs [30-32, 35-37, 39-42, 45, 46, 49, 54, 56, 59, 61-64]. The majority of studies (*k* = 32) recruited samples from specialist hospital clinics and *k* = 1 [53] collected participants from a school setting. Moreover, *k* = 31 collected data from a single centre and were conducted in the following countries: United States (*k* = 9) [49, 53, 55-57, 60-63], United Kingdom (*k* = 7) [17, 25, 33-35, 51, 52], Canada (*k* = 5) [31, 32, 39-41, 46, 47, 64], Germany (*k* = 4)[36-38, 45, 48], Denmark (*k* = 2) [30, 58], Australia (*k* = 2) [44, 54], Italy (*k* = 1) [59], and Poland (*k* = 1) [42]. Moreover, *k* = 1 study [50] was conducted in both the United Kingdom and Canada, and *k* = 1 study [43] was conducted in 10 centres across the following eight counties: Australia, Canada, France, Hungary, Italy, Germany, United Kingdom, and United States.

Of the *N* = 1452 participants, 83.5% (*n* = 1213) were assigned female sex. Of the *k* = 4 studies [31, 32, 50, 51, 57] reporting ethnicity, the majority of participants were of White ethic origin. The majority of studies (*k* = 17) [30-32, 39-41, 43-47, 50-54, 56, 59, 60, 64] diagnosed participants with FTLB but the remaining studies used different terminology to describe this diagnosis such as: functional tics (*k* = 8) [17, 25, 33-35, 57, 58, 62], functional tic disorder (*k* = 2) [49, 61], psychogenic tics (*k* = 1) [42], psychogenic movements resembling tics (*k* = 1) [63], MSMI-FTLB (*k* = 1) [36, 37], tic-like behaviours following social media consumption (TLB-SM) (*k* = 1) [48], abrupt onset tic-like movements (*k* = 1) [55], and functional tic-like vocalisations (*k* = 1) [38]. The majority of studies (*k* = 21) [25, 31-35, 39-41, 44, 46-50, 52-55, 57-61] included participants who received a diagnosis of FTLB during the COVID-19 pandemic, *k* = 4 studies [17, 38, 42, 63] included participants diagnosed prior to COVID-19, and *k* = 8 studies [30, 36, 37, 43, 45, 51, 56, 62, 64] did not specify when participants were diagnosed. A variety of professionals were reported to provide participants with a diagnosis of FTLB including a neurologist (*k* = 12) [17, 30-37, 39-42, 44, 52, 57], neuro-paediatrician (*k* = 2) [44, 58], psychologist (*k* = 3) [44, 51, 52], psychiatrist (*k* = 4) [36, 37, 44, 51, 52], and unspecified clinicians (*k* = 9) [25, 43, 45, 47, 49, 53, 54, 62, 63]. Diagnosis provider was not specified in *k* = 10 studies [38, 46, 48, 50, 55, 56, 59-61, 64].

Table 2 Overview of included studies

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Anderson, 2023 [30]: DK	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children and adults (<20yrs) **Comparator(s):** TS/CTD **FU:** n/a	**Sample Size:** *N* = 53 **Mean Age (SD):** 14.9 (2.0), range 11-20 **Female Sex (%):** 50 (94.3) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 15 (28.3) unspecified, 14 (26.4) anxiety, 10 (18.9) ADHD, 10 (18.9) OCD, 7 (13.2) ASD	**Dx:** FTLB **Criteria used:** NR **Assessed by:** Neurologist **Received diagnosis:** May 2020 – Apr 2022	13.7 (2.4), range 6-19.8	0.77 (high)
Armstrong-Javors, 2024 [49]: US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** TS/CTD/TTD **FU:** n/a	**Sample Size:** *N* = 19 **Mean Age (SD):** NR **Female Sex (%):** 18 (94.7) **Gender (%):** 18 (94.7) female, 7 (36.8) sexual orientation/gender minority **Ethnicity (%):** NR **Comorbid (%):** 9 (47.4) ADHD, 8 (42.1) OCD, 7 (36.8) FNS	**Dx:** FTD **Criteria used:** ESSTS [16] **Assessed by:** Clinicians **Received diagnosis:** May 2018 – Jan 2022	14.2, range 10-17	0.59 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Baizabal-Carvallo, 2023 [62]: US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Adults (≥18yrs) **Comparator(s):** TS **FU:** n/a	**Sample Size:** *N* = 21 **Mean Age (SD):** 35.7 (16.0) **Female Sex (%):** 10 (47.6) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 5 (23.8) OCD, 3 (14.3) ADHD	**Dx:** FT **Criteria used:** Fahn and Williams [65] **Assessed by:** Clinician **Received diagnosis:** NR	31.6 (15.3)	0.77 (high)
Baizabal-Carvallo, 2014 [63]: US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Adults (≥18yrs) **Comparator(s):** TS, PMD **FU:** n/a	**Sample Size:** *N* = 9 **Mean Age (SD):** 26.3 (16.9) **Female Sex (%):** 5 (55.6) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 9 (100) PMD	**Dx:** PMRT **Criteria used:** Fahn and Williams [65] **Assessed by:** Clinician **Received diagnosis:** Jan 2009 – Jul 2012	34.1 (17.3), range 16-66	0.55 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Berg, 2024 [31, 32]:CA	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children and adults **Comparator(s):** TS, Controls **FU:** n/a	**Sample Size:** *N* = 35 **Mean Age (SD):** 17.5 (3.1) **Female Sex (%):** 32 (91.4) **Gender (%):** 20 (57.1) cisgender, 15 (42.5) transgender and gender diverse **Ethnicity (%)[a]:** (21.2) ethnic minority **Comorbid (%):** 8/28pps (28.6) ADHD, 3/28pps (10.7) FNS	**Dx:** FTLB **Criteria used:** ESSTS [16] **Assessed by:** Neurologist **Received diagnosis:** Oct 2020 – Jun 2022	15.2 (3.7)	0.95 (high)
Buts, 2022 [50]: CA, UK	**Design:** Case series **Recruitment:** Clinic **Population:** Children (≤17yrs) **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 34 **Mean Age (SD):** 14.0 (1.4) **Female Sex (%):** 32 (94.1) **Gender (%):** NR **Ethnicity (%):** 27 (79.4) Caucasian **Comorbid (%):** 23 (67.6) anxiety, 8 (23.5) depression, 7 (20.6) ADHD, 5 (14.7) learning difficulty, 4 (11.8) ASD, 4 (11.8) OCD, 1 (2.9) intellectual disability	**Dx:** FTLB **Criteria used:** Self-developed **Assessed by:** NR **Received diagnosis:** Nov 2020 – Apr 2021	13.7	0.70 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Cavanna, 2022 [25]: UK	**Design:** Case series **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** Within-subjects **FU:** n/a	**Sample Size:** *N* = 10 **Mean Age (SD):** 18.4 (2.9), range 13-24 **Female Sex (%):** 9 (90.0) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 8 (80.0) OCB, 7 (70.0) anxiety, 7 (70.0) FNS, 5 (50.0) affective disorder, 3 (30.0) OCD, 3 (30.0) ADHD, 2 (20.0) ASD	**Dx:** TS + FT **Criteria used:** NHIS GTS[66] **Assessed by:** Clinician **Received diagnosis:** Mar 2020 – Oct 2022	16.9 (2.3), range 12-22	0.73 (medium)
Cavanna, 2023 [33]: UK	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** FMD **FU:** n/a	**Sample Size:** *N* = 66 **Mean Age (SD):** 23.1 (10.5), range 13-63 **Female Sex (%):** 47 (71.2) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 46 (69.7) anxiety, 22 (33.3) NES, 20 (30.3) affective disorder	**Dx:** FT **Criteria used:** NHIS GTS [66] (adapted for FTLB) **Assessed by:** Neurologist **Received diagnosis:** Apr 2020 – Apr 2023	21.1 (10.6), range 11-61	0.73 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Cavanna, 2023 [34]: UK	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 105 **Mean Age (SD):** 23.2 (10.7), range 13-63 **Female Sex (%):** 76 (72.4) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 73 (69.5) anxiety, 43 (41.0) FND, 42 (40.0) affective disorder, 34 (32.4) NES, 28 (26.7) ASD, 24 (2.9) OCB, 24 (22.9) TS, 22 (21.0) FMD, 19 (18.1) ADHD, 10 (9.5) OCD	**Dx:** FT **Criteria used:** NHIS GTS [66] (adapted for FTLB) **Assessed by:** Neurologist **Received diagnosis:** Apr 2020 – Mar 2023	21.4 (10.8), range 11-61yrs	0.64 (medium
Cavanna, 2023 [35]: UK	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** TS **FU:** n/a	**Sample Size:** *N* = 83 **Mean Age (SD):** 23.2 (10.7), range 13-63 **Female Sex (%):** 59 (71.1) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 58 (69.9) anxiety, 39 (47.0) FND, 32 (38.6) affective disorder, 31 (37.3) NES, 21 (25.3) ASD, 17 (20.5) FMD, 9 (10.8) ADHD, 6 (7.2) OCB, 3 (3.6) OCD	**Dx:** FT **Criteria used:** NHIS GTS [66] **Assessed by:** Neurologist **Received diagnosis:** Apr 2020 – Mar 2023	21.2 (10.9), range 11–61 years	0.77 (high)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Demartini, 2015 [17]: UK	**Design:** Case series **Recruitment:** Clinic **Population:** Children (≥16yrs) and adults **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 11 **Mean Age (SD):** 37.2 (13.5), range 16-65 **Female Sex (%):** 3 (27.3) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 8 (72.7) FNS, 5 (45.5) depression, 3 (27.3) anxiety	**Dx:** FT **Criteria used:** NR **Assessed by:** Neurologist **Received diagnosis:** Jan 2011 – Oct 2013	NR	0.60 (medium)
Ducroizet, 2025 [52]: UK	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** None **FU:** NR	**Sample Size:** *N* = 43 **Mean Age (SD):** 14.2 (1.8) **Female Sex (%):** 43 (100) **Gender (%):** 40 (93.0) cisgender, 2 (4.7) non-binary, 1 (2.3) transgender **Ethnicity (%):** NR **Comorbid (%):** 30 (69.8) anxiety or OCD, 26 (60.5) FNS, 8 (18.6) ASD, 5 (11.6) depression, 4 (9.3) ADHD	**Dx:** FTLB **Criteria used:** ESSTS [16] **Assessed by:** Neurologist, psychiatrist, and psychologist **Received diagnosis:** Jan 2020 – Mar 2023	13.0 (1.7)	0.73 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Duncan, 2024 [51]: UK	**Design:** Experimental **Recruitment:** Clinic **Population:** Children (≤17yrs) **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 58 **Mean Age (SD):** 14.3 (2.1) **Female Sex (%):** 59 (71.1) **Gender (%):** 52 (85.9) cisgender, 6 (10.5) non-binary or transgender **Ethnicity (%):**[a] (50.9) White British, (5.3) Asian Indian, (1.8) Black British, (1.8) mixed ethnicity **Comorbid (%):** NR	**Dx:** FTLB **Criteria used:** NR **Assessed by:** Psychologist and psychiatrist **Received diagnosis:** NR	NR	0.91 (high)
Firestone, 2023 [53]: US	**Design:** Cross-Sectional **Recruitment:** School **Population:** Children (<18yrs) **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 8 **Mean Age (SD):** 16.0 **Female Sex (%):** 8 (100) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid:** 5 (63.5) depression, 2 (25.0) ADHD, 1 (12.5) OCD	**Dx:** FTLB **Criteria used:** Self-developed **Assessed by:** clinicians **Received diagnosis:** Sep 2021 – Nov 2021	16.0, range 15-17	0.40 (low)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Fremer, 2024 [36, 37]: DE	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** TS/CTD **FU:** *M* = 4.8 months (range: 6 days – 19 months)	**Sample Size:** *N* = 32 **Mean Age (SD):** 20.1, range 11-53 **Female Sex (%):** 16 (50.0) **Gender (%):** 2 (6.3) non-binary **Ethnicity (%):** NR **Comorbid (%):** 15 (46.9) OCB, 13 (40.6) anxiety, 10 (31.3) depression, 8 (25.0) sleeping difficulties, 6 (18.8) personality disorder, 5 (15.6) ASD, 3 (9.4) ADHD, 3 (9.4) ID, 2 (6.3) post-traumatic stress disorder	**Dx:** MSI-FTLB **Criteria used:** NR **Assessed by:** Neurologist and psychiatrist **Received diagnosis:** May 2019 – Sept 2021	19.2 (11.0)	0.82 (high)
Ganos, 2016 [38]: DE	**Design:** Case series **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 13 **Mean Age (SD):** NR, range 10-56 **Female Sex (%):** 4 (30.8) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 5 (38.5) FND, 4 ADHD, 1 (7.7) OCD	**Dx:** Functional tic-like vocalisations **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** 1995 - 2015	Range 5-50	0.60 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Han, 2022 [54]: AU	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** TS/CTD **FU**: range: 2 months – 3 years	**Sample Size:** *N* = 22 **Mean Age (SD):** NR **Female Sex (%):** 22 (100) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 21 (95.5) anxiety or depression, 6 (27.3) TS, 5 (22.7) OCD, 3 (13.6) ADHD, 2 (9.1) ASD	**Dx:** FTLB **Criteria used:** NR **Assessed by:** Clinicians **Received diagnosis:** 2018 – Jul 2021	13.8	0.68 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Howlett, 2022 [39-41]:CA	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** PTD **FU:** 6 months	**Sample Size:** *N* = 29 *Adolescents (n = 20)* **Mean Age (SD):** 14.3 **Female Sex (%):** 19 (95.0) **Gender (%):** 11 cis-gender (55.0), 9 (45.0) transgender, non-binary, or gender fluid **Ethnicity (%):** NR **Comorbid (%):** 15 (75.0) anxiety, 11 (55.0) depression, 5 (25.0) ADHD, 5 (25.0) OCD, 0 (0) FND *Adults (n = 9)* **Mean Age (SD):** 19.9 **Female Sex (%):** 8 (88.9) **Gender (%):** 7 (77.8) cisgender, 2 transgender (22.2) **Ethnicity (%):** NR **Comorbid (%):** 5 (55.6) anxiety, 4 (44.4) depression, 2 (22.2) ADHD, 1 (11.1) substance use, 1 (11.1) FMD, 0 (0) OCD	**Dx:** FTLB **Criteria used:** NR **Assessed by:** Neurologist **Received diagnosis:** 2012– 2021 (adolescents); Jan – June 2021 (adults)	**Adolescents:** 13.9 **Adults:** 15.3	0.73 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Hull, 2021 [55]: US	**Design:** Case series **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 6 **Mean Age (SD):** NR **Female Sex (%):** 6 (100) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 3 (50.0) FMD, 3 (50.0) depression, 2 (33.3) anxiety, 1 (16.7) post-traumatic stress disorder	**Dx:** Abrupt onset tic-like movements **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** Nov 2020 – Jan 2021	14.2, range 13-16	0.60 (medium)
Janik, 2014 [42]: PL	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** PTD **FU:** n/a	**Sample Size:** *N* = 5 **Mean Age (SD):** NR **Female Sex (%):** 1 (20.0) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 1 (20.0) conversion disorder, 1 (20.0) depression, 1 (20.0) personality disorder, 1 (20.0) ADHD, 1 (20.0) OCD	**Dx:** PT **Criteria used:** NR **Assessed by:** Neurologist **Received diagnosis:** 1998 - 2012	34.0 (16.6), range 17-54	0.55 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Larsh, 2022 [56]: US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** TS **FU:** n/a	**Sample Size:** *N* = 89 **Mean Age (SD):** 15.6 (2.0) **Female Sex (%):** 83 (93.2) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 66 (74.4) OCD or anxiety, 45 (50.6) ADHD, 26 (29.2) OCB	**Dx:** FTLB **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** 2021	NR	0.86 (high)
Martino, 2023 [43]: AU, CA, DE FR, HU, IT, UK, US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 294 **Mean Age (SD):** 15.1 (5.0), range 8-53 **Female Sex (%):** 255 (86.7) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 195 (66.3) anxiety disorder, 94 (32.0) FNS, 81 (27.6) depressive disorder, 71 (24.1) ASD, 68 (23.1) ADHD, 56 (19.0) PTD, 27 (9.2) OCD	**Dx:** FTLB **Criteria used:** Self-developed **Assessed by:** Clinician **Received diagnosis:** Oct 2019- Jun 22	NR	0.73 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Mathew, 2023 [57]: US	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** None **FU:** *M* = 198 days	**Sample Size:** *N* = 29 **Mean Age (SD):** 15.9 (1.40) **Female Sex (%):** 26 (89.7) **Gender (%):** 2 (6.9) non-binary, 1 (3.4) transgender **Ethnicity (%):** 25 (86.2) White, 2 (6.9) Black 1 (3.4) Hispanic, 1 (3.4) other **Comorbid (%)**: 20 (69) mood disorder, 19 (65.5) anxiety, 10 (34.5) FNS, 3 (10.3) ADHD	**Dx:** FT **Criteria used:** NR **Assessed by:** Neurologist **Received diagnosis:** Mar 2020 – Dec 2021	NR	0.73 (medium)
Maxwell, 2023 [44]: AU	**Design:** Case series **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** 2– 8 months	**Sample Size:** *N* = 8 **Mean Age (SD):** NR **Female Sex (%):** 8 (100) **Gender (%):** 1 (12.5) non-binary **Ethnicity (%):** NR **Comorbid (%)**: 8 (100) anxiety, 3 (37.5) major depressive disorder, 2 (25.0) ASD, 1 (12.5) OCB, 1 (12.5) selective mutism, 1 (12.5) functional paralysis	**Dx:** FTLB **Criteria used:** ESSTS [16] **Assessed by:** Psychologist and neurologist/paediatrician/psychiatrist **Received diagnosis:** 2019 - 2023	Range 12-20	0.59 (medium)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Müller-Vahl, 2024 [45]: DE	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** TS **FU:** n/a	**Sample Size:** *N* = 71 **Mean Age (SD):** 21.5, range 11-55 **Female Sex (%):** 27 (38.0) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 41 (57.7) OCS, 26 (36.6) ADHD, 24 (33.8) MUS, 23 (32.4) depression, 19 (26.7) anxiety, 17 (23.9) OCD, 2 (2.8) ASD	**Dx:** TS + FTLB **Criteria used:** Self-developed **Assessed by:** Clinicians **Received diagnosis:** 2002 – 2021	20.8 (11.8), range: 5–52	0.64 (medium)
Nilles, 2024 [46]: CA	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children and adults (<20yrs) **Comparator(s):** PTD **FU:** n/a	**Sample Size:** *N* = 41 **Mean Age (SD):** 16.1, range 11-20 **Female Sex (%):** 40 (97.6) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** NR	**Dx:** FTLB **Criteria used:** Self-developed **Assessed by:** NR **Received diagnosis:** NR	NR	0.86 (high)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Nilles, 2024 [47]: CA	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** 6 and 12 months	**Sample Size:** *N* = 83 **Mean Age (SD):** 18 (5.6), range 11-53 **Female Sex (%):** 80 (96.4) **Gender (%):** 64 (77.1) cisgender, 11 (13.3) transgender, 8 (9.6) non-binary **Ethnicity (%):** NR **Comorbid (%):** 54 (65.1) anxiety, 35 (42.2) major depressive disorder, 29 (34.9) ADHD, 19 (22.9) FNS, 6 (7.2) OCD, 3 (3.6) ASD	**Dx:** FTLB **Criteria used:** NR **Assessed by:** Clinician **Received diagnosis:** Oct 2020 – Dec 2022	NR	0.82 (high)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Okkels, 2023 [58]: DK	**Design:** Case series **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** None **FU:** *M* = 127 days (range: 14-266 days)	**Sample Size:** *N* = 28 **Mean Age (SD):** 14.7, range 11-18.9 **Female Sex (%):** 27 (96.4) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):**[a] (66.0) PTSD, (47.1) OCD, (41.2) ADHD, (35.3) anxiety, (35.3) depression, NR (28.6) sleep difficulties, (17.9) NR dyslexia, NR (<13.0) ASD, NR (<13.0) PTD	**Dx:** FT **Criteria used:** NR **Assessed by:** Neuro-paediatrician **Received diagnosis:** May 2020 – Jun 2021	NR	0.75 (high)
Paulus, 2021[48]: DE	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) and adults **Comparator(s):** None **FU:** n/a	**Sample Size:** *N* = 13 **Mean Age (SD):** 16.5 (3.1), range 12 -24 **Female Sex (%):** 5 (38.5) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 3 (28.3) OCD, 1 (7.7) ADHD, 1 (7.7) ASD	**Dx:** TLB-SM **Criteria used:** Self-developed **Assessed by:** NR **Received diagnosis:** NR	15.3 (3.0)	0.86 (high)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Prato, 2023 [59]: IT	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** TS/CTD **FU:** 6 and 12 months	**Sample Size:** *N* = 11 **Mean Age (SD):** 14.8 (2.6), range 11-18 **Female Sex (%):** 8 (72.7) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 7 (63.6) FNS	**Dx:** FTLB **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** Jun 2021- Jun 2022	14.0 (2.6), range 11-18	0.68 (medium)
Szejko, 2024 [64]: CA	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Adults (≥18yrs) and adults **Comparator(s):** TS **FU:** n/a	**Sample Size:** *N* = 40 **Mean Age (SD):** 20.7 (3.2) **Female Sex (%):** 36 (90.0) **Gender (%):** 31 (77.5) female, 2 (5.0) male, 7 (17.5) gender minority **Ethnicity (%):** NR **Comorbid (%):** NR	**Dx:** FTLB **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** NR	16.8 (5.7)	0.91 (high)

First Author, Year: Country	Study Characteristics	Sample Characteristics	Diagnosis Characteristics	Mean age of FTLB onset (*SD*)	Quality Score (rating)
Tomczak, 2024 [60]: US	**Design:** Longitudinal **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** None **FU:** *M* = 518 days (range:137-894 days)	**Sample Size:** *N* = 56 **Mean Age (SD):** NR **Female Sex (%):** 54 (96.4) **Gender (%):** 31 (55.4) cisgender, 25 (44.6) gender diverse **Ethnicity (%):** NR **Comorbid (%):** 52 (92.9) anxiety, 40 (71.4) depression, 25 (44.6) ADHD, 18 (32.1) NES, 13 (23.2) OCD, 13 (23.2) dissociative disorder, 7 (12.5) paralysis or gait impairment, 4 (7.1) ASD, 4 (7.1) psychogenic tremor	**Dx:** FTLB **Criteria used:** NR **Assessed by:** NR **Received diagnosis:** Mar 2020 – Onwards	14 (1.9)	0.82 (high)
Trau, 2022 [61]: US	**Design:** Cross-Sectional **Recruitment:** Clinic **Population:** Children (<18yrs) **Comparator(s):** PTD, MTD **FU:** n/a	**Sample Size:** *N = 36* **Mean Age (SD):** NR **Female Sex (%):** 34 (94.4) **Gender (%):** NR **Ethnicity (%):** NR **Comorbid (%):** 32 (89.9) anxiety, 25 (69.4) ADHD, 20 (55.6) OCB, 6 (16.7) FNS	**Dx:** FTD **Criteria used:** Self-developed **Assessed by:** NR **Received diagnosis:** May 2020 – Dec 2021	14.0 (2.0)	0.77 (high)

Abbreviations: DK Denmark, US United States, CA Canada, UK United Kingdom, DE Germany, AU Australia, PL Poland, FR France, HU Hungary, IT Italy, NR not reported, pps participants, TS Tourette syndrome, CTD chronic tic disorder, TTD transient tic disorder, PTD primary tic disorder, MTD mixed tic disorder, PMD psychogenic movement disorder, FMD functional movement disorder, FND functional neurological disorder, FNS functional neurological symptoms, FTLB functional tic-like behaviours, FTD functional tic disorder, FT functional tics, PMRT psychogenic movements resembling tics, MSMI-FTLB mass social media-induced illness functional tic-like behaviours, PT psychogenic tics, TLB-SM tic like behaviours following social media consumption, ADHD attention deficit hyperactivity disorder, ASD autism spectrum disorder, OCB obsessive compulsive behaviours, OCD obsessive compulsive disorder, NES non-epileptic seizures, MUS medically unexplained symptoms, NHIS GTS National Hospital Interview Schedule for the Assessment of Gilles de la Tourette Syndrome, ESSTS European society for the study of Tourette syndrome

[a] Number of participants is not specified as percentages only provided by the study and do not match the total sample size, suggesting some participants may not have been asked about characteristics.

1.4.3 Quality Appraisal

Quality assessment ratings are provided for each study (Table 2) and ranged from high to low quality, but only *k* = 1 study [53] was identified as low quality. Individual item scores for each study are shown in Online Resource 2. The selection strategy for all studies was likely to introduce bias as participants were recruited via specialist movement disorder clinics or via clinicians known to them. The majority of studies (*k* = 22) [30, 33-35, 39-41, 43-47, 49, 50, 52, 54, 56-59, 61-64] appeared to have a small sample size and failed to report effect sizes or variance estimates to assess whether samples were adequately powered. In particular, *k* = 5 studies [17, 25, 38, 42, 53, 55] were identified as having extremely small sample sizes that were not representative of the whole FTLB population, with the majority being case series. Another common flaw was the lack of detailed reporting of outcome measures and questions/response options given to participants to generate results, with only *k* = 8 studies [25, 31, 32, 48, 51, 56, 60, 62, 64] providing sufficient detail to minimise the likelihood of measurement error.

1.4.4 Sex and Gender of Participants with FTLB

All studies included the assigned sex of participants (Table 2) and *k* = 27 of these [30-35, 39-41, 43, 44, 46-61, 63, 64] reported the majority of participants with FTLB were of assigned-female sex, with rates ranging from 55.5% to 100%. However, *k* = 1 high-quality study [36, 37] found an equal ratio between assigned-female and assigned-male sex, and *k* = 5 studies [17, 38, 42, 45, 62] ranging from high to medium quality reported a lower incidence of FTLB in assigned-female sex participants (ranging from 20.0% to 47.6%) in comparison to those assigned-male sex.

Only *k* = 11 studies [31, 32, 36, 37, 39-41, 44, 47, 49, 51, 52, 57, 60, 64] provided information on the gender identity of participants and were considered either high or medium quality research. The majority of these studies (*k* = 6) [31, 32, 39-41, 47, 51, 52, 60] recorded the number of participants identifying as cisgender, with prevalence ranging from 55.4% to 93.0% of samples. In total, *k* = 9 studies [31, 32, 36, 37, 39-41, 44, 47, 51, 52, 57, 60] used terminology such as non-binary, transgender, gender diverse, or gender fluid to describe participants that identified as a different gender to their assigned sex. The terms sexual orientation and gender identity minority (*k* = 1) [49] and gender minority (*k* = 1) [64] were also used to describe this identity. Of the *n* = 432 participants described in the *k* = 11 studies, *n* = 143 (33.1%) identified as gender diverse, with individual studies reporting a prevalence of between 2.3% and 44.6% (Table 2). The majority of studies (*k* = 6) [31, 32, 49, 51, 52, 57, 60] reporting gender identity

included children participants only, followed by *k* = 4 studies [36, 37, 39-41, 44, 47] including mixed aged samples, and *k* = 1 [64] using adult participants only.

1.4.4.1 Sex and Gender of Participants with FTLB Compared to CTDs

When compared to CTDs, *k* = 13 studies [30-32, 36, 37, 39-41, 45, 46, 54, 56, 59, 61-64] found participants with FTLB were more likely to be assigned-female sex. Interestingly, *k* = 1 study [42] reported a higher incidence of FTLB in male-assigned sex. Whilst this study was considered medium quality, the score was on the cut-off between low and medium quality, raising questions towards the reliability and validity of the results. Only *k* = 1 study [31, 32] investigated differences in rates of gender identity between participants with FTLB and CTDs. This study [31, 32] was of the highest quality research and found higher rates of gender diverse individuals in the FTLB group.

1.4.5 Age of Onset of FTLB

Overall, *k* = 24 studies [25, 30-37, 39-42, 44, 45, 48-50, 52-55, 59-64] varying from high to low quality reported the age participants developed FTLB, and the average age ranged from 13 years old to 31 years old (Table 2). Studies investigating participants diagnosed with FTLB during COVID-19 (*k* = 20) [25, 30-37, 39-41, 44-50, 52-55, 57-61] found age of onset ranged from 5 years old to 61 years old. A similar age of onset was found in studies including participants diagnosed prior to COVID-19 (*k* = 3) [38, 42, 63], which ranged from 5 years old to 66 years old. However, *k* = 1 study [49] directly compared age of onset between participants who developed FTLB before COVID-19 (*M* age = 12.5) and during the pandemic (*M* age = 16.0).

1.4.5.1 Age of Onset of FTLB Compared to CTDs

Overall, *k* = 11 studies [30-32, 35-37, 39, 45, 54, 59, 61, 62, 64] compared symptom onset in participants with FTLB to those with CTDs and each study found age of onset was significantly higher in participants with FTLB, regardless of when the diagnosis was given. Moreover, *k* = 1 high quality study [35] included participants with Tourette syndrome and comorbid FTLB, and found onset of FTLB were later in 100% of cases, and on average developed 14 years after tics associated with CTDs.

1.4.6 Acuteness of FTLB

There were *k* = 3 studies [17, 42, 48] that found 100% of participants reported a rapid onset of FTLB compared to *k* = 9 studies [25, 31-35, 38, 45, 52, 59] which found between 54.0% and 93.0% of participants experienced an acute or subacute onset of FTLB. However, *k* = 3

studies [36, 37, 57, 58] reported only a minority of participants experienced sudden and rapid onset of FTLB.

1.4.6.1 Acuteness of FTLB Compared to CTDs

When comparing onset of symptoms, *k* = 1 study [45] of medium quality evidence found FTLB had an acute/subacute onset in comparison to tics associated with CTDs which, reportedly, developed more gradually. This is supported by another high quality study [35], in which none of the participants with CTDs reported experiencing a sudden or rapid onset of symptoms in comparison to those with FTLB who reported an acute onset.

1.4.7 Triggers Precipitating Onset of FTLB

A total of *k* = 12 studies [17, 30, 33-35, 38, 43, 45, 53, 58-60] ranging from high to low quality investigated whether participants recalled a precipitating event prior to onset of FTLB. The majority of studies found participants reported a psychological trigger including stress/anxiety (*k* = 4) [34, 43, 53, 60], significant life events (*k* = 3) [38, 43, 58], relationship and work/school difficulties (*k* = 3) [43, 45, 59], bullying (*k* = 1) [38], and traumatic experiences (*k* = 1) [58] in the days and months before symptom onset. The number of participants reporting a psychological trigger ranged from 45.5% to 79.2%. Moreover, *k* = 1 medium quality study [33] highlighted social media as a possible trigger for FTLB, as 47.0% of *n* = 66 children reported FTLB developed following social media consumption. Participants also named the COVID-19 pandemic as a particular trigger for FTLB onset in *k* = 4 studies [43, 58-60] of both high and medium quality. Moreover, 50.0% of participants in *k* = 1 [53] study reported testing positive for COVID-19 prior to experiencing symptoms of FTLB, however this study was considered to have the lowest quality of evidence and thus this finding should be treated with caution.

1.4.7.1 Triggers Precipitating Onset of FTLB Compared to CTDs

Only *k* = 1 study [30] compared the presence of a trigger prior to symptom onset between participants with FTLB and CTDs. This study [30] was found to be high quality research and revealed children and adults with FTLB were more likely to experience a psychological trigger before symptom onset compared to CTDs.

1.4.8 Prevalence of Comorbid Anxiety in FTLB

Table 2 provides the proportion of participants in each study with specific comorbid diagnoses. The majority of studies (*k* = 20) [17, 25, 30, 33-37, 43-45, 47, 50, 52, 54-58, 60, 61] reported the prevalence of anxiety disorders in participants with FTLB via medical records, self-

report, or clinical assessments. Whereas *k* = 1 study [39-41], rated as medium quality, used screening tools to diagnose anxiety, including the multidimensional anxiety scale for children version 2 and the generalized anxiety disorder-7 (GAD-7) questionnaire for adult participants. Rates of co-occurring anxiety disorders found in studies reporting child samples (*k* = 10) [39-41, 50, 52, 54-58, 60, 61] ranged from 23.5% to 95.5%, whereas, rates of co-occurring anxiety disorders in studies reporting mixed (*k* = 11) [17, 25, 30, 33-37, 43-45, 47] and adult samples (*k* = 1) [39-41] ranged from 26.4% to 100%. Only *k* = 1 study [53] used the GAD-7 in conjunction with medical records to identify current anxiety symptoms; scores indicated a higher proportion of participants met diagnostic criteria for comorbid anxiety compared to pre-existing records although this study was assessed as low quality research and thus the findings may be questionable.

1.4.8.1 Prevalence of Comorbid Anxiety in FTLB Compared to CTDs

Only a few studies (*k* = 3) [35, 54, 61], ranging from high to medium quality, found significantly higher rates of anxiety disorders in participants with FTLB compared to CTDs, and *k* = 1 medium quality study [39-41] reported a strong association between comorbid anxiety and diagnosis of FTLB. Alternatively, *k* = 4 studies [30, 36, 37, 45, 56] of both high and medium quality evidence found similar rates of co-occurring anxiety disorders in participants with FTLB compared to CTDs, even in participants with both Tourette syndrome and comorbid FTLB.

1.4.9 Prevalence of Comorbid Depression in FTLB

When reporting rates of comorbid depression in participants with FTLB, various terminology was used with the majority of studies (*k* = 12) [17, 36, 37, 39-42, 45, 50, 52-55, 58, 60] referring to this disorder as depression, followed by affective disorder (*k* = 4) [25, 33-35], major depressive disorder (*k* = 3) [43, 44, 47], and mood disorder (*k* = 1) [57]. Only *k* = 1 study [39-41] used the child depression inventory and patient health questionnaire (PHQ-9) to aid diagnosis, and *k* = 1 study [53] required participants to complete the PHQ-9 and found current incidence of depression was higher than pre-existing diagnosis of depression, but as aforementioned, this study was found to be the lowest quality research. A higher prevalence of comorbid depression was found in studies reporting child samples (*k* = 8) [39, 40, 50, 52-55, 57, 58, 60] with rates ranging from 11.6% to 95.5% in comparison to studies including mixed age (*k* = 11) [17, 25, 33-37, 42-45, 47] and adult samples (*k* = 1) [39-41] which reported a prevalence rate ranging between 20.0% and 50.0%.

1.4.9.1 Prevalence of Comorbid Depression in FTLB Compared to CTDs

Findings were mixed when reporting differences in prevalence of depression in participants with FTLB and CTDs. For instance, *k* = 1 study [54] of medium quality research reported significantly higher rates of comorbid depression in participants with FTLB compared to CTDs but no differences were found in *k* = 3 studies [35-37, 45] varying between high and medium quality evidence. However, *k* = 1 study of medium quality research [39-41] explored the relationship between comorbidities and FTLB and found a diagnosis of depression increased the likelihood of receiving a diagnosis of FTLB in children.

1.4.10 Prevalence of Comorbid OCD in FTLB

Only *k* = 1 study [39-41], identified as medium quality, used screening tools such as the Children's Yale-Brown Obsessive Compulsive Scale and Obsessive-Compulsive Inventory, alongside medical records to diagnose comorbid OCD in participants with FTLB. The prevalence of co-occurring OCD in adult samples (*k* = 2) [39-41, 62] ranged between 0% and 23.8% whereas studies combining child and adult prevalence (*k* = 10) [25, 30, 34, 35, 38, 42, 43, 45, 47, 48] found higher rates ranging between 7.7% and 30.0%. Moreover, the highest prevalence, ranging from 11.8% to 74.4% was found in studies reporting rates of OCD in children (*k* = 8) [39-41, 49, 50, 52, 53, 56, 58, 60]. However, *k* = 1 medium quality study [52] and *k* = 1 high quality study [56] combined anxiety and OCD rates, and if they were removed prevalence of OCD would only range between 11.8% and 47.1%. Similarly, the rates of obsessive compulsive behaviours (OCB) in participants with FTLB ranged from 2.9% to 80.0% across studies including mixed age samples (*k* = 6) [25, 34-37, 44, 45] and child participants only (*k* = 2) [56, 61].

1.4.10.1 Prevalence of Comorbid OCD in FTLB Compared to CTDs

Study findings varied when comparing prevalence of comorbid OCD and OCB in participants with FTLB and CTDs, as *k* = 2 high quality studies [35-37] found participants with FTLB had significantly lower rates of OCD/OCB compared to CTDs. Whereas *k* = 1 medium quality study [45] found OCD was more prevalent in participants with FTLB, but rates of OCB were no different in those with FTLB or CTDs. Furthermore, *k* = 2 studies [30, 56] rated as high quality found no differences in prevalence rates of OCD/OCB in either participant group.

1.4.11 Prevalence of Comorbid Neurodevelopmental Disorders in FTLB

Comorbid ADHD was diagnosed via medical records and clinical assessment in the majority of studies (*k* = 22) [25, 30-32, 34-38, 42, 43, 45, 47-50, 52-54, 57, 58, 60-62], and only *k* = 1 study, rated as medium quality, used the Conners and adult self-rating report scale for

ADHD to aid diagnosis [39-41]. Similar to other comorbidities, the prevalence of ADHD was higher in studies with child participants (*k* = 10) [39, 40, 49, 50, 52-54, 57, 58, 60, 61] in comparison to adults (*k* = 2) [39, 41, 62] and mixed age samples (*k* = 12) [25, 30-32, 34-38, 42, 43, 45, 47, 48], with rates ranging from 9.3% to 69.0%, 14.3% to 22.0%, and 4.0% to 37.0%, respectively.

A total of *k* = 10 studies [25, 30, 34-37, 43-45, 47, 48, 52] including both children and adults in their sample reported the prevalence of autism spectrum disorder (ASD) in participants with FTLB and found this ranged from 3.0% to 26.7%. Studies including child samples only (*k* = 4) [50, 52, 54, 58] found similar rates of comorbid ASD reporting a range of 9.0% to 18.6% in participants with FTLB.

Only *k* = 4 studies [34, 43, 54, 58] reported the prevalence of co-occurring CTDs in participants with FTLB (not including studies recruiting participants with both Tourette syndrome and FTLB) and this ranged from 13.0% to 27.3%. An additional *k* = 6 studies [43, 44, 49, 53, 59, 60, 64] reported the number of participants who had a history of tics or previously diagnosed CTDs in childhood, and this ranged from 0% to 45.5%.

1.4.11.1 Prevalence of Comorbid Neurodevelopmental Disorders in FTLB Compared to CTDs

A total of *k* = 3 studies [35-37, 54] of ranging from high to medium quality found participants with FTLB were less likely to have a diagnosis of ADHD in comparison to those with CTDs. However, *k* = 3 studies [30, 45, 61] found minimal differences in rates of ADHD between participants with FTLB and CTDs. In contrast, *k* = 1 high quality study [35] found participants with FTLB were significantly more likely to have an ASD diagnosis compared to participants with CTDs. On the other hand, *k* = 3 studies [36, 37, 45, 54] varying from high to medium quality found no differences in prevalence rates of ASD between FTLB and CTDs.

1.4.12 Prevalence of Comorbid FND in FTLB

Multiple studies reported the prevalence of FND and/or subtypes of this diagnosis (e.g. FMD) but were unclear whether participants were classed as only having one or multiple types. Thus, prevalence rates are grouped based on the terminologies used in studies to prevent participants being counted multiple times. The majority of studies used the term FNS (*k* = 9) [17, 25, 43, 47, 49, 52, 57, 59, 61] and found prevalence ranged between 17.0% to 72.7% in participants with FTLB. In comparison, rates of FND were lower varying from 0% to 38.5% across *k* = 5 studies [31, 32, 34, 35, 38-40] alongside FMD which prevalence was found to range between 11.1% to 50.0% in *k* = 4 studies [34, 35, 39, 41, 55]. In addition, *k* = 4 studies [33-35, 60]

found between 32.1% and 37.3% of participants had a comorbid diagnosis of non-epileptic seizures, and *k* = 2 studies [44, 57] found 12.5% of participants had co-occurring functional paralysis. In addition, *k* = 1 study [63] reported 100% of participants had a co-occurring psychogenic movement disorder and *k* = 1 study [45] found 33.8% of participants were diagnosed with medically unexplained symptoms. Rates of co-occurring psychological tremor and gait impairment were low in *k* = 1 study [60], and *k* = 1 study [42] found only *n* = 1 participant to have comorbid conversion disorder although this was of the lowest quality research.

1.4.12.1 Prevalence of Comorbid FND in FTLB Compared to CTDs

Only *k* = 2 high quality studies [35, 61] compared the prevalence of FND in participants with FTLB against participants with CTDs and both found significantly higher rates of this comorbidity in the former compared to the latter group of participants.

1.4.13 Prevalence of Additional Comorbid Conditions in FTLB

Multiple studies included prevalence rates of additional co-occurring diagnoses including post-traumatic stress disorder (*k* = 3) [36, 37, 55, 58], intellectual disabilities (*k* = 2) [36, 37, 50], specific learning difficulties (*k* = 2) [50, 58], personality disorders (*k* = 2) [36, 37, 42], substance misuse (*k* = 1) [39, 41], selective mutism (*k* = 1) [44], and sleep difficulties (*k* = 2) [36, 37, 58], however rates of these were low.

1.4.14 Types of Tics that Occur in FTLB

Participants with FTLB reported a variety of simple and complex motor and phonic tics across *k* = 29 studies [17, 25, 30, 34-40, 42-50, 53-59, 61-63], however the level of detail varied with some studies reporting the number of participants with specific tics, and others only reporting the presence of motor/phonic or simple/complex tics (Table 3). The prevalence of coprolaliac tics ranged from 0% to 87.5% across *k* = 16 high to medium quality studies [17, 25, 34-40, 42-48, 54, 57], whereas rates of copropraxia ranged from 0% to 53.8% in high to medium quality studies [17, 25, 34-37, 44-48, 54, 57]. However, *k* = 6 studies [30, 49, 50, 58, 59, 61] ranging from high to medium quality research reported rates of coprophenomena tics in general, with prevalence ranging from 22.2% to 76.5%. Moreover, *k* = 8 studies [25, 34-37, 43, 50, 58, 61] reported whether FTLB followed a rostro-caudal progression, and *k* = 7 studies found the majority of participants did not experience this slow progression of tics from the head to lower extremities. Participants reported multiple body parts were affected in *k* = 5 studies [43, 44, 48, 57, 62] that ranged from high to medium quality and specific tics such as blocking, forced touching, and throwing objects were found in *k* = 11 studies [17, 25, 35-37, 43, 44, 46, 47, 55, 61].

1.4.14.1 Differences in Types of Tics That Occur in FTLB Compared to CTDs

In studies that compared participants with FTLB to those with CTDs, rates of coprophenomena were higher in those diagnosed with FTLB (*k* = 6) [30, 35, 45, 46, 54, 61]. Studies of high quality research found FTLB were more likely to include blocking and throwing tics (*k* = 3) [35, 46, 61] and less likely to have simple tics and rostro-caudal progression (*k* = 3) [30, 35, 61]. However, findings differed as to whether FTLB were more or less likely to have complex motor tics in comparison to CTDs in studies of high to medium quality (*k* = 4) [30, 35, 54, 62].

Table 3 Overview of the types of tics reported in studies of participants with FTLB

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Anderson, 2023 [30]	***N*** = 53 **Dx:** FTLB **Population:** Children	**Simple:** 39 (73.6) **Complex:** 44 (83.0) **Specific tics:** 23 (43.4) self-injurious tics	**Simple:** 30 (56.6) **Complex:** 30 (56.6) **Specific tics:** 25 (47.2) unrestrained speech	16 (30.2) unspecified coprophenomena **NOSIB:** NR	NR
Armstrong-Javors 2024 [49]	***N*** = 19 **Dx:** FTD **Population:** Children and adults	**Simple:** NR **Complex**: NR **Specific tics:** 42 (48.8) self-injurious tics	**Simple:** NR **Complex:** NR **Specific tics:** NR	5 (26.3) unspecified coprophenomena **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Baizabal-Carvallo, 2023 [62]	***N*** = 21 **Dx:** FT **Population:** Adults	**Simple:** 21 (100) **Complex:** 2 (9.5) **Specific tics:** 6 (28.6) eye blinking, 4 (19.0) facial grimacing, 3 (14.3) eyerolling	**Simple:** 3 (14.3) **Complex**: 0 (0) **Specific tics:** NR	**Coprolalia:** NR **Copropraxia:** NR **NOSIB:** NR	15 (71.4%) report tics occur in face, 10 (47.6%) report tics occur in neck, 8 (38.1%) report tics occur in shoulders, 6 (23.8%) report tics occur in arms, 5 (23.8%) report tics occur in trunk, 2 (9.5%) report tics occur in legs
Baizabal-Carvallo, 2014 [63]	***N*** = 9 **Dx:** PT **Population:** Adults	9 (100) unspecified motor tics **Specific tics:** 4 (44.4) facial grimacing, 4 (44.4) shoulder movements	3 (33.3) unspecified phonic tics	**Coprolalia:** NR **Copropraxia:** NR **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Buts, 2022 [50]	***N*** = 34 **Dx:** FTLB **Population:** Children	**Simple:** NR **Complex:** NR **Specific tics**: NR	**Simple:** NR **Complex:** NR **Specific tics:** NR	26 (76.5) unspecified coprophenomena	21 (61.8%) report first tics had rostro-caudal distribution
Cavanna, 2022 [25]	***N*** = 10 **Dx:** TS + FT **Population:** Children and adults	**Simple:** 6 (60.0) **Complex:** 10 (100) Specific tics: 8 (80.0) forced touching, 7 (70.0) self-injurious tics, 7 (70.0) palipraxia	**Simple:** 6 (60.0) **Complex:** 8 (80.0) Specific tics: 6 (60.0) echolalia/echopraxia/palilalia	**Coprolalia:** 8 (80.0) **Copropraxia:** 5 (50.0) **NOSIB:** 8 (80.0)	2 (20.0%) report rostro-caudal distribution
Cavanna, 2023 [34]	***N*** = 105 **Dx:** FT **Population:** Children and adults	**Simple:** 91 (86.7) **Complex:** 85 (81.0) **Specific tics:** 41 (39.0) self-injurious tics, 16 (15.2) throwing tics, 12 (11.4) forced touching	**Simple:** 83 (79.0) **Complex:** 79 (75.2) **Specific tics:** NR	**Coprolalia:** 51 (48.6) **Copropraxia:** 21 (20.0) **NOSIB:** NR	16 (15.2%) report rostro-caudal distribution

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Cavanna, 2023 [35]	***N*** = 83 **Dx:** FT **Population:** Children and adults	**Simple:** 72 (86.7) **Complex:** 67 (80.7) **Specific tics:** 27 (32.5) self-injurious tics, 16 (19.3) throwing tics, 12 (14.4) blocking tics, 8 (9.6) forced touching	**Simple:** 65 (78.3) **Complex:** 65 (78.3) **Specific tics:** NR	**Coprolalia:** 44 (53.0) **Copropraxia:** 15 (18.1) **NOSIB:** NR	13 (15.7%) report rostro-caudal distribution
Demartini, 2015 [17]	***N*** = 11 **Dx**: FT **Population:** Children and adults	**Simple:** 5 (45.5) **Complex:** 79 (75.2) **Specific tics:** 4 (36.4) blocking tics, 3 (27.3) head movements, 2 (18.2) eye blinking	**Simple:** NR **Complex:** NR **Specific tics:** NR	**Coprolalia:** 0 (0) **Copropraxia:** 0 (0) **NOSIB:** NR	NR
Firestone, 2023 [53]	***N*** = 8 **Dx:** FTLB **Population:** Children	**Simple:** 8 (100) **Complex:** NR **Specific tics:** 8 (100) head jerks, 5 (62.5) shoulder shrugs, 3 (37.5) eye-blinking, 3 (37.5%) abdominal tensing	**Simple:** 1 (12.5) **Complex:** 2 (25.0) **Specific tics:** 2 (25.0) syllables/words/phrases, 1 (12.5) nose whistling	**Coprolalia:** NR **Copropraxia:** NR **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Fremer, 2024 [36, 37]	***N*** = 32 **Dx:** MSMI-FTLB **Population:** Children and adults	**Simple:** 19 (59.4) **Complex:** 32 (100) **Specific tics:** 24 (75.0) head movements, 20 (62.5) arm movements, 20 (62.5) throwing objects/food, 15 (46.9) touching others, 15 (46.9) torso movements, 15 (46.9) self-injurious tics, 12 (37.5) hurting/hitting others, 12 (37.5) hand movements, 12 (37.5) destroying objects, 12 (37.5) kicking/hitting objects, 11 (34.4) face movements, 10 (31.3) leg movements, 8 (25.0) altered gait	**Simple:** 28 (87.5) **Complex:** 32 (100) **Specific tics:** 11 (34.4) animal sounds, 9 (28.1) syllables, 7 (21.9) mouth clicking, 6 (18.8) whistling, 5 (15.6) screaming	**Coprolalia:** 8-13 (25.0 – 40.6) **Copropraxia:** 17 (53.1) **NOSIB:** NR	1 (3.1%) report rostro-caudal distribution

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Ganos, 2016 [38]	***N*** = 13 **Dx:** Functional tic-like vocalisations **Population:** Children and adults	11 (84.6) unspecified motor tics	**Simple:** 9 (69.2) **Complex:** NR **Specific tics:** 10 (76.9) palilalia, 5 (38.5) echolalia	**Coprolalia:** 7 (53.8) **Copropraxia:** NR **NOSIB:** 2 (15.4)	NR
Han, 2022 [54]	***N*** = 22 **Dx:** FTLB **Population:** Children	**Simple:** NR **Complex:** NR **Specific tics:** 17 (77.3) head jerks/nods, 11 (50.0) self-injurious tics ,7 (31.8) eye blinking/facial twitching, 5 (22.7) shoulder shrugging, 3 (13.6) tongue thrusting, 2 (9.1) thumping of chest	**Simple:** NR **Complex:** NR **Specific tics:** 10 (45.5) complex words/phrases, 7 (31.8) whistling	**Coprolalia:** 17 (77.3) **Copropraxia:** 10 (45.5) **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Howlett, 2022 [39-41]	***N*** = 29 **Dx:** FTLB **Population:** Children (*n*=20) and adults (*n*=9)	**Simple:** NR **Complex:** 13 (65.0) adolescents, 8 (88.8) adults **Specific tics:** 14 (70.0) self-injurious tics (adolescents)	**Simple:** NR **Complex:** 18 (90.0) adolescents, 8 (89.9) adults **Specific tics:** NR	**Coprolalia:** 11 (55.0) adolescents, (*n* and % adults NR) **Copropraxia:** NR **NOSIB:** NR	NR
Hull, 2021 [55]	***N*** = 6 **Dx:** Abrupt onset tic-like movements **Population:** Children	**Simple:** NR **Complex:** NR **Specific tics:** 4 (66.7) neck flexion/extension ,4 (66.7) self-injurious tics, 3 (50.0) adduction of arms, 3 (50.0) punching contralateral palm, 2 (33.3) extension of thumbs, 2 (33.3) throwing objects, 2 (33.3) blowing kisses	6 (100) unspecified phonic tics **Specific tics:** NR	**Coprolalia:** NR **Copropraxia:** NR **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Janik, 2014 [42]	***N*** = 5 **Dx:** PT **Population:** Children and adults	**Simple:** 0 (0) **Complex:** 4 (80.0) **Specific tics:** NR	**Simple:** NR **Complex:** NR **Specific tics:** 1 (20.0) palilalia/echolalia	**Coprolalia:** 2 (40.0) **Copropraxia:** NR **NOSIB:** NR	NR
Larsh, 2022 [56]	***N*** = 89 **Dx:** FTLB **Population:** Children	**Simple:** NR **Complex:** NR **Specific tics:** 42 (48.8) self-injurious tics	**Simple:** NR **Complex:** NR **Specific tics:** NR	**Coprolalia**: NR **Copropraxia:** NR **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Martino, 2023 [43]	***N*** = 294 **Dx:** FTLB **Population:** Children and adults	**Simple:** 261 (88.8) **Complex:** 251 (85.4) **Specific tics:** 48 (16.3) finger movements, 51 (17.3) flinging/throwing objects, 37 (12.6) chest thumping, 118 (40.1) self-injurious tics, 89 (35.7) hitting own body, other people or objects	**Simple:** 241 (81.9) **Complex:** 239 (81.3) **Specific tics:** 141 (48.0) nonsensical language, 85 (28.0) whistling, 71 (24.1) context-dependent words, 57 (19.4) echolalia, 56 (19.0) palilalia, 42 (14.3) tongue clicking, 38 (14.3) sniffing, 38 (12.9) animal sounds	**Coprolalia:** 150 (51.0) **Copropraxia:** NR **NOSIB:** NR	187 (63.6%) report tics occur in cervical region, 125 (42.5%) report tics occur ocular region, 104 (35.4%) report tics occur in shoulder region, 39 (13.3%) report tics occur as whole-body movements, 47 (16.0%) report rostro-caudal distribution
Mathew, 2023 [57]	***N*** = 29 **Dx:** FT **Population:** Children	**Simple:** NR **Complex:** <85%[a] **Specific tics:** 0 (0) echopraxia, (<25)[a] injurious tics towards self or others	**Simple:** NR **Complex:** <50%[a] **Specific tics:** <10%[a] echolalia	**Coprolalia:** 5 (18.6) **Copropraxia:** 3 (10.3) **NOSIB**: NR	Between 87.5% and 95.0% report tics occur in trunk and limbs

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Maxwell, 2023 [44]	***N*** = 8 **Dx:** FTLB **Population:** Children and adults	**Simple:** 3 (37.5) **Complex:** 7 (87.5) **Specific tics:** 5 (62.5) throwing things, 4 (50.0) self-injurious tics, 3 (37.5) winking, 3 (37.5) shoulder shrugs, 3 (37.5) hitting/kicking or slapping, 3 (37.5) head bang/jerk/nod, 2 (25.0) echopraxia, 1 (12.5) blinking, 1 (12.5) grinding teeth, 1 (12.5) jaw protrusion, 1 (12.5) hold legs to chest, 1 (12.5) clapping, 1 (12.5) jumping, 1 (12.5) stomping feet, 1 (12.5) bending over	**Simple:** 3 (37.5) **Complex:** 6 (75.0) **Specific tics:** 1 (12.5) squeaking, 1 (12.5) change accent/tone, 1 (12.5) popping/clicking sounds, 1 (12.5) throat clearing, 1 (12.5) snorting ,1 (12.5) echolalia	**Coprolalia:** 7 (87.5) **Copropraxia:** 2 (25.0) **NOSIB:** 3 (37.5)	1 (12.5%) report whole body movements
Müller-Vahl, 2024 [45]	***N*** = 71 **Dx:** TS + FTLB **Population:** Children and adults	**Simple:** NR **Complex:** 57 (80.3) **Specific tics:** 29 (40.8) self-injurious tics	**Simple:** NR **Complex:** 28 (39.4) **Specific tics:** NR	**Coprolalia:** 27 (38.0) **Copropraxia:** 7 (9.9) **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Nilles, 2024 [46]	***N*** = 41 **Dx:** FTLB **Population:** Children and adults	**Simple:** NR **Complex:** NR **Specific tics:** 37 (90.2) head tics, 25 (61.0) eye blinking, 22 (53.7) self-injurious tics, 22 (53.7) simple and 14 (34.0) complex arm movements, 22 (53.7) shoulder shrugs, 18 (43.9) simple nose movements, 18 (43.9) facial grimacing, 16 (39.0) simple eye movements, 14 (34.1) simple mouth movements, 12 (29.3) complex hand movements, 10 (24.4) complex head movements, 8 (19.5) blocking tics, 3 (7.3) writing tics	**Simple:** NR **Complex:** NR **Specific tics:** 20 (48.8) enunciation of words, 15 (36.6) sniffing, 13 (31.7) echolalia, 11 (26.8) whistling, 10 (24.4) syllables, 7 (17.1) clicking, 6 (14.6) popping, 5 (12.2) throat clearing, 4 (9.8) phrases, 3 (7.3) coughing	**Coprolalia:** 18 (43.9) **Copropraxia:** 15 (36.6) **NOSIB:** NR	NR

Nilles, 2024 [47]	***N*** = 83 **Dx:** FTLB **Population:** Children and adults	**Simple:** NR **Complex:** NR **Specific tics:** 71 (85.5) head jerks, 41 (49.4) shoulder shrugs, 43 (51.8) simple and 24 (28.9) complex arm movements, 35 (42.2) self-injurious tics, 32 (38.6) simple and 6 (7.2) complex eye movements, 31 (37.3) nose movements, 30 (36.1) facial grimacing, 27 (32.5) simple and 3 (3.6) complex mouth movements, 25 (30.1) simple and 12 (14.5) complex leg/foot movements, 24 (28.9) hand movements, 22 (55.4) eye blinking, 20 (24.1) head movements, 15 (18.1) blocking tics, 13 (15.7) bending or gyrating, 10 (12.0), abdominal tensing, 6 (7.2) dystonic postures, 3 (3.6) shoulder movements, 3 (3.6) writing tics, 2 (2.4) disinhibited behaviour	**Simple:** NR **Complex:** NR **Specific tics:** 45 (54.2) words, 27 (32.5) echolalia, 26 (31.3) syllables, 25 (30.1) whistling, 23 (27.7) sniffing, 18 (21.7) clicking, 17 (20.5) popping, 16 (19.3) palilalia, 15 (18.1) humming, 15 (18.1) speech blocking, 13 (15.7) mouth noises, 12 (14.5) throat clearing, 12 (14.5) phrases, 11 (13.3) animal noises, 8 (9.6) disinhibited speech, 7 (8.4) screeching, 5 (6.0) coughing, 4 (4.8) grunting, 4 (4.8) wheezing, 4 (4.8) barking, 3 (3.6) hiccups, 3 (3.6) forceful exhalation, 2 (2.4) kissing noises, 2 (2.4) gasping, 1	**Coprolalia:** 36 (43.4) **Copropraxia:** 25 (30.1) **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
			(1.2) gulping, 1 (1.2) burping, 0 (0) snorting		
Okkels, 2023 [58]	***N*** = 28 **Dx:** FT **Population:** Children	**Simple:** 18 (64.3) **Complex:** 26 (92.9) **Specific tics:** 18 (69.2) injurious tics towards self or others	**Simple:** 10 (35.7) **Complex:** 20 (71.4) **Specific tics:** NR	9 (32.1) unspecified coprophenomena NOSIB: NR	0 (0%) report rostro-caudal distribution
Paulus, 2021 [48]	***N*** = 13 **Dx:** TLB-SM **Population:** Children and adults	**Simple:** NR **Complex:** 12 (92.3) **Specific tics:** 5/9pps (55.6) echopraxia	**Simple:** 9/12pps (75.0) **Complex:** 5/12pps (41.7) **Specific tics:** 5/10pps (50.0) echolalia	**Coprolalia:** 5 (38.5) **Copropraxia:** 7 (53.8) **NOSIB:** NR	12 (92.3%) report tics are slow and tonic 2 (15.4%) report tics occur only in trunk and extremities 0 (0%) report tics only occur in face/head/neck
Prato, 2023 [59]	***N*** = 11 **Dx:** FTLB **Population:** Children	**Simple:** NR **Complex:** 11 (100) **Specific tics:** NR	**Simple:** NR **Complex:** 7 (63.6) **Specific tics:** NR	7 (63.6) unspecified coprophenomena **NOSIB:** NR	NR

First Author, Year	Sample Characteristics	Motor Tics, *n* (%)	Phonic Tics, *n* (%)	Coprophenomena, *n* (%)	Tic Distribution
Trau, 2022 [61]	***N*** = 36 **Dx:** FTD **Population:** Children	**Simple:** NR **Complex:** NR **Specific tics:** 21 (58.4) injurious tics, 6 (16.7) throwing tics, 4 (11.1) blocking tics	**Simple:** NR **Complex:** NR **Specific tics:** 26 (72.2) broad/extended words	8 (22.2) unspecified coprophenomena **NOSIB:** NR	1 (2.8%) report rostro-caudal progression

Abbreviations: FTLB functional tic-like behaviours, FTD functional tic disorder, FT functional tics, MSMI FTLB mass social media-induced illness functional tic like-behaviours, PT psychogenic tics, TLB-SM tic like behaviours following social media consumption, TS Tourette syndrome, pps participants, NR not reported, NOSIB non-obscene socially inappropriate behaviours

[a] Study does not provide exact number of participants or percentages

1.4.15 Intensity, Impairment, and Severity of FTLB

Studies reported mixed findings as to whether FTLB waxed and waned. For instance, *k* = 1 mixed sample study [17] of medium quality found none of the participants experienced waxing and waning of FTLB whereas another medium quality study (*k* = 1) [50] reported the presence of this feature in 32.0% of child participants. Similarly, FTLB were found to be intermittent in the majority of participants in *k* = 1 medium quality study [34] but the number or participants reporting fluctuating symptoms of FTLB varied across high to medium studies (*k* = 3) [25, 38, 48], with rates ranging from 0% to 100% of participants.

Participants indicated varying levels of impairment from FTLB (*k* = 2) [45, 57] including missing school (*k* = 4) [31, 32, 44, 52, 54] or requiring home-schooling (*k* = 2) [52, 56]. The Children's Global Assessment Scale (CGAS), a clinician-rated measure of overall functioning, was used in *k* = 3 high to medium quality studies [50-52] of child cohorts and participants scored an average of 44-51, indicating moderate impairment in most social aspects of life [67]. Moreover, *k* = 1 study [52] found the mean CGAS score improved from 51.1 to 57.1 at follow up but functioning remained in the moderate range. Studies also used the Yale Global Tic Severity Scale (YGTSS) [68] to measure current or worst-ever impairment, however *k* = 4 studies [25, 39-41, 44, 47] ranging from medium to high quality did not specify which time-period they rated. Average participant scores ranged from 17 to 32, indicating impairment varied between minimal, mild and moderate [68]. However, *k* = 1 study [31, 32], which was rated to have the highest quality evidence, reported both current and worst-ever ratings and participants averaged scores of 20 and 40 respectively. Moreover, *k* = 1 study [44] of medium quality evidence found the mean YGTSS impairment score reduced from 31.3 to 0 following a course of integrated-cognitive behavioural intervention for functional tics (i-CBiT).

Participants reported the presence of self-injurious tics (such as hitting/slapping self) in *k* = 17 studies [25, 30, 34-37, 39-41, 43-47, 54-58, 61], with a minimum of 20.0% of cases being affected in each study (Table 3). FTLB were described as painful in *k* = 1 high quality study [56], and another high quality study (*k* = 1) [36, 37] using a mixed sample found 46.9% of participants had been injured by their FTLB but did not require medical attention. Alternatively, *k* = 5 studies [44, 50, 54, 56, 57] varying in high to medium quality found a number of participants (ranging from 12.5% to 48.0%) required medical attention or hospitalisation due to the severity of FTLB. Participants described FTLB as occurring in long bursts, clusters, or as tic attacks in *k* = 3 studies of children [54, 55, 61] and in *k* = 4 mixed cohorts [30, 35, 44, 45] all of which were either deemed as high or medium quality research.

Only *k* = 11 studies [25, 31, 32, 39-41, 44, 45, 47, 50, 51, 59, 61, 63] used clinician rated tools to measure symptom severity in participants with FTLB and mean scores are reported in Table 4. Comparisons were unable to be made due to studies using different measures or reporting different subscales of the same tool, and not specifying whether scores were based on current or historical symptoms. However, out of the *k* = 9 high to medium quality studies [25, 31, 32, 39-41, 44, 47, 50, 51, 59, 61] using the YGTSS, *k* = 5 [39-41, 44, 47, 59, 61] found a significant reduction in symptom severity at follow ups.

1.4.15.1 Intensity, Impairment and Severity of FTLB Compared to CTDs

Only *k* = 1 medium quality study [34] looking into the intensity of FTLB and tics in CTDs found participants with CTDs reported greater intermittency in symptoms. Alternatively, *k* = 1 study [31, 32] rated as the highest quality comparing impairment of CTDs and FTLB found no differences in participants with either diagnosis. However, higher rates of self-injurious tics (*k* = 3) [35, 45, 61] were found in participants with FTLB when compared to CTDs, and greater symptom severity was associated with an increased likelihood of FTLB diagnosis (*k* = 1) [39, 41], all of which were found in studies ranging from high to medium quality.

Table 4 Mean scores on severity measures in included studies

Author, Year	Sample Characteristics	Severity Measure
Baizabal-Carvallo, 2014 [63]	***N*** = 9 **Dx:** PT **Population:** Adults **FU:** n/a	**Measure:** Global Severity Rating and Rush Video-Based Tic Rating Scales **Total Possible Score:** 6 **Mean Score (SD):** 2.8 (1.1)
Berg, 2024 [31, 32]	***N*** = 35 **Dx:** FTLB **Population:** Children and adults **FU:** n/a	**Measure:** YGTSS – Global Score **Total Possible Score:** 100 **Mean Score (SD):** Current: 44.3 (24.4), Worst-ever: 82.8 (16.8)
Buts, 2022 [50]	***N*** = 34 **Dx:** FTLB **Population:** Children **FU:** n/a	**Measure:** YGTSS – Severity Score **Total Possible Score:** 100 **Mean Score (SD):** 62.6 (19.0)
Cavanna, 2022 [25]	***N*** = 10 **Dx:** TS + FT **Population:** Children and adults **FU:** n/a	**Measure:** YGTSS – Severity Score **Total Possible Score:** 50 **Mean Score (SD):** 25.4 (10.2)

Author, Year	Sample Characteristics	Severity Measure
Duncan, 2024 [51]	***N*** = 58 **Dx:** FTLB **Population:** Children **FU:** n/a	**Measure:** YGTSS – Global Score **Total Possible Score:** 100 **Mean Score (SD):** 63.4 (18.4)
Howlett, 2022 [39-41]	***N*** = 29 **Dx:** FTLB **Population:** Children (*n*=20) and adults (*n*=9) **FU:** 6 months	*Adolescents* **Measure:** YGTSS – Global Score **Total Possible Score:** 100 **Mean Score (SD):** 64.5 at initial visit, 32.5 at follow up *Adults* **Measure:** YGTSS – Global Score **Total Possible Score:** 100 **Mean Score (SD):** 63.7 at initial visit, 32.5 at follow up
Maxwell, 2023 [44]	***N*** = 8 **Dx:** FTLB **Population:** Children and adults **FU:** 2– 8 months	**Measure:** YGTSS – Severity Score **Total Possible Score:** 50 **Mean Score (SD):** 35.0 (11.0) pre-therapy, 8.5 (6.4) post-therapy

Author, Year	Sample Characteristics	Severity Measure
Müller-Vahl, 2024 [45]	***N*** = 71 **Dx:** TS + FTLB **Population:** Children and adults FU: n/a	**Measure:** Shapiro TS Severity Scale **Total Possible Score:** 9 (1= mild and 9=severe) **Mean Score (SD):** NR – 63% scored as mild, 4% mild to moderate, 7% moderate, 6% moderate to severe, 6% severe severity
Nilles, 2024 [47]	***N*** = 41 **Dx:** FTLB **Population:** Children and adults FU: 6 and 12 months	**Measure:** YGTSS – Severity Score **Total Possible Score:** 50 **Mean Score (SD):** 29.8 (10.0) at initial visit, 20.9 (12.4) at 6 month follow up, 14.6 (13.6) at 12 month follow up
Prato, 2023 [59]	***N*** = 11 **Dx:** FTLB **Population:** Children **FU:** 6 and 12 months	**Measure:** YGTSS – Severity Score **Total Possible Score:** 50 **Mean Score (SD):** 32.5 (14.9) at initial visit, 29.7 (9.9) at 6 month follow up, 24.4 (11.7) at 12 month follow up
Trau, 2022 [61]	***N*** = 36 **Dx:** FTD **Population:** Children **FU:** n/a	**Measure:** YGTSS – Global Score **Total Possible Score:** 100 **Mean Score (SD):** 53.0 (25.0) at initial visit, 49 (29.0) at follow up

Abbreviations: Dx Diagnosis, FU follow up, FTLB functional tic-like behaviours, FTD functional tic disorder, FT functional tics, PT psychogenic tics, TS Tourette syndrome, YGTSS Yale Global Tic Severity Scale,

1.4.16 Specific Triggers of FTLB

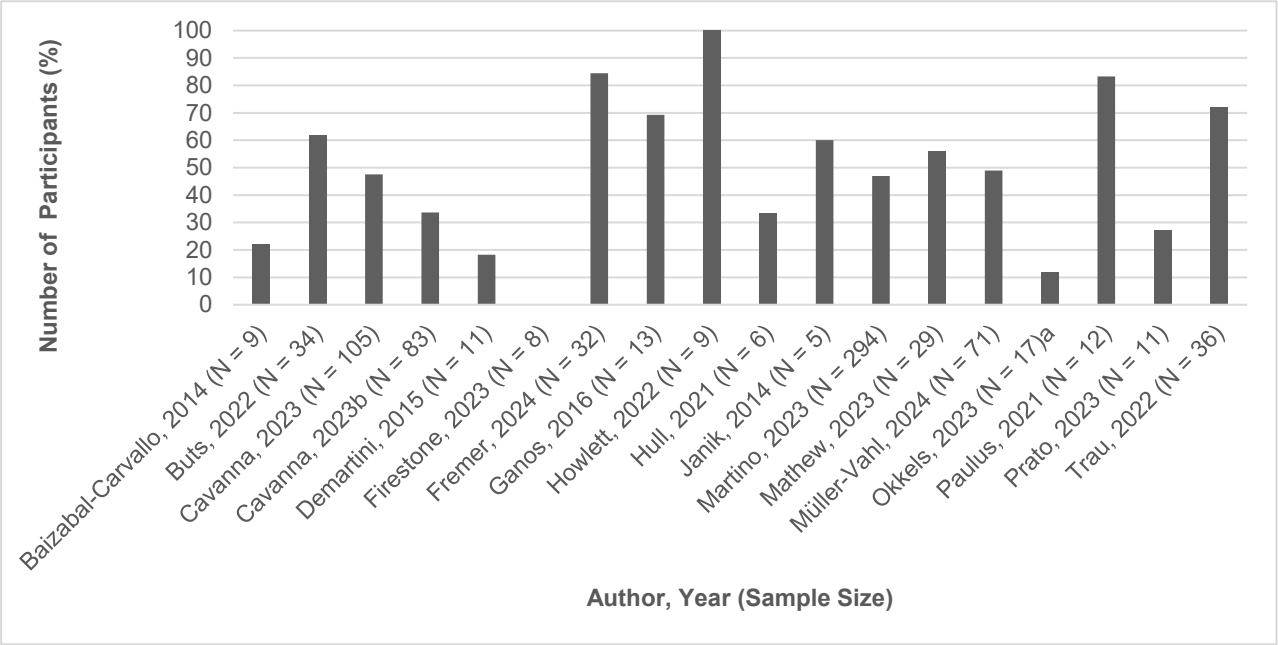
Participants identified triggers for FTLB, with the majority (*k* = 4) reporting symptoms worsened around other people, particularly relatives [36, 37, 48, 53, 57]. This is followed by stress and anxiety which reportedly affected 32.0% to 74.7% of participants (*k* = 3) [43, 45, 53], although these studies ranged from medium to low quality. Alternatively, participants in *k* = 1 high quality study [36, 37] reported being in the presence of other people improved their symptoms. Practicing sports, concentrating on tasks, relaxing, gaming, being with pets, and being alone were also identified as activities that improved FTLB (*k* = 1) [36, 37]. However, participants also reported specific triggers that worsened FTLB such as specific sounds or words/phrases (*k* = 2) [36, 37, 55], seeing a particular type of person (e.g. policeman) (*k* = 1) [36, 37], certain places, daily activities, and body positions (*k* = 1) [45]. Sensory triggers such as loud noises, extreme temperatures, and flashing lights were identified as triggering increased FTLB in *n* = 5 of *N* = 6 children in *k* = 1 medium quality study [55]. Only *k* = 1 study [43], considered to have medium quality evidence, compared triggers of tics between FTLBs and CTDs and found children with FTLB reported higher rates of tic-contingent triggers compared to adults with FTLB.

1.4.17 Prevalence of Premonitory Urges and Suppressibility in FTLB

A total of *k* = 18 studies [17, 34-39, 41-43, 45, 48, 50, 55, 57-59, 61-63] found participants experienced a premonitory urge prior to expressing FTLB, however prevalence varied amongst studies (Fig. 2). For instance, *k* = 1 study [53] found no participants recalled experiencing a premonitory urge, although this study was rated to have the lowest quality evidence and thus may not be an accurate representation of this population. The premonitory urge for tics scale (PUTS) was administered to adult participants in *k* = 1 medium quality study [39, 41] and to a sample of children in a high quality study (*k* = 1) [58]; both found participants had a mean score of 26. Furthermore, *k* = 1 high quality research study [36, 37] investigated the duration of premonitory urges and found they typically lasted around 67 seconds and ranged between 1 second and 1 hour.

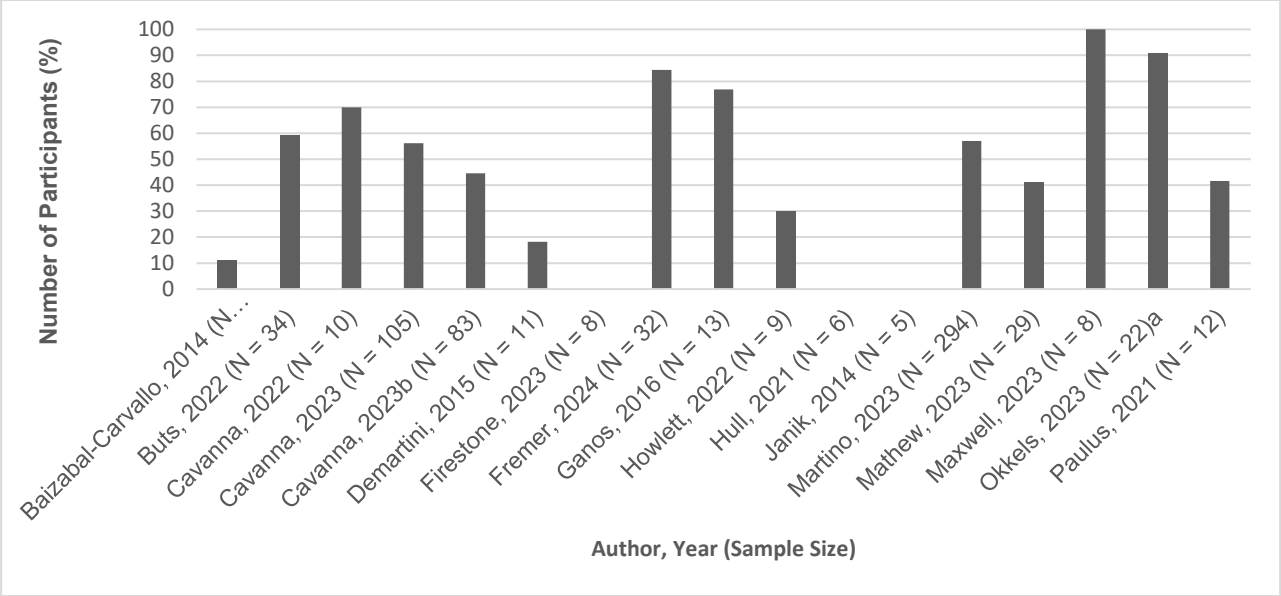
Mixed findings were reported for participants' ability to suppress FTLB as prevalence rates varied (Fig. 3) across *k* = 17 studies [17, 25, 34-39, 41-44, 48, 53, 55, 57, 58, 62, 63]. In *k* = 1 medium quality study [45] 70.0% of participants with Tourette syndrome and comorbid FTLB were able to suppress tics, but as the researchers did not specify whether this ability was for Tourette syndrome or FTLB, it was not grouped with the other studies. Similar to premonitory urges, participants in *k* = 1 high quality study [36, 37] reported being able to suppress FTLB for an average of 71 minutes, ranging from 1 second to 8 hours, and *k* = 1 study [44] of medium

quality found all participants reported improved suppressibility following a course of i-CBiT, with the 87.5% reporting FTLB were suppressible for at least one hour.



[a] This study had a total sample size of *N* = 28 participants, but only *n* = 17 were asked about premonitory urges.

Fig. 2 Rates of premonitory urges in participants with FTLB



[a] This study had a total sample size of *N* = 28, but only *n* = 22 participants were asked whether FTLB were suppressible.

Fig. 3 Percentage of participants reporting suppressibility of FTLB

1.4.17.1 Prevalence of Premonitory Urges and Suppressibility in FTLB Compared to CTDs

Evidence from *k* = 1 [61] high and *k* = 1 [34] medium quality study suggests premonitory urges are less frequent in participants with FTLB compared to CTDs, however this is contradicted by *k* = 1 high quality study [64] which found no differences in PUTS scores between participants with either diagnosis. Alternatively, *k* = 3 studies [34, 35, 45] ranging from high to medium quality investigated suppressibility in FTLB found participants with this diagnosis were less likely to suppress tics compared to participants with CTDs.

1.4.18 Interventions

Participants reported receiving pharmaceutical medication to treat FTLB (Table 5) in the majority of studies (*k* = 20) [17, 25, 31-35, 38-43, 47, 48, 50, 54-57, 59, 60, 63]. Studies with mixed aged samples (*k* = 7) [31, 32, 34, 39-41, 43, 46, 48], child only samples (*k* = 3) [54, 55, 60], and adult only samples (*k* = 1) [63] reported prescribing common anti-tic medications such as risperidone and aripiprazole as well as anti-anxiety, anti-depressant, and anti-psychotic medications in all aged samples across a variety of high and medium quality studies. Pharmacotherapy was found to have a positive effect for participants in *k* = 3 [48, 55, 60] studies of mixed aged and child only samples. However, *k* = 3 studies [17, 38, 63] including adults only and mixed aged samples reported none of the participants experienced symptom improvement following medication.

Participants also reported receiving psychological interventions in the majority of studies including cognitive behavioural therapy (CBT) (*k* = 7) [34, 39-41, 47, 57, 59, 60], comprehensive behavioural intervention for tics (CBIT) or i-CBIT (*k* = 5) [39-41, 44, 57, 58, 60], FND or stress management (*k* = 2) [54, 63], group psychoeducation (*k* = 1) [51], and unspecified psychotherapy (*k* = 6) [17, 33, 35-37, 42, 56]. Between 40.6% to 100% of participants across multiple studies ranging from high to medium quality (*k* = 5) [36, 37, 44, 58, 59, 63] found symptoms improved following psychological therapy. A minority of participants in high and medium quality studies (*k* = 2) [31, 32, 38] reported using cannabinoids to treat FTLB in both child and adult populations, however only *k* = 1 study [38] reported the effect of this treatment. Moreover, *k* = 2 studies of high and medium quality [36, 37, 43] including both children and adults found FTLB improved without treatment, however a high number of participants were found to relapse following remission, and *k* = 1 study [44] of medium quality evidence found psychological intervention led to worsened mood or FNS in 50.0% of participants in a mixed aged sample.

Table 5 Overview of interventions prescribed to treat FTLB and associated outcomes

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Baizabal-Carvallo, 2014 [63]	***N*** = 9 **Dx:** PT **Population:** Adults	***n:*** 3 (33.3%) **Types:** 1 levetiracetam, 1 risperidone, 1 tetrabenazine	***n:*** 7 (77.8%) **Types:** Psychotherapy and stress management	• No observed benefit from pharmacological intervention • *n* = 4 participants (57.1%) improved with psychological treatment
Berg, 2024 [31, 32]	***N*** = 35 **Dx:** FTLB **Population:** Children and adults	***n:*** 26 (74.3%) **Types:** 18 SSRIs, 6 antidepressants, 6 antipsychotics, 3 alpha-agonists, 2 stimulants, 2 cannabinoids, 1 lorazepam, and 1 gabapentin	***n:*** NR **Types:** n/a	NR
Buts, 2022 [50]	***N*** = 34 **Dx:** FTLB **Population:** Children	***n:*** 15 (44.1%) **Types:** NR	***n:*** NR **Types:** n/a	NR

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Cavanna, 2022 [25]	***N*** = 10 **Dx:** TS + FT **Population:** Children and adults	***n:*** 4 (40.0%) **Types:** 2 SSRIs, 1 second generation anti-dopaminergic agents, 1alpha 2 agonists, 1 beta blockers	***n:*** NR **Types:** n/a	NR
Cavanna, 2023 [33]	***N*** = 66 **Dx:** FT **Population:** Children and adults	***n:*** 32 (48.5%) **Types:** NR	***n:*** 25 (37.9%) **Types:** Psychotherapy	NR
Cavanna, 2023 [34]	***N*** = 105 **Dx:** FT **Population:** Children and adults	***n:*** 58 (55.2%) **Types:** 21% anti-dopaminergic agents, 11% benzodiazepines, 7% alpha-2 agonists, 6% serotonergic agents, 5% pregabalin, 4% beta blockers, 9% other	***n:*** 41 (39.0%) **Types:** Psychotherapy using CBT for anxiety and affective disorders	NR

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Cavanna, 2023 [35]	***N*** = 83 **Dx:** FT **Population:** Children and adults	***n:*** 47 (56.6%) **Types:** NR	***n:*** 32 (38.6%) **Types:** Psychotherapy	NR
Demartini, 2015 [17]:	***N*** = 11 **Dx:** FT **Population:** Children and adults	***n:*** 2(18.2%) **Types:** NR	***n:*** 0 (0%) **Types:** n/a	• No observed benefit from medications
Duncan, 2024 [51]	***N*** = 58 **Dx:** FTLB **Population:** Children	***n:*** NR **Types:** n/a	***n:*** 58 (100%) **Types:** One-off 2.5hr group focused on psychoeducation and CBT strategies	• Goal based outcomes improved from pre-group (*M* = 4.02, SD = 2.1) to post-group (*M* = 6.53, SD = 1.7)

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Fremer, 2024 [36, 37]	***N*** = 32 **Dx:** MSMI-FTLB **Population:** Children and adults	***n:*** NR **Types:** n/a	***n:*** 9 (28.1%) **Types:** psychotherapy, 4 (12.5%) unspecified treatment	• 40.6% improved with psychological or unspecified treatment • 12.5% improved without treatment • 12.5% improved following diagnosis
Ganos, 2016 [38]	***N*** = 13 **Dx:** Functional tic-like vocalisations **Population:** Children and adults	***n:*** 10 (76.9%) **Types:** NR	***n:*** NR **Types:** n/a	• No symptom improvement in 100% of participants using pharmacotherapy • 23.1% reported cannabis led to symptom improvement

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Han, 2022 [54]	***N*** = 22 **Dx:** FTLB **Population:** Children	***n:*** 17 (77.3%) **Types:** Alpha 2 agonists, antidepressants, antipsychotics	***n:*** 17 (77.3%) **Types:** FND management	• At last follow up, 68.2% participants had persistent symptoms • 18.2% reported partial improvements • 13.6% complete resolution of FTLB • Does not specify whether improved due to pharmacotherapy or psychological therapy

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Howlett, 2022 [39-41]	***N*** = 29 **Dx:** FTLB **Population:** Children (*n*=20) and adults (*n*=9)	***n:*** Minimum of 7 out of 15 adolescents (46.7%) and minimum of 3 out of 9 adults (33.3%) **Types:** Alpha-agonists, antipsychotics, SSRIs, non-SSRIs, psychostimulants	***n:*** 11 of 15 (73.3% adolescents, 6 (66.7%) adults **Types:** 53% adolescents and 44% adults received CBT for anxiety and depression, 20% adolescents and 44% adults received CBIT	NR
Hull, 2021 [55]	***N*** = 6 **Dx:** Abrupt onset tic-like movements **Population:** Children	***n:*** 4 (66.7%) **Types:** guanfacine, fluphenazine, diazepam, pimozide, clonidine	***n:*** NR **Types:** n/a	• 50.0% of participants receiving pharmacotherapy reported symptom improvement
Janik, 2014 [42]	***N*** = 5 **Dx:** PT **Population:** Children and adults	***n:*** 5 (100%) **Types:** NR	***n:*** 4 (80.0%) **Types:** Psychotherapy	NR

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Larsh, 2022 [56]	***N*** = 89 **Dx:** FTLB **Population:** Children	***n:*** 36 (41.9%) **Types:** NR	***n:*** 54 (62.1%) **Types:** NR	NR
Martino, 2023 [43]	***N*** = 294 **Dx:** FTLB **Population:** Children and adults	***n:*** 122 (41.5%) **Types:** clonidine, aripiprazole, guanfacine, risperidone	***n:*** NR **Types:** n/a	• No clinical benefit was seen in 73-89% of participants receiving pharmacotherapy • 60 (20.4%) made spontaneous recovery, but 38 (63.3%) relapsed
Mathew, 2023 [57]	***N*** = 29 **Dx:** FT **Population:** Children	***n:*** 9 (31.0%) **Types:** NR	***n:*** 15 (51.7%) **Types:** 34.5% CBT, 17.2% CBIT	• 82.8% of participants reported improvement since diagnosis • 10.3% reported no improvement • Does not specify whether improved due to pharmacotherapy or psychological therapy

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Maxwell, 2023 [44]	***N*** = 8 **Dx:** FTLB **Population:** Children and adults	***n:*** NR **Types:** n/a	***n:*** 8 (100%) **Types:** Individual i-CBiT focusing on ACT and CBT strategies	• 100% of participants experienced improved tic frequency, suppressibility, and severity • 50.0% tic free post treatment • 50.0% experienced worsened mood or FNS following treatment • School attendance improved in 100% of those affected *(n = 4)*

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Nilles, 2024 [47]	***N*** = 83 **Dx:** FTLB **Population:** Children and adults	***n:*** Minimum of 18 (21.7%) **Types:** alpha agonists, psychostimulants, antipsychotics, SSRIs, trazadone	***n:*** 38 of 57 (66.6%) **Types:** CBT for anxiety and depression Findings: n/a	• Rates of alpha agonists, psychostimulants, and antipsychotics reduced at follow up • Rates of SSRIs and trazadone increased at follow up
Okkels, 2023 [58]	***N*** = 28 **Dx:** FT **Population:** Children	***n:*** NR **Types:** n/a	***n:*** 28 (100%) **Types:** CBIT and psychoeducation	• 100% of participants reported a reduction in tic severity or disappearance of tics following psychological treatment
Paulus, 2021 [48]	***N*** = 13 **Dx:** TLB-SM **Population:** Children and adults	***n:*** 6 (46.2%) **Types:** antipsychotics	***n:*** NR **Types:** n/a	• 33.3% of participants receiving pharmacotherapy reported symptom improvement

Authors (Year, Country)	Study Design	Pharmacotherapy	Psychological Therapy	Findings
Prato, 2023 [59]	***N*** = 11 **Dx:** FTLB **Population:** Children	***n:*** 8 (72.7%) **Types:** n/a	***n:*** 7 (63.6%) **Types:** CBT	NR
Tomczak, 2024 [60]	***N*** = 56 **Dx:** FTLB **Population:** Children	***n:*** 55 (98.2%) **Types:** SSRIs, anti-anxiety, anti-depressants, antipsychotics, alpha agonists	***n:*** 48 (85.7%) **Types:** 39.6% CBIT, 60.4% psychological therapy without CBIT	• 78.9% of participants receiving CBIT improved at follow up • 79.3% receiving psychological therapy without CBIT improved at follow up • 72.7% receiving pharmacotherapy improved at follow up

Abbreviations: FTLB functional tic-like behaviours, FTD functional tic disorder, FT functional tics, MSMI-FTLB mass social media-induced illness functional tic-like behaviours, PT psychogenic tics, TLB-SM tic like behaviours following social media consumption, pps participants, NR not reported, SSRIs selective serotonin reuptake inhibitors, CBT cognitive behavioural therapy, CBIT comprehensive behavioural intervention for tics, i-CBiT integrated-cognitive behavioural intervention for functional tics

1.5 Discussion

1.5.1 Summary of Evidence

This review aimed to systematically evaluate the literature and identify the prevalence of FTLB and the associated features of this phenomenon in line with proposed diagnostic criteria and compared them to features of CTDs. Overall, *k* = 33 studies [17, 25, 30-64] investigating patients with FTLB were included and reported findings on the prevalence, associated comorbidities, clinical features, expression of symptoms, or common interventions used to treat FTLB. Table 6 summarises the key clinical features of FTLB compared to CTDs based on the evidence identified within this review.

Narrative synthesis showed that the majority of studies reported an assigned-female sex predominance of FTLB which contrasts the typical 3-4:1 assigned-male to assigned-female sex seen within CTDs [8]. Whilst this suggests assigned-female sex is a common feature of FTLB, the studies [17, 36-38, 42, 45, 62] reporting lower or equal rates of FTLB in the assigned-male sex suggests sex cannot be a sole indicator of FTLB [16]. Synthesis also highlighted the prevalence of FTLB among individuals identifying as transgender, non-binary, or gender diverse. However, few studies reported the gender identity of participants, making it difficult to assess overall prevalence within this population. The finding that gender identity was reported in more studies including children [31, 32, 49, 51, 52, 57, 60] and both children and adults [36, 37, 39-41, 44, 47] compared to adult only [64] samples may echo the recent surge in young people identifying as transgender, non-binary, or gender diverse. However, it could also reflect later generations increased acceptance towards gender minorities and thus may find it easier to describe their identity [69]. A recent review found high rates of transgender individuals in FND, indicating a possible link between non-cisgender individuals and manifestation of functional illnesses [70], however comparing rates of gender minorities in individuals with FTLB and CTDs is difficult as most research into CTDs fail to report or investigate gender identity, meaning the rates of gender minorities in this population are unknown.

Table 6 Summary of the key differences in clinical features of FTLB compared to CTDs, ordered by features with the most evidence and quality ratings

FTLB Patients Compared to CTD Patients	Quality Ratings of Studies	References
More likely to be of assigned female sex[a]	High - Medium	30-32, 36, 37, 29-41, 45, 46, 54, 56, 59, 61-64
Older age symptom onset	High - Medium	30-32, 35-37, 39, 45, 54, 59, 61, 62, 64
Increased likelihood of coprophenomena, self-injurious, blocking and throwing tics	High - Medium	30, 35, 45, 46, 54, 61
Tics are more likely to be intermittent and severe	High - Medium	34, 39, 41
Less likely to be able to suppress tics	High - Medium	34, 35, 45
More likely to be gender diverse	High	31, 32
More likely to have acute/rapid onset of symptoms	High - Medium	35, 45
Higher incidence of comorbid FNS	High	35, 61
Onset and symptoms more likely to have psychological or tic-contingent trigger	High	30

[a] *k* = 1 medium quality study found higher male incidence in FTLB compared to CTDs

The studies included in this review found onset of FTLB was significantly later in comparison to CTDs [30-32, 35-37, 39, 45, 54, 59, 61, 62, 64] suggesting that FTLB may follow a different trajectory to CTDs, and indicates symptom onset at 12 years old or later is a common feature of FTLB [16]. It has been widely documented that mental health difficulties increased in COVID-19 [71] and multiple studies have reported surges in the number of individuals presenting with FND, including FTLB, during COVID-19 [20, 72]. This trend was reflected in the

studies included in this review as most participants received a diagnosis of FTLB during the pandemic [25, 31-35, 39-41, 44, 46-50, 52-55, 57-61] and COVID-19 was identified as a precipitating factor in several studies [43, 58-60], suggesting COVID-19 may be a causal factor in onset of FTLB. Moreover, in comparison to CTDs which are classed as neurodevelopmental, studies included in this review suggest onset of FTLB is linked to psychosocial triggers [17, 30, 33-35, 38, 43, 45, 53, 58-60], such as stress and anxiety (including pandemic related stressors) and social media consumption. This indicates a possible difference in the development of FTLB compared to CTDs which may aid differential diagnosis. However, not all participants recalled a psychosocial trigger prior to onset of FTLB suggesting this cannot be the sole cause of the phenomenon. Instead, a biopsychosocial framework has been proposed as a way of understanding the interaction between neurobiological, psychological, and social factors in increasing susceptibility to FTLB [73]. Studies within this review highlighted the prevalence of co-occurring or pre-existing CTDs in participants with FTLB [25, 35, 43-45, 49, 53, 54, 58-60, 64], suggesting a possible functional overlay between the two, and indicating a history of CTDs may be a causal factor in the onset of FTLB.

Despite FTLB typically being associated with rapid symptom onset, the studies in this review indicate this feature varies between individuals. However, the number of studies investigating this characteristic were limited and this may be because participants were diagnosed with FTLB prior to this being suggested as a diagnostic criterion [16]. Thus, it may be that the rates of acute onset are higher in individuals with FTLB but are not consistently being recorded by clinicians, highlighting the impact that measurement bias can have on studies. CTDs are known for having a gradual onset of tics, and studies in this review found onset was more rapid in FTLB when compared to CTDs [35, 45], suggesting this is a possible difference between the two diagnoses.

Overall, narrative synthesis highlighted children with FTLB had higher rates of comorbid anxiety, depression, OCD, ADHD, and ASD compared to studies including older participants. Whilst this indicates age may be a factor in prevalence of comorbidities in FTLB, it may instead reflect the high rates of mental health difficulties seen within young people in the general population, particularly since the COVID-19 pandemic [74]. Despite co-occurring anxiety and depressive disorders being classed as a minor diagnostic criteria for FTLB [16], studies in this review reported varying rates of these disorders in participants and mixed evidence was found when comparing rates to participants with CTDs. This suggests prevalence of co-occurring anxiety and depression may not be specific to those with FTLB, particularly as rates of these disorders are frequently reported in individuals with CTDs [8, 15]. Similarly, rates of OCD, ADHD, and ASD found in included studies varied, with some studies finding a higher prevalence in participants with FTLB, whereas others found no differences. This indicates individual

differences in comorbidities may exist between FTLB and CTDs and are not specific to either diagnosis. Alternatively, co-occurring FNS are considered to be extremely prevalent in individuals with FTLB and is a minor criterion for diagnosing this disorder [16]. The studies in this review found high rates of FND/FNS in participants with FTLB, and these were higher in comparison to CTDs, suggesting co-occurring of FND/FNS is a common feature of FTLB.

Complex tics such as coprophenomena, self-injurious behaviours, and throwing objects have been associated with FTLB, as these tics rarely occur in CTDs [75]. Narrative synthesis identified coprophenomena was common in participants with FTLB and a higher incidence was found in comparison to CTDs, suggesting presence of coprophenomena may indicate greater likelihood of FTLB diagnosis. Similarly, studies in this review found a high prevalence of self-injurious tics in participants with FTLB, particularly in comparison to CTDs, suggesting symptom severity may be higher in the former diagnosis. Studies also reported higher incidences of requiring medical help as a result of tics, although this was only reported in studies including child samples, indicating (parents with) children may be more likely to seek medical advice or have greater symptom severity compared to adults with FTLB. Whilst studies in this review found participants with FTLB experienced a range of complex tics, findings were mixed as to whether rates of simple or complex tics were higher in FTLB or CTDs [30, 35, 54, 62]. This suggests that types of tics observed in FTLB may not be a sole diagnostic indicator and should be considered in conjunction with a range of other criteria [76].

Narrative synthesis highlighted impairment from FTLB varies between participants over time, and this appeared similar to participants with CTDs, suggesting this is not a sole indicator of diagnostic certainty. Premonitory urges are frequently reported in individuals with CTDs [77]. However, the studies in this review indicated the presence of premonitory urges varied between participants with FTLB, but similar frequency and impairment ratings on the PUTS were found in child and adult studies, indicating no age differences in this characteristic [39, 41, 58]. Suppressibility also varied across participants within the included studies, however this ability was reported less frequently in comparison to CTDs, suggesting it is not a consistent feature of FTLB [39, 41, 58].

Pharmacotherapy is a common intervention for CTDs and individuals can be prescribed antidopaminergic medications to help reduce tic-related symptoms or anti-anxiety medications to target comorbid conditions that may interfere with tics [78]. Previous research has indicated pharmacotherapy for tics (particularly antidopaminergic medication) is not suitable for treating patients with FTLB [79]. Whilst this review highlighted individuals with FTLB are prescribed similar pharmacological medications to individuals with CTDs, the findings were mixed, indicating effects may vary on an individual basis. Similarly, psychological interventions were

found to have varying response rates, but studies indicated this may be a useful treatment for patients with FTLB. One study [51] evaluated the effectiveness of a virtual group delivering psychoeducation and CBT techniques to children with FTLB and found participants felt closer to achieving their goals following treatment, suggesting it may be a useful intervention. This echoes recent research which highlights the benefits of psychoeducation in improving understanding and symptom reduction [76, 80]. Similarly, a case series [44] highlighting the benefits of individual i-CBiT, which included strategies from acceptance and commitment therapy, found all participants had noticeable improvement scores on the YGTSS for impairment and severity. Although this was conducted on a small sample size and cannot be generalised to the wider population of people with FTLB, it indicates promising developments in effective interventions for this disorder. Furthermore, research suggests early diagnosis is crucial in promoting recovery of FND [80], and this was reflected in two studies [36, 37, 43] in which participants were found to recover following assessment with no active treatment.

1.5.2 Critique of the Studies

Most studies included in this review had a small sample size or did not provide power calculations, meaning adequate sample size for statistical analysis could not be determined. This suggests studies with significant results may be underpowered and should be interpreted with caution, and findings reported in these studies may not be generalisable to the wider population. Moreover, all studies demonstrated selection bias when recruiting their samples, and thus participants may not be representative of all individuals with FTLB, as those in studies may present with more severe symptoms or be a specific subtype. Similarly, few studies reported the ethnicity of participants, thus it is unclear whether samples represent participants from a variety of ethnicities, again indicating the findings cannot be generalised to others and may be unrepresentative.

Only $k = 1$ study [39-41] used validated screening tools to diagnose participants with comorbid psychiatric disorders, whereas the remaining studies relied upon medical records, self-report, or clinical examination. It is possible researchers and participants may have been subject to recall bias which may have impacted the reporting of comorbidities in studies, indicating they could be under or over reported. Moreover, the lack of detailed reporting of methods used to gather outcomes in the majority of studies impacts the reliability of results as they may not be reproducible, reducing the external validity of the findings.

1.5.3 Critique of the Review

This review is the first to systematically examine the literature on FTLB and includes all associated terminology to describe this diagnosis. A large number of studies were included, including studies in which FTLB was diagnosed prior to COVID-19 to ensure synthesis reflected the overall phenomenological picture of this disorder. However, the studies included in this review varied in their design and the outcomes they reported, resulting in a meta-analysis not being conducted. This, alongside the highly descriptive nature of study findings, made grouping studies and narrative synthesis difficult.

1.5.4 Conclusion

This systematic review highlights that incidences of FTLB have been documented and described throughout the last eleven years despite the differing terminologies. It shows features such as higher age of onset and coprophenomena are associated with FTLB. However, this review also highlights types of tics, the presence of premonitory urges, suppressibility, and comorbid conditions may not be specific to FTLB and may be similar to rates observed in CTDs. Furthermore, the review shows common interventions for CTDs produced mixed results for patients with FTLB and suggests novel interventions may need to be developed. This review indicates further research is required, particularly on larger samples to allow results to be generalised. Future research should aim to document gender identity and ethnicity in people with FTLB and CTDs to allow statistical comparisons to be made. In addition, researchers should seek to conduct experimental designs, such as randomised control trials, to evaluate the impact of psychological interventions including psychoeducation and i-CBiT on people with FTLB.

1.6 References

1. American Psychiatric Association (2013) Somatic Sympton and Related Disorders. In: Diagnostic and Statistical Manual of Mental Disorders, 5th edn. https://psychiatryonline.org/doi/book/10.1176/appi.books.9780890425596
2. Kanaan RAA (2016) Freud's hysteria and its legacy. In: Hallett M, Stone J, Carson A (eds) Handbook of Clinical Neurology. Elsevier, pp 37-44. https://doi.org/10.1016/b978-0-12-801772-2.00004-7
3. Park JE (2024) Functional Movement Disorders: Updates and Clinical Overview. J Mov Disord 17:251-261. https://doi.org/10.14802/jmd.24126
4. Espay AJ, Aybek S, Carson A, Edwards MJ, Goldstein LH, Hallett M, LaFaver K, LaFrance WC, Jr., Lang AE, Nicholson T, Nielsen G, Reuber M, Voon V, Stone J, Morgante F (2018) Current Concepts in Diagnosis and Treatment of Functional Neurological Disorders. JAMA Neurol 75:1132-1141. https://doi.org/10.1001/jamaneurol.2018.1264
5. Ludwig L, Pasman JA, Nicholson T, Aybek S, David AS, Tuck S, Kanaan RA, Roelofs K, Carson A, Stone J (2018) Stressful life events and maltreatment in conversion (functional neurological) disorder: systematic review and meta-analysis of case-control studies. Lancet Psychiatry 5:307-320. https://doi.org/10.1016/s2215-0366(18)30051-8
6. American Psychiatric Association (2013) Neurodevelopmental disorders. In: Diagnostic and statistical manual of mental disorders, 5th edn. https://psychiatryonline.org/doi/book/10.1176/appi.books.9780890425596
7. Leckman JF, Peterson BS, Pauls DL, Cohen DJ (1997) Tic Disorders. In Rutter M, Taylor E, Hersov L (eds): Child and Adolescent Psychiatry: Modern Approaches, 3rd edn. London, Blackwell Scientific, pp 455–466. https://doi.org/10.1016/s0193-953x(05)70348-8
8. Freeman RD, Fast DK, Burd L, Kerbeshian J, Robertson MM, Sandor P (2000) An international perspective on Tourette syndrome: selected findings from 3,500 individuals in 22 countries. Dev Med Child Neurol 42:436-447. https://doi.org/10.1017/s0012162200000839
9. Ueda K, Black KJ (2021) A Comprehensive Review of Tic Disorders in Children. J Clinic Med 10:2479. https://www.mdpi.com/2077-0383/10/11/2479
10. Eapen V, Cavanna AE, Robertson MM (2016) Comorbidities, Social Impact, and Quality of Life in Tourette Syndrome. Front Psychiatr 7:97. https://doi.org/10.3389/fpsyt.2016.00097
11. Gill CE, Kompoliti K (2020) Clinical Features of Tourette Syndrome. J Child Neurol 35:166-174. https://doi.org/10.1177/0883073819877335

12. Leckman JF, Bloch MH, Scahill L, King RA (2006) Tourette syndrome: the self under siege. J Child Neurol 21:642-649. https://doi.org/10.1177/08830738060210081001
13. Banaschewski T, Woerner W, Rothenberger A (2003) Premonitory sensory phenomena and suppressibility of tics in Tourette syndrome: developmental aspects in children and adolescents. Dev Med Child Neurol 45:700-703. https://doi.org/10.1017/s0012162203001294
14. Brandt VC, Beck C, Sajin V, Baaske MK, Bäumer T, Beste C, Anders S, Münchau A (2016) Temporal relationship between premonitory urges and tics in Gilles de la Tourette syndrome. Cortex 77:24-37. https://doi.org/10.1016/j.cortex.2016.01.008https://www.sciencedirect.com/science/article/pii/S0010945216000186
15. Hirschtritt ME, Lee PC, Pauls DL, Dion Y, Grados MA, Illmann C, King RA, Sandor P, McMahon WM, Lyon GJ, Cath DC, Kurlan R, Robertson MM, Osiecki L, Scharf JM, Mathews CA (2015) Lifetime prevalence, age of risk, and genetic relationships of comorbid psychiatric disorders in Tourette syndrome. JAMA Psychiatry 72:325-333. https://doi.org/10.1001/jamapsychiatry.2014.2650
16. Pringsheim T, Ganos C, Nilles C, Cavanna AE, Gilbert DL, Greenberg E, Hartmann A, Hedderly T, Heyman I, Liang H, Malaty I, Malik O, Debes NM, Vahl KM, Munchau A, Murphy T, Nagy P, Owen T, Rizzo R, Skov L, Stern J, Szejko N, Worbe Y, Martino D (2023) European Society for the Study of Tourette Syndrome 2022 criteria for clinical diagnosis of functional tic-like behaviours: International consensus from experts in tic disorders. Eur J Neurol 30:902-910. https://doi.org/10.1111/ene.15672
17. Demartini B, Ricciardi L, Parees I, Ganos C, Bhatia KP, Edwards MJ (2015) A positive diagnosis of functional (psychogenic) tics. Eur J Neurol 22:527. https://doi.org/10.1111/ene.12609
18. Müller-Vahl KR, Pisarenko A, Jakubovski E, Fremer C (2022) Stop that! It's not Tourette's but a new type of mass sociogenic illness. Brain 145:476-480. https://doi.org/10.1093/brain/awab316
19. Robinson S, Hedderly T (2016) Novel Psychological Formulation and Treatment of "Tic Attacks" in Tourette Syndrome. Front Pediatr 4:46. https://doi.org/10.3389/fped.2016.00046
20. Heyman I, Liang H, Hedderly T (2021) COVID-19 related increase in childhood tics and tic-like attacks. Arch Dis Child 106:420-421. https://doi.org/10.1136/archdischild-2021-321748
21. Amorelli G, Martino D, Pringsheim T (2022) Rapid Onset Functional Tic-Like Disorder Outbreak: A Challenging Differential Diagnosis in the COVID-19 Pandemic. J Can Acad Child Adolesc Psychiatr 31:144-181.

22. Owen T, Silva J, Grose C, Bailey A, Robinson S, Anderson S, Ludlow A, Sharp S, Toghill L, Hedderly T (2022) Case report: Advice for schools on managing functional tic-like behaviours. Front Psychiatry 13:1001459 https://doi.org/10.3389/fpsyt.2022.1001459
23. Rigas A, Mainka T, Pringsheim T, Münchau A, Malaty I, Worbe Y, Cavanna AE, Lees AJ, Lang AE, Martino D, Ganos C (2023) Distinguishing functional from primary tics: a study of expert video assessments. J Neurol Neurosurg Psychiatry 94:751-756. https://doi.org/10.1136/jnnp-2022-330822
24. Ganos C, Martino D, Espay AJ, Lang AE, Bhatia KP, Edwards MJ (2019) Tics and functional tic-like movements: Can we tell them apart? Neurol 93:750-758. https://doi.org/10.1212/wnl.0000000000008372
25. Cavanna AE, Damodaran L, Purpura G, Nacinovich R (2022) Tourette Syndrome with Functional Overlay: A Case Series. Arch Med Health Sciclin 10:312-316. DOI*:* 10.4103/amhs.amhs_247_22
26. Andersen K, Cavanna AE, Szejko N, Müller-Vahl KR, Hedderly T, Skov L, Mol Debes N (2024) A Critical Examination of the Clinical Diagnosis of Functional Tic-like Behaviors. Mov Disord Clin Pract 11:1065-1071. https://doi.org/10.1002/mdc3.14150
27. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, Shamseer L, Tetzlaff JM, Akl EA, Brennan SE, Chou R, Glanville J, Grimshaw JM, Hróbjartsson A, Lalu MM, Li T, Loder EW, Mayo-Wilson E, McDonald S, McGuinness LA, Stewart LA, Thomas J, Tricco AC, Welch VA, Whiting P, Moher D (2021) The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 372:n71. https://doi.org/10.1136/bmj.n71
28. Kmet LM, Cook LS, Lee RC (2004) Standard quality assessment criteria for evaluating primary research papers from a variety of fields. HTA Initiatve.
29. Mayo-Wilson E, Li T, Fusco N, Dickersin K (2018) Practical guidance for using multiple data sources in systematic reviews and meta-analyses (with examples from the MUDS study). Res Synth Methods 9:2-12. https://doi.org/10.1002/jrsm.1277
30. Andersen K, Jensen I, Okkels KB, Skov L, Debes NM (2023) Clarifying the Differences between Patients with Organic Tics and Functional Tic-Like Behaviors. Healthc 11:1489 https://doi.org/10.3390/healthcare11101481
31. Berg L, Martino D, L'Erario ZP, Pringsheim T (2024) Symptom Severity and Health Impacts of Functional Tic-Like Behaviors in Youth. Pediatr Neurol 155:68-75. https://doi.org/10.1016/j.pediatrneurol.2024.03.016
32. Berg L, Pringsheim TM, Lerario M, Martino D (2024) Psychological Factors Associated with Functional Tic-like Behaviours during the COVID-19 Pandemic. Res Child Adolesc Psychopathol 52:1157-1172. https://doi.org/10.1007/s10802-024-01184-y

33. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) Functional tics: Expanding the phenotypes of functional movement disorders? Eur J Neurol 30:3353-3356. https://doi.org/10.1111/ene.15967
34. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) New-onset functional tics during the COVID-19 pandemic: Clinical characteristics of 105 cases from a single centre. Eur J Neurol 30:2411-2417. https://doi.org/10.1111/ene.15867
35. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) Neurodevelopmental versus functional tics: A controlled study. J Neurol Sci 451:120725. https://doi.org/10.1016/j.jns.2023.120725
36. Fremer C, Szejko N, Pisarenko A, Haas M, Laudenbach L, Wegener C, Müller-Vahl KR (2022) Mass social media-induced illness presenting with Tourette-like behavior. Front Psychiatry 13:963769. https://doi.org/10.3389/fpsyt.2022.963769
37. Fremer C, Szejko N, Pisarenko A, Haas M, Laudenbach L, Wegener C, Müller-Vahl KR (2024) What distinguishes patients with mass social media-induced illness presenting with Tourette-like behavior from those with Tourette syndrome? Results of a prospective cohort study. Eur Arch Psychiatry Clinic Neurosci 274:227-233. https://doi.org/10.1007/s00406-023-01603-z
38. Ganos C, Edwards MJ, Müller-Vahl K (2016) "I swear it is Tourette's!": On functional coprolalia and other tic-like vocalizations. Psychiatry Res 246:821-826. https://doi.org/10.1016/j.psychres.2016.10.021
39. Howlett M, Martino D, Nilles C, Pringsheim T (2022) Prognosis of rapid onset functional tic-like behaviors: Prospective follow-up over 6 months. Brain Behav 12:2606. https://doi.org/10.1002/brb3.2606
40. Pringsheim T, Ganos C, McGuire JF, Hedderly T, Woods D, Gilbert DL, Piacentini J, Dale RC, Martino D (2021) Rapid Onset Functional Tic-Like Behaviors in Young Females During the COVID-19 Pandemic. Mov Disord 36:2707-2713. https://doi.org/10.1002/mds.28778
41. Pringsheim T, Martino D (2021) Rapid onset of functional tic-like behaviours in young adults during the COVID-19 pandemic. Eur J Neurol 28:3805-3808. https://doi.org/10.1111/ene.15034
42. Janik P, Milanowski Ł, Szejko N (2014) Psychogenic tics: Clinical characteristics and prevalence. Psychiatria Polska 48:835-845.
43. Martino D, Hedderly T, Murphy T, Müller-Vahl KR, Dale RC, Gilbert DL, Rizzo R, Hartmann A, Nagy P, Anheim M, Owen T, Malik O, Duncan M, Heyman I, Liang H, McWilliams A, O'Dwyer S, Fremer C, Szejko N, Han VX, Kozlowska K, Pringsheim TM (2023) The spectrum of functional tic-like behaviours: Data from an international registry. Eur J Neurol 30:334-343. https://doi.org/10.1111/ene.15611

44. Maxwell A, Zouki J-J, Eapen V (2023) Integrated cognitive behavioral intervention for functional tics (I-CBiT): case reports and treatment formulation. Front Pediatr 11:1265123. https://doi.org/10.3389/fped.2023.1265123
45. Müller-Vahl KR, Pisarenko A, Fremer C, Haas M, Jakubovski E, Szejko N (2024) Functional Tic-Like Behaviors: A Common Comorbidity in Patients with Tourette Syndrome. Mov Disord Clinic Pract 11:227-237. https://doi.org/10.1002/mdc3.13932
46. Nilles C, Martino D, Berg L, Fletcher J, Pringsheim T (2024) What are the Key Phenomenological Clues to Diagnose Functional Tic-Like Behaviors in the Pandemic Era? Mov Disord Clinic Pract 11:398-402. https://doi.org/10.1002/mdc3.13977
47. Nilles C, Szejko N, Martino D, Pringsheim T (2024) Prospective follow-up study of youth and adults with onset of functional tic-like behaviours during the COVID-19 pandemic. Eur J Neurol 31:e16051. https://doi.org/10.1111/ene.16051
48. Paulus T, Bäumer T, Verrel J, Weissbach A, Roessner V, Beste C, Münchau A (2021) Pandemic Tic-like Behaviors Following Social Media Consumption. Mov Disord 36:2932-2935. https://doi.org/10.1002/mds.28800
49. Ducroizet A, Eccles C, Lancaster R, Kowalczyk A, Owen T, Sopena S, Malik O, Hedderly T (2025) Outcomes of functional tics in adolescents: a single-centre tertiary study. Arch Dis Child. https://doi.org/10.1136/archdischild-2024-327408
50. Armstrong-Javors A, Realbuto E, Dy-Hollins ME, Scharf JM (2024) Increase in Functional Tic Presentations in Sexual Orientation and Gender Identity Minority Youth During Coronavirus Disease 2019. Pediatr Neurol 155:182-186. https://doi.org/10.1016/j.pediatrneurol.2024.03.027
51. Buts S, Duncan M, Owen T, Martino D, Pringsheim T, Byrne S, McWilliams A, Murphy T, Malik O, Liang H, Heyman I, Hedderly T (2022) Paediatric tic-like presentations during the COVID-19 pandemic. Arch Dis Child 107:17. https://doi.org/10.1136/archdischild-2021-323002
52. Duncan M, Pearman Z, Harrold K, Warren A, Evans S, McAllister E, Heyman I, Shavel-Jessop S, Murphy T, Liang H (2024) Evaluation of a psychoeducation group for children presenting with functional tic-like behaviours. Clinic Child Psychol Psychiatry 29:1011-1025. https://doi.org/10.1177/13591045241237829
53. Firestone MJ, Holzbauer S, Conelea C, Danila R, Smith K, Bitsko RH, Klammer SM, Gingerich S, Lynfield R (2023) Rapid onset of functional tic-like behaviors among adolescent girls-Minnesota, September-November 2021. Front Neurol 13:1063261. https://doi.org/10.3389/fneur.2022.1063261
54. Han VX, Kozlowska K, Kothur K, Lorentzos M, Wong WK, Mohammad SS, Savage B, Chudleigh C, Dale RC (2022) Rapid onset functional tic-like behaviours in children and adolescents during COVID-19: Clinical features, assessment and biopsychosocial

treatment approach. J Paediatr Child Health 58:1181-1187. https://doi.org/10.1111/jpc.15932

55. Hull M, Parnes M (2021) Tics and TikTok: Functional Tics Spread Through Social Media. Movement disorders clinical practice 8:1248-1252. https://doi.org/10.1002/mdc3.13267
56. Larsh TR, Wu SW, Gilbert DL (2022) Comparison of Impairment in Functional Tic Disorders Versus Tourette Syndrome. Pediatr Neurol 134:83-84. https://doi.org/10.1016/j.pediatrneurol.2022.06.013
57. Mathew A, Abu Libdeh A, Patrie J, Garris J (2023) Outcome in Pediatric Functional Tic Disorders Diagnosed During the COVID-19 Pandemic. J Neuropsychiatr Clinic Neurosci 35:393-397. https://doi.org/10.1176/appi.neuropsych.20220186
58. Okkels KB, Skov L, Klansø S, Aaslet L, Grejsen J, Reenberg A, Sørensen CB, Debes NMMM (2023) Increased Number of Functional Tics Seen in Danish Adolescents during the COVID-19 Pandemic. Neuropediatr 54:113-119. https://doi.org/10.1055/a-1985-6862
59. Prato A, Saia F, Milana MC, Scerbo M, Barone R, Rizzo R (2023) Functional tic-like behaviours during the COVID-19 pandemic: Follow-up over 12 months. Front Pediatr 10:1003825. https://doi.org/10.3389/fped.2022.1003825
60. Tomczak KK, Worhach J, Rich M, Swearingen Ludolph O, Eppling S, Sideridis G, Katz TC (2024) Time is ticking for TikTok tics: A retrospective follow-up study in the post-COVID-19 isolation era. Brain Behav 14:3451. https://doi.org/10.1002/brb3.3451
61. Trau SP, Quehl L, Tsujimoto THM, Lin F-C, Singer HS (2022) Creating a Patient-Based Diagnostic Checklist for Functional Tics During the COVID-19 Pandemic. Neurol Clinic Pract 12:365-376. http://dx.doi.org/10.1212/CPJ.0000000000200067
62. Baizabal-Carvallo JF, Alonso-Juarez M, Jankovic J (2023) Contrasting features between Tourette syndrome and secondary tic disorders. J Neural Transmi (Vienna, Austria : 1996) 130:931-936. https://doi.org/10.1007/s00702-023-02642-5
63. Baizabal-Carvallo JF, Jankovic J (2014) The clinical features of psychogenic movement disorders resembling tics. J Neurol, Neurosurg & Psychiatry 85:573-575. https://doi.org/10.1136/jnnp-2013-305594
64. Szejko N, Fletcher J, Martino D, Pringsheim T (2024) Premonitory Urge in Patients with Tics and Functional Tic-like Behaviors. Mov Disord Clinic Pract 11:276-281. https://doi.org/10.1002/mdc3.13951
65. Fahn S, Williams DT (1988) Psychogenic dystonia. Adv Neurol 50:431-455.
66. Robertson MM, Eapen V (1996) The National Hospital Interview Schedule for the assessment of Gilles de la Tourette Syndrome. Int J Methods Psychiatr Res 6:203-226. https://psycnet.apa.org/doi/10.1002/(SICI)1234-988X(199612)6:4%3C203::AID-MPR165%3E3.3.CO;2-F

67. Shaffer D, Gould MS, Brasic J, Ambrosini P, Fisher P, Bird H, Aluwahlia S (1983) A children's global assessment scale (CGAS). Arch Gen Psychiatr 40:1228-1231. https://doi.org/10.1001/archpsyc.1983.01790100074010
68. Leckman JF, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, Cohen DJ (1989) The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity. J Am Acad Child Adolesc Psychiatr 28:566-573. https://doi.org/10.1097/00004583-198907000-00015
69. Graham P (2023) Transgender children and young people: how the evidence can point the way forward. BJPsych Bull 47:98-104. https://doi.org/10.1192/bjb.2022.3
70. Lerario MP, Fusunyan M, Stave CD, Roldán V, Keuroghlian AS, Turban J, Perez DL, Maschi T, Rosendale N (2023) Functional neurological disorder and functional somatic syndromes among sexual and gender minority people: A scoping review. J Psychosom Res 174:111491. https://doi.org/10.1016/j.jpsychores.2023.111491
71. Vindegaard N, Benros ME (2020) COVID-19 pandemic and mental health consequences: Systematic review of the current evidence. Brain, Behav, & Immun 89:531-542. https://doi.org/10.1016/j.bbi.2020.05.048
72. Janssen-Aguilar R, Galíndez-de la Portilla JP, Gómez-Alcorta R, Hernández-Palestina M, Mendez-Dominguez N, Ruiz-Chow A, Crail-Meléndez D (2022) Impact of the COVID-19 pandemic on the incidence of patients with functional neurological disorder seen in a neurological emergency department. Psychiatr Clin Neurosci 76:595-596. https://doi.org/10.1111/pcn.13459
73. Berg L, Pringsheim T, Martino D (2022) Sudden Onset Tic and Tic-Like Presentations in Older Adolescents and Adults. Curr Dev Disord Rep 9:1-10. https://doi.org/10.1007/s40474-022-00263-3
74. Kauhanen L, Wan Mohd Yunus WMA, Lempinen L, Peltonen K, Gyllenberg D, Mishina K, Gilbert S, Bastola K, Brown JS, Sourander A (2023) A systematic review of the mental health changes of children and young people before and during the COVID-19 pandemic. Eur Child Adolesc Psychiatr 32:995-1013. https://doi.org/10.1007/s00787-022-02060-0
75. Cavanna AE, Spini L, Ferrari S, Purpura G, Riva A, Nacinovich R, Seri S (2024) Functional Tic-like Behaviors: From the COVID-19 Pandemic to the Post-Pandemic Era. Healthc (Basel) 12:1106 https://doi.org/10.3390/healthcare12111106
76. Malaty IA, Anderson S, Bennett SM, Budman CL, Coffey BJ, Coffman KA, Greenberg E, McGuire JF, Müller-Vahl KR, Okun MS, Quezada J, Robichaux-Viehoever A, Black KJ (2022) Diagnosis and Management of Functional Tic-Like Phenomena. J Clin Med 11:6470 https://doi.org/10.3390/jcm11216470

77. Brandt V, Essing J, Jakubovski E, Müller-Vahl K (2023) Premonitory Urge and Tic Severity, Comorbidities, and Quality of Life in Chronic Tic Disorders. Mov Disord Clinic Pract 10:922-932. https://doi.org/10.1002/mdc3.13742

78. Srour M, Lespérance P, Richer F, Chouinard S (2008) Psychopharmacology of tic disorders. J Can Acad Child Adolesc Psychiatr 17:150-159.

79. Nilles C, Pringsheim TM, Martino D (2022) The recent surge of functional movement disorders: social distress or greater awareness? Curr Opin Neurol 35:485-493. https://doi.org/10.1097/wco.0000000000001074

80. Vassilopoulos A, Mohammad S, Dure L, Kozlowska K, Fobian AD (2022) Treatment Approaches for Functional Neurological Disorders in Children. Curr Treat Options Neurol 24:77-97. https://doi.org/10.1007/s11940-022-00708-5

Chapter 2 Empirical Study

The Association Between Interoception, Tics, Anxiety, and Quality of Life in Young People with Chronic Tic Disorders and Functional Tic-Like Behaviours

Journal Specification: The following chapter has been prepared for submission to the Journal of Neurology. The guidelines for authors are shown in Appendix A. However, font style and size has not been followed and instead the standard university template font (Aptos) has been selected for ease of readability for the examiner.

Word Count: 7376

(excluding abstract, keywords, figures, tables, captions and references)

Acknowledgements: Tourettes Action Charity provided funding for this study. Also, the name of the author legally changed during the doctorate and thus their old name will be displayed on study materials presented in the appendices.

2.1 Abstract

Background: Adults with chronic tic disorders (CTDs) have been found to have atypical interoception. Few studies have investigated interoception in children with CTDs and findings are mixed. Limited studies have explored interoception in adults with functional movement disorders. No studies have investigated interoception in functional tic-like behaviours (FTLB). This study aims to assess and compare interoceptive processes across multiple bodily domains in young people with CTDs, FTLB, and controls. Group differences in attentional control, quality of life, tic-related factors, and comorbidities are explored, alongside associations with interoceptive processes.

Methods: *N*=53 young people aged 10-17 completed the study. *n*=23 with CTDs (Mean age=12.70, *SD*= 2.40, *n*=18 male), *n*=7 with FTLB (Mean age=16.00, *SD*=1.41, *n*=0 male), and *n*=23 controls (Mean age=12.74, *SD*=2.12, *n*=14 male). Self-report measures assessed quality of life, psychiatric comorbidities, premonitory urges, tic frequency, attentional control, and interoceptive beliefs. Participants completed cardiovascular and respiratory tasks of interoceptive accuracy, and scores were correlated with confidence ratings to assess interoceptive insight.

Results: While interoceptive beliefs significantly differed between the three groups (*F*=-2.76, *p*=.010), interoceptive accuracy on the cardiovascular (*F*=0.21, *p*=.979) and respiratory (*F*=1.88, *p*=.188) tasks did not. Interoceptive beliefs were positively associated with attentional control (*r*=.42, *p*=.002) and were a predictor of quality of life in young people with tics.

Conclusions: Unlike adults, interoceptive accuracy does not appear to differ in young people with CTDs compared to controls. In young people with tics, it appears the belief in one's interoceptive skills is more important to quality of life than actual interoceptive skills.

Keywords: chronic tic disorders, functional tic-like behaviours, tourette syndrome, interoception, young people

2.2 Introduction

2.2.1 Chronic Tic Disorders

Chronic tic disorders (CTDs) including Tourette syndrome are neurodevelopmental conditions characterised by the presence of tics, which are defined as rapid, non-rhythmic, repetitive, and sudden movements and sounds [1]. Tics commonly develop during childhood, and males are four times more likely to be affected than females [2]. High rates of comorbid neurodevelopmental and psychiatric disorders such as attention deficit hyperactivity disorder (ADHD), obsessive compulsive disorder (OCD), autism spectrum disorder (ASD), and mood disorders are associated with CTDs [2, 3]. Moreover, young people with tics report poorer quality of life, less opportunities at school, and lower self-esteem compared to those without tics [4].

Tics can be simple or complex and vary in frequency, usually following a pattern of waxing and waning in the course of a day, a week, and throughout childhood [1, 5]. Children over the age of 10 often report experiencing a premonitory urge prior to tics, and this is often described as an uncomfortable sensation, tingling, or building pressure in the body [6, 7]. Tics are considered a response to premonitory sensations as they reduce the intensity and discomfort of the urge and provide symptom relief [8]. Tics can be voluntarily suppressed for short periods of time and behavioural therapies focus on increasing this ability in young people to help manage tic severity and frequency [9]. However, the ability to suppress tics was found to be more prevalent in children older than 10 years old, and this may be because younger children either lack the ability to notice premonitory urges that signal incoming tics or have not developed the language to describe these sensations yet [9,10]. Another explanation of why premonitory urges develop only later in childhood or adolescence might be that they are not the cause of tics but develop as a consequence of having and anticipating tics [11]. This is currently unknown.

2.2.2 Functional Tic-Like Behaviours

In contrast to CTDs, functional tic-like behaviours (FTLB) are not considered a neurological disorder and are instead classed as a functional movement disorder (FMD), which is a type of functional neurological disorder [12]. Whilst cases of FTLB have been documented earlier than the 21st century, they remained rare [13]. However, during the COVID-19 pandemic, the number of young people presenting with FTLB at specialist tic clinics rapidly increased from 3-4 referrals a year, to 3-4 referrals per week [14]. It is suggested this may be due to the high levels of disruption, social isolation, and anxiety young people experienced as a result of the

pandemic and subsequent lockdowns worldwide [15, 16]. In comparison to the tics observed in CTDs, FTLB have been found to significantly affect more females than males and suddenly emerge in adolescence, rather than early childhood [16, 17]. Moreover, FTLB present with more complex and severe tics, often consisting of self-injurious tics, and do not follow the typical waxing and waning trajectory [17, 18]. Premonitory urges have also been found to be less prevalent in patients with FTLB and patients often report an inability to suppress tics [17, 18]. However, similar to the tics observed in CTDs, FTLB are highly suggestible and are affected by stress, fatigue, and anxiety [13, 19]. Researchers have also found that these tics are similarly improved by concentration and distraction, and there is evidence to suggest attention plays an integral role in symptom expression and maintenance [17, 19, 20]. Similar to CTDs, comorbidities such as ADHD, ASD, and OCD have been observed in patients with FTLB, but they are reported to have higher incidences of anxiety and depressive disorders [16, 21]. Moreover, young people with FTLB report high school absenteeism and debilitating symptoms [22], indicating their quality of life is similarly impaired.

2.2.3 Interoception

Many neurodevelopmental and psychiatric disorders are associated with lower interoceptive abilities [23]. Interoception is the ability to detect, interpret, integrate, and regulate internal signals experienced and perceived within the body [24]. Interoception requires both high and low levels of processing within the central nervous system to perceive and interpret internal bodily sensations, suggesting it is multidimensional [23, 25]. Lower interoceptive abilities are associated with poorer emotional expression and regulation, and have been linked to psychiatric disorders including anxiety, ASD, ADHD, and CTDs [23]. Similar brain structures such as the insular cortex, thalamus, and amygdala involved in interoceptive processes have been found to be active when patients with tics experience premonitory urges, suggesting atypical interoceptive processing is linked to CTDs [5, 8, 24]. Suksasilp and Garfinkel [25] propose interoception is made up of three distinctive domains; interoceptive accuracy (objective accuracy measured by performance on behavioural tasks), interoceptive beliefs (subjective accuracy assessed using self-report measures), and interoceptive insight (metacognitive awareness identified by alignment between objective and subjective measures). Interoceptive accuracy is predominantly measured using variations of the heartbeat counting task (HCT) [26] which requires participants to silently count their heartbeats for set time intervals. Similarly, interoceptive beliefs can be measured using self-report questionnaires designed to assess awareness of interoceptive signals or via individual self-reported confidence ratings judging performance on accuracy tasks [25]. Studies have found premonitory urges are associated with greater awareness of internal bodily sensations and interoceptive beliefs [27-

29], but mixed findings have been observed in interoceptive accuracy scores between adults with CTDs and healthy controls [27, 29]. Alternatively, Rae et al. [29] found adults with CTDs overestimated their interoceptive abilities compared to their objective performance, suggesting they have lower interoceptive insight. Similarly, a number of studies investigating interoception in adults with FMD using the HCT [26] have found patients with FMD have lower interoceptive accuracy [30-32] and reduced interoceptive beliefs compared to healthy controls [30, 31]. However, Millman et al. [33] found no differences in interoceptive accuracy or interoceptive beliefs measured by HCT [26] confidence ratings between adults with FMD and healthy controls. Whilst this suggests mixed findings as to whether atypical interoception is associated with FMD, small sample sizes have meant subgroup analyses cannot be conducted, meaning findings cannot be generalised to specific FMD, such as FTLB.

The above studies are limited to only adult samples and to date, there are only two studies exploring the role of interoception in young people with tics. Pile et al. [34] used standard and manipulated versions of the HCT [26] and found no differences in interoceptive accuracy in young people with CTDs compared to healthy controls. Contradictory to adult findings, premonitory urges were not associated with interoception, suggesting they are unrelated; however, this finding may be a reflection of the age-related differences in premonitory urge presence as it is known to develop in later years [10, 34]. Studies have been criticised for assessing one domain of interoceptive accuracy rather than multiple, as this ability is thought to vary within individuals [25]. Thus, Schütteler et al. [35] assessed interoceptive accuracy in young people with CTDs using the HCT [26] and a novel muscle tension task requiring participants to tense facial muscles and report observed tension. Whilst no significant differences in interoceptive accuracy were found between participants with CTDs and healthy controls, interoceptive accuracy was associated with premonitory urges, indicating a possible relationship between the two [35]. These findings are similar to those observed in adults with CTDs and suggest further research is needed to explore the link between interoception and tics in young people. Schütteler et al. [35] found the two interoceptive accuracy tasks were unrelated, supporting research stating it is domain-specific and should incorporate multiple accuracy tasks to investigate the extent of this ability [25]. Furthermore, studies into both children and adults with CTDs are limited by small sample sizes and thus, are unable to account for comorbidities in the findings which may skew the results.

2.2.4 Aims and Hypotheses

This research aimed to expand on previous research by testing young people with CTDs, young people with FTLB with or without comorbid CTDs, and compare them to controls on two interoceptive accuracy tasks (cardiovascular and respiratory). Confidence ratings for trials on

each task and self-report measures were incorporated to assess interoceptive beliefs, and correlations between objective and subjective scores were calculated to assess interoceptive insight. Self-report measures assessing attentional control, presence of tics, premonitory urges, psychiatric comorbidities, and quality of life were administered to explore differences between groups of participants and relationships between interoceptive processes.

We formed four hypotheses for this study. Firstly, we hypothesised that young people with CTDs would exhibit reduced interoceptive accuracy compared to controls. Secondly, we hypothesised that young people with FTLB would exhibit reduced interoceptive accuracy compared to young people with CTDs and controls. Thirdly, we hypothesised that young people with FTLB would exhibit reduced interoceptive insight relative to young people with CTDs and controls. And finally, we hypothesised young people with CTDs would exhibit a positive relationship between atypical interoceptive processes, quality of life, tic related factors, and anxiety symptoms.

2.3 Method

2.3.1 Ethics

This study was approved by the University of Southampton Ethics and Research Governance Committee (ERGO ID: 97471; Appendix B and C).

2.3.2 Power Analysis

A-priori power analysis was conducted to determine the minimum sample size needed to show an effect, assuming an effect size of $\eta p^2 = 0.16$, similar to Pile et al. [34] who investigated interoception in adults with Tourette syndrome. G* Power version 3.1.9.2 [36] was used to calculate this power analysis and proposed a total sample size of $N = 54$ participants ($n = 18$ participants per group) would be required to obtain an effect size with 80% power and at an $\alpha = 0.05$ significance level for a one-way analysis of variance (ANOVA).

2.3.3 Participants

A total of $N = 116$ individuals expressed an interest in this study and 46.6% ($n = 54$) were recruited. Prior to data collection $n = 1$ participant withdrew from the study, resulting in a total of $N = 53$ participants completing the study. Fig. 4 provides an overview of the recruitment process. Initially, $n = 27$ participants were recruited for the CTD sample, however on meeting the researcher (KT-C), five were identified as having possible FTLB as they recalled a late age of tic onset and described complex tics commonly seen in patients with FTLB. These participants

were assessed additionally by VB who was experienced in diagnosing FTLB. The presence of FTLB was confirmed in all five participants and they were re-allocated to the FTLB group to avoid confounding results of participants with only CTDs. Thus *n* = 23 participants with a diagnosis of a CTD were included in the CTD group and *n* = 7 participants with FTLB with or without a comorbid diagnosis of CTDs were allocated to the FTLB group. In addition, *n* = 23 participants with no history of tics nor diagnosed with CTDs or FTLB were allocated to the control group.

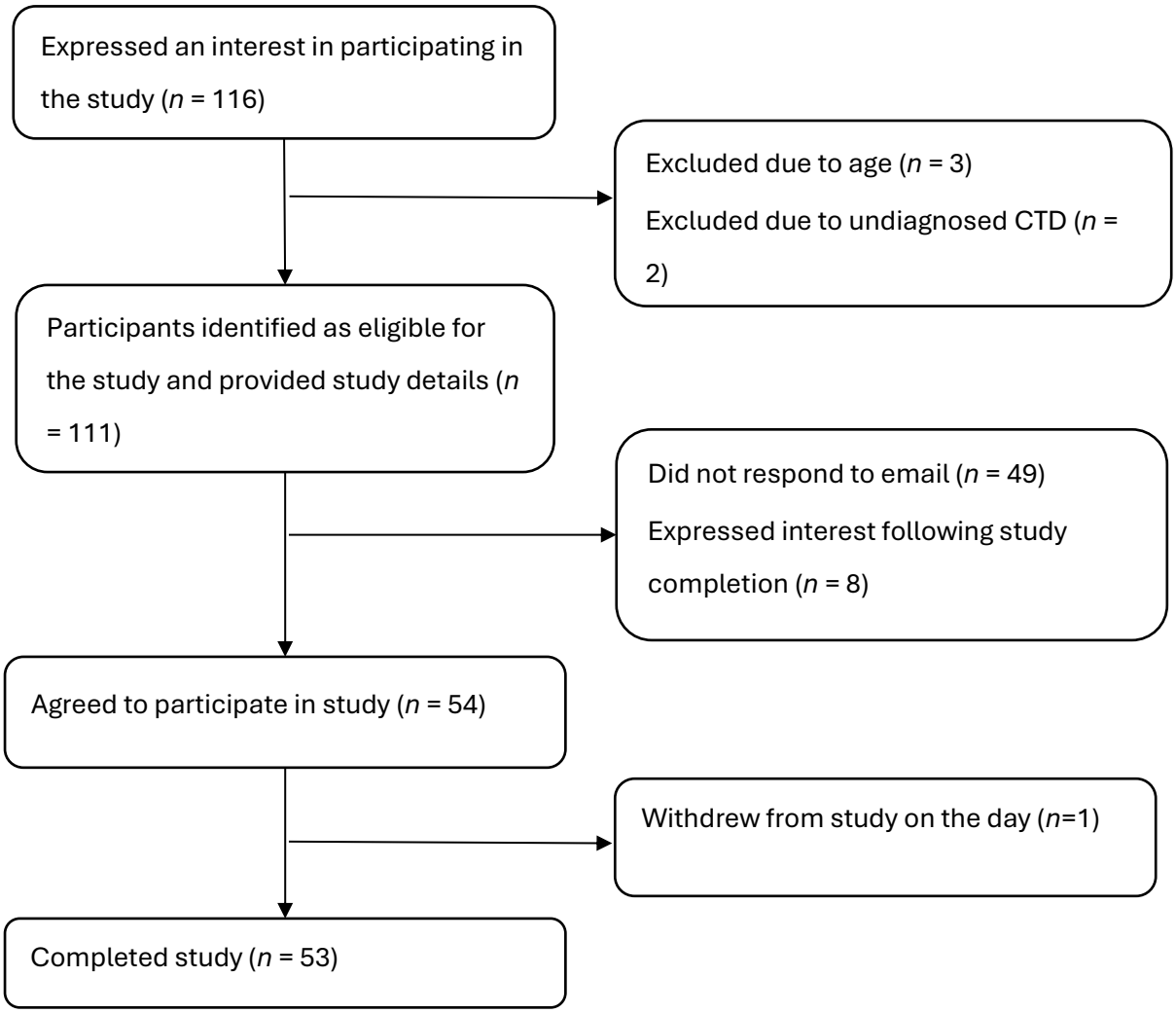


Fig. 4 Flow chart of the recruitment process

2.3.4 Measures

2.3.4.1 Demographics

Participants were provided with a demographic questionnaire used to collect information on participants' age, gender, and ethnicity, alongside details of diagnoses to ensure participants were correctly allocated to one of the three experimental groups (Appendix D).

Moreover, the matrix reasoning and vocabulary subtests of the Weschler Abbreviated Scale of Intelligence Second Edition (WASI-II) [37] were administered to provide an estimate of participant IQ.

2.3.4.2 Attentional Control

The Attentional Control Scale for Children (ASC-C; Appendix E) is a self-report measure consisting of 20 items assessing attentional control, and is split into two subscales: attentional focusing and attentional shifting [38, 39]. Items are scored on a scale ranging from 0 (never) to 3 (always) and some items are reverse-coded. Items are summed to produce subscale scores and a total score; higher scores indicate greater attention control. Melendez et al. [40] found the ACS-C to have satisfactory internal consistency (α = .74), and this study found both the attentional focusing and attentional shifting subscales to be satisfactory (α = .76, α = .78, respectively).

2.3.4.3 Tic Specific Measures

The Motor tic, Obsessions and compulsions, Vocal tic Evaluation Survey (MOVES) [41] assesses the presence and frequency of tics, along with associated obsessive compulsive symptoms (Appendix F). It consists of 20 items scored from 0 (never) to 3 (always), and higher scores indicate increased frequency of tics. The tic and obsessive-compulsive subscales were also calculated for this study, and both were found to have good internal consistency (α = .88, α = .82, respectively).

The Premonitory Urge for Tics Scale (PUTS) [42] measures premonitory experiences prior to tic onset (Appendix G). It consists of nine items scored from 1 (not at all) to 4 (very much) which when summed together produces a total score. Higher scores indicate greater intensity and severity of premonitory urges. This study found the PUTS to have good internal consistency (α = .88), similar to Pile et al. [34] who reported an internal consistency of α = .82.

The Yale Global Tic Severity Scale (YGTSS) [43] is a clinician-rated measure used to assess the frequency, impairment, and severity of tics. For this study, only the impairment section of the YGTSS was administered (Appendix H). The researcher asked questions about the impact of tics and gave participants a score between 0 and 50. Higher scores indicate greater levels of impairment.

2.3.4.4 Comorbid Psychiatric Symptoms

The Revised Child Anxiety and Depression Scale (RCADS; Appendix I) is a self-report scale measuring anxiety and depression in young people [44]. It consists of 47 items split into six

subscales and produces a total anxiety and total internalising (anxiety and depression) score. Responses range from 0 (never) to 3 (always) and are summed and converted into standardised t-scores with higher scores indicating greater severity. Previous research has found excellent internal consistency (α = .92) for the total anxiety subscale but questionable internal consistency (α = .63) for the total internalising subscale [34]. Alternatively, this study found excellent internal consistency for the depression (α = .90), generalised anxiety disorder (α = .92), panic (α = .90), total anxiety (α = .96), and total internalising (α = .96) subscales, and good internal consistency for the social phobia (α = .88), separation anxiety (α = .80), and OCD (α = .81) subscales.

The Swanson, Nolan, and Pelham Version IV Rating Scale (SNAP-IV) is a parent-report measure assessing symptoms of ADHD [45]. It consists of 27 items (Appendix J) split into three subscales: inattention, hyperactivity, and oppositional, and responses are rated on a 0 (not at all) to 3 (very much) scale and summed to produce combined ADHD and subscale scores. Higher scores indicate greater symptom severity. Excellent internal consistency was found for both the inattention (α = .93) and hyperactivity (α = .91) subscales, and good internal consistency was found for the oppositional subscale (α = .89). Similarly, Pile et al. [34] found the overall SNAP-IV had excellent internal consistency (α = .93).

2.3.4.5 Quality of Life

The Paediatric Quality of Life Inventory (Peds-QL; Version 4.0) is a self-report measure used to assess children's quality of life in four areas: physical, emotional, social, and school (Appendix K) [46]. Items are rated on a five-point scale (never to always) and can be split into two subscales (physical and psychological functioning) or summed to produce an overall quality of life score. Higher scores indicate greater functioning and quality of life. Upton et al. [47] found the physical and psychological functioning subscales to have acceptable (α = .70) and excellent (α = .90) internal reliability, respectively. This study found good internal consistency for both subscales (α = .84 and α = .89).

2.3.4.6 Interoceptive Beliefs

The Interoceptive Accuracy Scale for Children (IAS-C; Appendix L) is a child-version of the IAS developed by Murphy et al. [48]. It consists of 20 items assessing children's ability to accurately perceive bodily signals, and items are rated on a scale of 1 (strongly disagree) to 5 (strongly agree). Items are summed to produce a total score, and higher scores indicate increased perception of interoceptive cues, providing a measure of interoceptive beliefs. The IAS-C is not yet validated for use within the child population but has previously demonstrated

good internal consistency (α = .86) [49]. This study found a similar internal consistency of α = .87, further suggesting it has good reliability.

2.3.5 Experimental Tasks

The HCT [26] was used to assess cardiac interoceptive accuracy. Participants wore a pulse oximeter (Contec; CMS50E) on their non-dominant index finger. Similar instructions (Appendix M) to previous studies [29, 34, 50] were applied with participants instructed to silently count their heartbeats for four blocks of three randomised time trials (25, 35, 45 seconds). No exteroceptive cues (e.g. pulse-taking) were permitted during the task. Breaks of 30 seconds and two minutes were provided between each trial and block, respectively. Following each trial participants reported the number of heartbeats they counted. The actual number of heartbeats were recorded via the pulse oximeter. Participants were also asked to rate their confidence in perceived accuracy on a scale of 0 (no confidence/awareness) to 10 (complete confidence/awareness) as another measure of interoceptive beliefs. Participants also reported the length of time they thought each trial lasted to provide a measure of time estimation. This was controlled for in the analysis using a partial correlation to determine whether participants' awareness of time influenced interoceptive accuracy of heartbeats [51]. Accuracy scores were calculated for each trial and averaged across the four blocks to produce a mean value for each participant [50, 51], as per the formula below:

$$\frac{1}{12}\sum \frac{1 - |number\ of\ actual\ heartbeats - number\ of\ reported\ heartbeats|)}{(number\ of\ actual\ heartbeats + number\ of\ reported\ heartbeats)/2}$$

Accuracy scores ranged from 0 to 1, with values closer to 1 indicating better interoceptive accuracy. Reported confidence scores for each trial were also averaged across the four blocks to produce mean confidence ratings.

The respiratory task designed by Murphy et al. [52] was administered as a measure of respiratory interoceptive accuracy and required participants to breathe into a standard peak flow meter (Clement Clarke International; 3104710) which calculates the speed of exhalation from the lungs. Similar instructions (Appendix M) were given as outlined by Murphy et al. [52]. For each trial, participants were asked to aim for a large exhalation which was recorded as their standard (100%) and then aim for a target of this breath (e.g. 50%) for their second exhalation and results were recorded. Participants then reported their perceived percentage of how much they achieved the standard on their second breath. Participants completed six blocks of three target trials (30%, 50%, 70%) in a randomised order and were provided breaks between blocks. Participants wore a blindfold and listened to white noise using noise-cancelling headphones (Soundcore: Q20i) connected via Bluetooth to a mobile phone to prevent auditory and visual

cues aiding their performance on the task. Participants were provided with a demonstration on how to use the peak flow meter and given a practice trial prior to commencing the task. They were instructed to sit up straight and hold the peak flow meter between their hands without touching the gauge. Disposable mouthpieces were used. Exhalations that fell between two points on the gauge were rounded up to the nearest value. For each trial, the value of the participant's second exhalation was divided by their standard exhalation and multiplied by 100 to give the actual percentage achieved on the second exhalation when aiming for the standard [52]. This was then inputted into the formula below [52] to calculate absolute error scores for each target trial. These were then averaged across the six blocks to provide an average error score for each participant, providing a measure of interoceptive accuracy.

$$\frac{1}{3}\sum \frac{(|actual\ percentage - participant's\ perceived\ percentage|)}{actual\ percentage}$$

Mean scores of 0 reflected perfect interoceptive accuracy and higher scores indicated reduced interoceptive accuracy.

2.3.6 Procedure

Participants were recruited via posters (Appendix N) advertised on social media, charity websites, and within schools. Potential participants (or their guardian) expressed their interest by contacting the researcher (KT-C) who provided participant information sheets for both young people and their guardian to read prior to agreeing to taking part in the study (Appendix O). Inclusion criteria for the study was either a diagnosis of a CTD or FTLBs (or no diagnosis or history of tics for controls) and participants had to be between the ages of 10 and 17. Participants were excluded if they had a learning disability or diagnosed with an additional neurological condition (e.g. epilepsy). The presence of gasping tics was originally a basis for exclusion, however this was later revised as the researchers felt these tics would not interfere with the respiratory task.

Following written agreement to take part, participants were provided with a unique participant code and sent a hyperlink to access an online Qualtrics survey. All participants were asked to complete the ACS-C [38, 39], IAS-C [48], and RCADS [44], and only those in the CTD or FTLB group were required to complete the MOVES [41] and PUTS [42]. Participants were advised the questionnaire would take 15-20 minutes to complete and were required to consent to taking part and enter their unique participant code at the start of the online survey. Participants who were unable to complete the questionnaires online were provided with paper copies to answer on the day of the experimental task.

The experimental part of the study took place either within a lab at the University of Southampton or, at their request, in participants' homes, and the researcher (KT-C) followed lone-working policies. Participants under the age of 16 required a guardian present to provide consent whereas participants aged over 16 were not required but were advised to have a guardian present. On the day, participants were given opportunities to re-read the participant information sheets, and consent was obtained from participants over 16 years old (Appendix P) and the guardian(s) of participants under 16 years old (Appendix Q). Participants under the age of 16 were provided an assent form to sign (Appendix R). Following this, participants were asked to complete the Peds-QL [46] whilst their guardian completed the SNAP-IV [45]. Participants above 16 years old with no guardian present were asked to return the SNAP-IV via email once their guardian completed it. Participants were then administered the two subtests of the WASI-II [37], and those in the CTD and FTLB group were provided an impairment rating score on the YGTSS [43]. Participants then completed the two interoceptive accuracy tasks. Overall, this part of the study took around 90 minutes. Breaks were provided throughout the day and participants received £30 in cash following completion of the study.

2.3.7 Data Analysis

Data was analysed using IBM SPSS Statistics Version 30.0.0.0 and inspected for normal distribution and outliers. Histograms, Q-Q plots, and P-P plots indicated data had a relatively normal distribution (Appendix S). Boxplots for all variables indicated no issues with homogeneity of variance but five extreme outliers were identified. Two were associated with participant 24's scores on the panic and OCD subscales of the RCADS. Both participant 27's and 52's average error scores on trials of 50% exhalations, alongside participant 15's mean error score on trials of 70% exhalations were outliers. All five outliers were winsorized by substituting scores that were three standard deviations away from the mean [53] to prevent biasing the results. Data screening indicated that parametric tests could be used for all variables.

Chi-Square tests were run to explore differences between categorical variables such as gender and number of comorbid diagnoses. Independent *t*-tests were conducted to investigate differences in tic-specific measures between the CTD and FTLB group. Multiple one-way ANOVAs were run to assess group differences in age, IQ, quality of life, psychiatric comorbidities, attentional control, and interoceptive processes. For multiple comparisons that had not been hypothesised prior to analysis, a Hochberg's GT2 analysis was selected to account for the unequal sample sizes. Pearson correlations were conducted to assess interoceptive insight and explore relationships between interoceptive processes and self-report measures. Multiple linear regression analysis was also conducted to investigate whether

interoceptive processes, premonitory urges, and psychiatric comorbidities impacted quality of life in participants with CTDs and FTLB.

2.4 Results

2.4.1 Participant Characteristics

The mean age of participants in the CTD group (*M* = 12.70, *SD* = 2.40) was similar to those in the control group (*M* = 12.74, *SD* = 2.12), and age ranged from 10 to 17 years in both groups. Participants in the FTLB group had a mean age of 16.00 (*SD* = 1.41) and ages ranged from 13 to 17, as recruitment could not be finalised. A one-way ANOVA found age significantly differed between the three groups F (2,50) = 6.90, p = .002, ηp^2 = .216, 95% confidence intervals (CI) [0.04, 0.38]. Due to the unequal sample sizes, a post-hoc Hochberg's GT2 analysis was conducted [53]. Participants in the FTLB group were found to be significantly older in comparison to the CTD group (p = .003) and controls (p = .003), with participants on average 3.30 and 3.26 years older than CTD and controls, respectively. Age was not significantly different between the CTD group and controls (p = 1.000).

The majority of participants in the CTD and control group identified as male in comparison to all (n = 7) participants in the FTLB group who identified as female (Table 7). A chi-square test revealed significant differences in gender between the groups $\chi 2^{(2)} = 13.74$, $p < .001$ with a large effect size of V = .51, 95% bootstrapped CI [0.35, 0.70]. The Bayes factor strongly supported the alternative hypothesis BF_{02} = 200. The ethnicity of participants is presented in Table 7and a chi-square test revealed no significant differences in ethnicity between groups ($\chi 2^{(10)} = 11.42$, p = .340, V = .328, 95% bootstrapped CI [0.24, 0.74]). Participants reported a range of comorbidities (Table 7) which ranged from 0 to 5 in the CTD group (*M* = 1.09, *SD* = 1.28) and 0 to 10 in the FTLB group (*M* = 2.76, *SD* = 2.76), whereas controls had between 0 and 2 diagnoses (*M* = 0.30, *SD* = 0.56). No significant differences in IQ were found between the CTD group (*M* = 112.00, *SD* = 12.44), FTLB group (*M* = 110.29, *SD* = 8.36), or control group (*M* = 103.04, *SD* = 15.54), F (2,49) = 2.60, p = .085, ηp^2 = .096, 95% CI [0.00, 0.25).

Participants in the CTD group reported a significantly younger age of tic onset (*M* = 5.12, *SD* = 2.86) in comparison to those in the FTLB group (*M* = 10.86, *SD* = 2.19), t (28) = - 4.79, p <.001, d = -2.07, 95% CI [-3.06, -1.05]. Onset of tics ranged from age 1 to 11 for participants in the CTD group and age 7 to 13 for FTLB group. Only n = 19 participants in the CTD group recalled the age they received their diagnosis (*M* = 8.47, *SD* = 2.37) and this was significantly younger than participants in the FTLB group (*M* = 13.71, *SD* = 1.60), t (24) = -5.39, p <0.001, d = -2.38, 95% CI [-3.46, -1.27].

Table 7 Demographics of participants

	CTD (*n* = 23)		**FTLB (*n* = 7)**		**Control (*n* = 23)**		**Total Sample (*N* = 53)**	
	n	**%**	***n***	**%**	***n***	**%**	***n***	**%**
Gender								
Male	18	78.3	0	0	14	60.9	32	60.4
Female	5	21.7	7	100	9	39.1	21	39.6
Ethnicity								
Asian, Asian British, or Asian Welsh	1	4.3	0	0	2	8.7	3	5.7
Black, Black British, Black Welsh, Caribbean, or African	1	4.3	0	0	2	8.7	3	5.7
Mixed or Multiple ethnic groups	3	13.0	0	0	1	4.3	4	7.5
White English, Welsh, Scottish, Northern Irish, or British	17	73.9	6	85.7	18	78.3	41	77.4
Other	1	4.3	1	14.3	0	0	2	2.8
Comorbidities								
ADHD	3	13.0	1	14.3	3	13.0	7	13.2
ASD	3	13.0	3	42.0	0	0	6	11.3

	CTD (*n* = 23)		FTLB (*n* = 7)		Control (*n* = 23)		Total Sample (*N* = 53)	
	n	**%**	***n***	**%**	***n***	**%**	***n***	**%**
Anxiety	3	13.0	4	57.1	0	0	7	13.2
Depression	1	4.3	0	0	0	0	1	1.9
OCD	4	17.4	2	28.6	0	0	6	11.3
Dyslexia	3	13.0	1	14.3	1	4.3	5	9.4
Dyspraxia	1	4.3	1	14.3	0	0	2	3.8
FNS	0	0	3	42.9	0	0	3	5.7

Abbreviations: CTD Chronic Tic Disorder, FTLB Functional Tic-Like Behaviours, ADHD Attention Deficit Hyperactivity Disorder, ASD Autism Spectrum Disorder, OCD Obsessive Compulsive Disorder, FNS Functional Neurological Symptoms

2.4.2 Self-Report Measures

Table 8 provides means, standard deviations, and statistical analyses between groups for each self-report measure and associated subscales. A one-way ANOVA revealed scores on the Peds-QL significantly differed between groups (Table 8). A post-hoc Hochberg GT2 analysis was conducted on the psychological functioning subscale of the Peds-QL and the FTLB group were found to have significantly lower scores (*MD* = 18.64, *SD* = 6.55, *p* = .019, 95% CI [2.49, 34.80]) than controls, suggesting poorer psychological quality of life. However, no significant differences were found between FTLB and CTD participants (*MD* = 13.72, *SD* = 6.55, *p* = .118, 95% CI [-2.44, 29.87]) or between controls and CTD participants (*MD* = 4.93, *SD* = 4.47, *p* = .616, 95% CI [-6.11, 15.97]) in psychological functioning. Moreover, multiple one-way ANOVAs found significant differences in scores on the generalised anxiety disorder and social phobia subscales of the RCADS between the three groups (Table 8). Hochberg GT2 post-hoc analyses revealed no significant differences between the FTLB and CTD group scores on the generalised anxiety disorder subscale (*MD* = 1.64, *SD* = 6.03, *p* = .990, 95% CI [-13.25, 16.53]) or social phobia subscale (*MD* = 2.79, *SD* = 5.15, *p* = .930, 95% CI [-9.91, 15.49]). No significant differences were also found between FTLB participants and controls in generalised anxiety disorder subscale scores (*MD* = 12.16, *SD* = 6.03, *p* = .139, 95% CI [-2.73, 27.05]) and social phobia subscale scores (*MD* = 12.35, *SD* = 5.15, *p* = .059, 95% CI [-0.35, 25.05]). However, the CTD group were found to have significantly more symptoms of generalised anxiety disorder (*MD* = 10.52, *SD* = 4.12, *p* = .041, 95% CI [-0.35, 20.69]) and social phobia (*MD* = 9.57, *SD* = 3.52, *p* = .026, 95% CI [0.89, 18.24]) in comparison to controls.

Table 8 Descriptive statistics and one-way ANOVA results of group differences on self-report measures

Measure	CTD	FTLB	Control	*F* (2, 50)	*p*	ηp² [95% CI]
	M (SD)	*M (SD)*	*M (SD)*			
ACS-C						
Attentional Focusing	20.2 (5.1)	20.9 (3.9)	21.1 (5.5)	0.19	.828	0.01 [0.00, 0.07]
Attentional Shifting	26.3 (6.9)	24.4 (6.5)	26.1 (4.6)	0.28	.760	0.01 [0.00, 0.09]
Total Score	46.5 (11.3)	45.3 (9.1)	47.2 (8.5)	0.10	.903	0.00 [0.00, 0.5]
IAS-C						
Total Score	82.4 (12.1)	68.0 (7.6)	83.5 (9.8)	6.08	.004*	0.20 [0.02, 0.36]
MOVES						
Tics	10.6 (5.8)	17.3 (5.1)	-	-2.76[a]	.010*	-1.19 [-2.08, -0.28]
Obsessive-Compulsive	9.1 (5.7)	12.6 (4.3)	-	-1.56[a]	.155	-0.63 [-1.49, 0.24]
Total Score	22.3 (12.9)	34.9 (10.7)	-	-2.33[a]	.027*	-1.01 [-1.88, -0.11]
Peds-QL						
Physical Functioning	78.4 (13.5)	54.9 (27.9)	79.6 (11.3)	2.50[b]	.116	0.24 [0.05, 0.40]

Measure	CTD	FTLB	Control	*F* (2, 50)	*p*	ηp² [95% CI]
	M (SD)	*M (SD)*	*M (SD)*			
Psychological Functioning	66.8 (14.9)	53.1 (24.1)	71. 7 (12.0)	4.06	.023*	0.14 [0.00, 0.30]
Total Score	70.8 (13.1)	53.7 (25.0)	74.5 (9.8)	2.51[c]	.116	0.19 [0.02, 0.36]
PUTS						
Total Score	22.6 (7.5)	27.7 (8.6)	-	-1.53[a]	.136	-0.66 [-1.52, 0.21]
RCADS						
Separation Anxiety	59.0 (16.8)	63.0 (17.8)	50.6 (10.4)	2.92	.063	0.11 [0.00, 0.26]
Generalised Anxiety Disorder	56.2 (17.0)	57.9 (13.2)	45.7 (10.3)	4.57[d]	.026*	0.14 [0.00, 0.30]
Panic	63.7 (19.5)	65.0 (21.5)	53.4 (14.5)	2.35	.106	0.09 [0.00, 0.23]
Social Phobia	56.8 (13.1)	59.6 (12.6)	47.2 (10.4)	4.93	.011*	0.17 [0.01, 0.33]
OCD	49.4 (13.1)	57.1 (13.3)	44.9 (11.4)	2.72	.075	0.10 [0.00, 0.25]
Depression	59.4 (15.7)	71.0 (21.2)	54.6 (16.9)	2.54	.089	0.09 [0.00, 0.24]
Total Anxiety	59.4 (17.5)	63.6 (17.1)	47.8 (11.7)	4.69	.014	0.16 [0.01, 0.32]
Total Internalising	60.2 (17.7)	66.3 (18.8)	49.3 (13.0)	4.28	.019	0.15 [0.00, 0.31]

Measure	CTD	FTLB	Control	*F* (2, 50)	*p*	ηp^2 [95% CI]
	M (SD)	*M (SD)*	*M (SD)*			
SNAP-IV						
Inattention	10.0 (6.5)	9.3 (10.2)	10.4 (7.6)	0.06	.947	0.00 [0.00, 0.03]
Hyperactivity	9.4 (6.7)	7.7 (6.0)	6.6 (7.0)	1.00	.374	0.04 [0.00, 0.16]
Oppositional	5.6 (5.2)	5.6 (5.5)	4.7 (4.6)	0.20	.820	0.01 [0.00, 0.76]
Combined ADHD	19.4 (12.4)	17.0 (16.0)	17.0 (13.6)	0.22	.803	0.01 [0.00, 0.80]
YGTSS						
Impairment	19.1 (11.3)	23.6 (8.0)	-	-0.97[a]	.340	-0.42 [-1.27, 0.44]

Abbreviations: CTD Chronic Tic Disorder, FTLB Functional tic-like behaviours, ACS-C Attentional Control Scale for Children, IAS-C Interoceptive Accuracy Scale for Children, MOVES Motor tic, Obsessions and compulsions, Vocal tic Evaluation Survey, Peds-QL Paediatric Quality of Life Inventory, PUTS Premonitory Urge for Tics Scale, RCADS Revised Child Anxiety and Depression Scale, OCD Obsessive Compulsive Disorder, SNAP-IV Swanson, Nolan, and Pelham Version IV Rating Scale, YGTSS Yale Global Tic Severity Scale, CI Confidence Intervals

[a] Independent t-test (two groups only), reporting t (28) instead of F statistic. Cohen's d effect size reported instead of partial eta-squared.

[b] Variances not equal, Welch F value reported instead with df (2, 14.7)

[c] Variances not equal, Welch *F* value reported instead with df (2, 14.6)

[d] Variances not equal, Welch *F* value reported instead with df (2, 16.5)

* $p < .05$.

2.4.3 Interoceptive Accuracy

A one-way ANOVA found no significant differences in mean HCT accuracy scores (Fig. 5) between the three groups, F (2,50) = 0.21, p = .979, ηp^2 = 0.00, 95% CI [0.00, 0.00]. No significant differences between groups in HCT accuracy scores remained after controlling for participants' time estimation[1], F (2,41) = 0.25, p = .778, ηp^2 = 0.01. One participant in the FTLB group was unable to complete the full six trials of the respiratory task due to physical health issues, and thus n = 52 remained for mean respiratory accuracy scores. Similar to the HCT, a one-way ANOVA revealed no significant differences in accuracy scores on the respiratory task (Fig. 5) between the three groups, F (2,49) = 1.88, p = .188, ηp^2 = 0.07, 95% CI [0.00, 0.20].

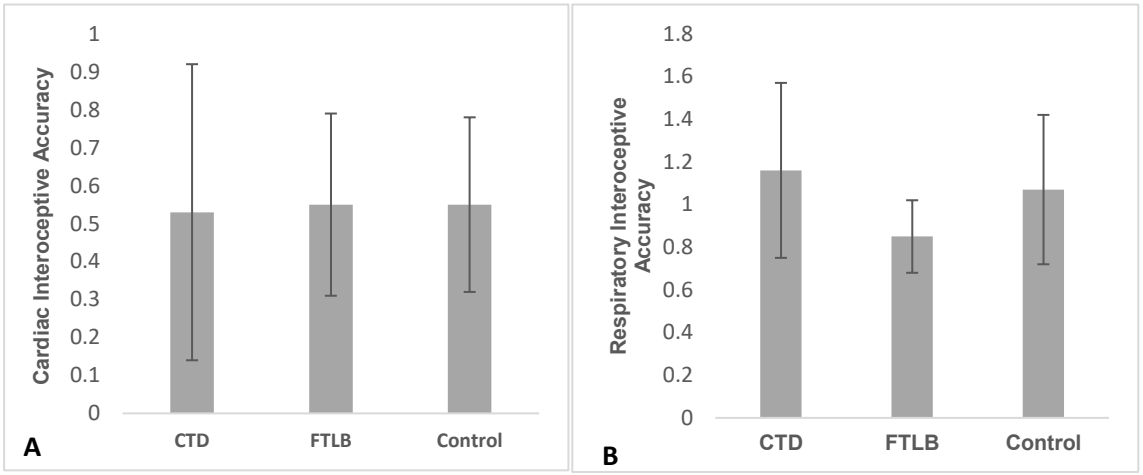


Fig. 5 Mean scores and standard deviations for each group on tasks of interoceptive accuracy **A** Cardiovascular interoceptive accuracy scores using the heartbeat counting task **B** Respiratory interoceptive accuracy scores

2.4.4 Interoceptive Beliefs

A one-way ANOVA revealed no significant differences in mean confidence ratings (Fig. 6) between the three groups, F (2,50) = 0.36, p = .703, ηp^2 = 0.01, 95% CI [0.00, 0.10]. Moreover, a Pearson correlation found accuracy scores on the HCT and mean confidence ratings were not

[1] A total of n = *8* participants reported no awareness of the time pass and offered no guess. Thus time estimation ability is based on n = 45.

significantly associated with each other, r = -.02, p = .896. Alternatively, a one-way ANOVA found a significant difference (p = .004) between groups in mean IAS-C scores (Table 8; Fig. 6). A post-hoc Hochberg GT2 analysis revealed the FTLB group had significantly lower scores on the IAS-C compared to the CTD group (*MD* = 14.43, *SD* = 4.58, p = .008, 95% CI [3.13, 25.74]) and controls (*MD* = 15.48, *SD* = 4.59, p = .004, 95% CI [4.17, 26.79]), suggesting a reduced ability to perceive internal bodily signals. No significant differences were found between participants with CTDs and controls (*MD* = -1.04, *SD* = 3.13), p = .982, 95% CI [-8.77, 6.68]. A Pearson correlation revealed confidence ratings on the HCT and IAS-C scores were not significantly correlated, r = -.09, p = .526, 95% CI [-.350, .187].

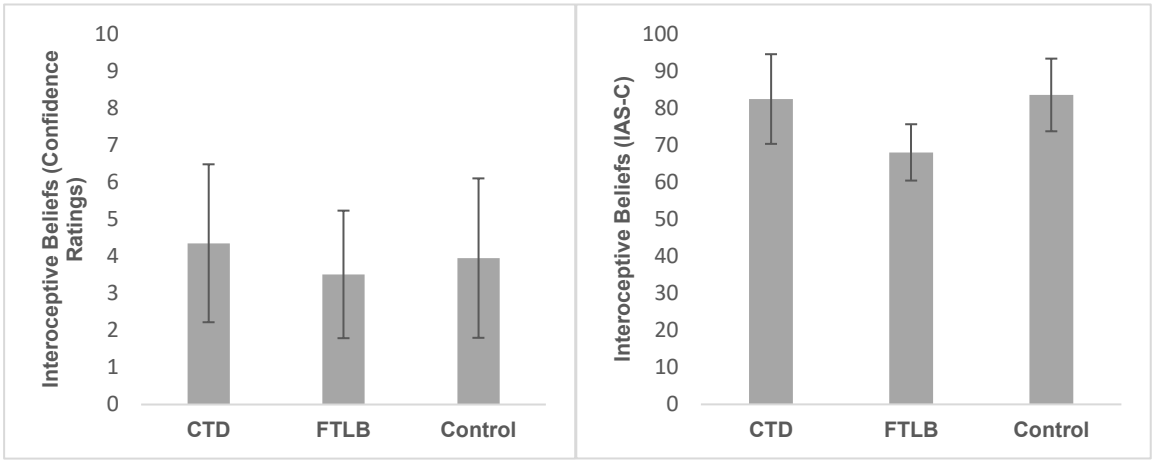


Fig. 6 Mean scores and standard deviations for each group on measures of interoceptive beliefs **A** Interoceptive beliefs measured using confidence ratings on the heartbeat counting task **B** Interoceptive beliefs measured using the IAS-C

2.4.5 Interoceptive Insight

For each participant, interoceptive insight was analysed using a Pearson correlation between accuracy scores and confidence ratings within the HCT. Mean scores for each group are shown in Fig. 7. A one-way ANOVA using individual participant interoceptive insight scores revealed no significant differences in interoceptive insight between the three groups, F (2,50) = 2.52, p = .091, ηp^2 = 0.09, 95% CI [0.00, 0.24].

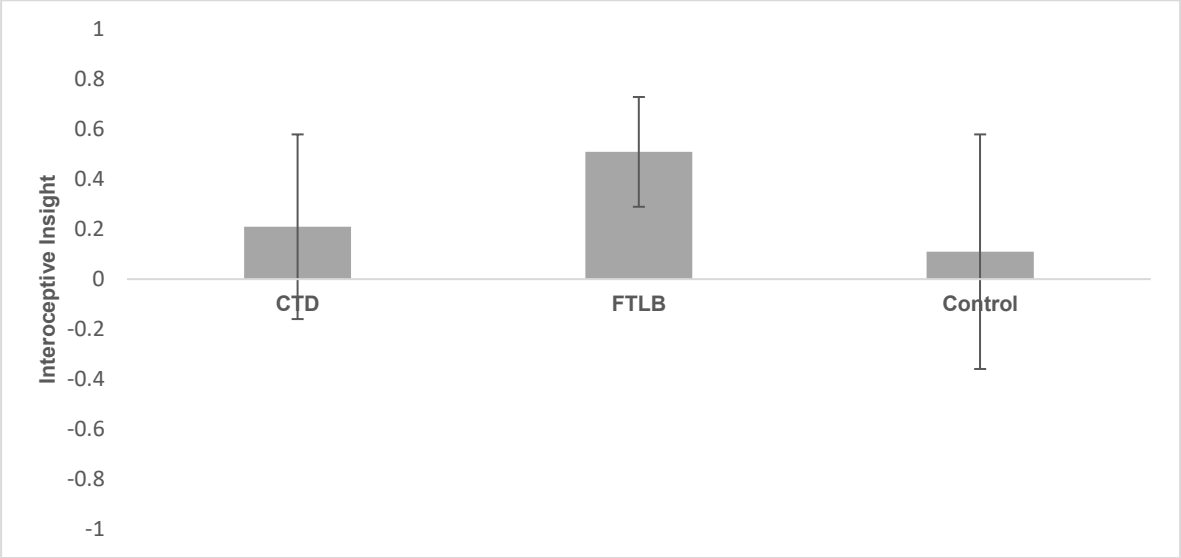


Fig. 7 Mean interoceptive insight scores and standard deviations for each group

2.4.6 Interoceptive Processes and Self-Report Measures

Pearson correlations were conducted to explore the relationship between interoceptive accuracy and self-report measures. Correlation analyses revealed respiratory interoceptive accuracy was significantly associated with higher self-reported interoceptive accuracy, $r = .37$, $p = .008$. No other self-report measures were significantly correlated with cardiac or respiratory interoceptive accuracy and results are shown in Table 9.

Table 9 Descriptive statistics and correlation coefficients for study variables

			1. HCT Accuracy Scores		2. Respiratory Task Accuracy Scores		3. IAS-C Total Score	
Variables	***M***	***SD***	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**
1. HCT Accuracy Scores	0.54	0.31						
2. Respiratory Task Accuracy Scores	1.08	0.37	-.71	.617 [-.34, .21]				
3. IAS-C Total Score	80.98	11.61	-.10	.481 -.360, .176]	.37	.008* [.103, .580]		
Self-Report Measures								
ACS-C Attentional Focusing	20.66	5.03	-.17	.228 [-.420, .107]	-.11	.458 [-.367, .173]	.20	.149 [-.073, .447]
ACS-C Attentional Shifting	25.96	5.86	-.11	.455 [-.365, .170]	.24	.094 [-.041, .477]	.52	<.001* [.286, .691]
ACS-C Total Score	46.62	9.72	-.15	.283 [-.404, .125]	.09	.541 [-.191, .351]	.42	.002* [.163, .616]

			1. HCT Accuracy Scores		2. Respiratory Task Accuracy Scores		3. IAS-C Total Score	
Variables	***M***	***SD***	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**
MOVES – Tics[b]	12.17	6.22	.17	.364 [-.201, .501]	-.08	.671 [-.436, .293]	-.34	.070 [-.621, .029]
MOVES – Obsessive-Compulsive[b]	9.93	5.56	.03	.861 [-.331, .389]	.07	.722 [-.305, .425]	-.27	.153 [-.573, .103]
MOVES Total Score[b]	25.23	13.41	.12	.525 [-.250, .461]	.00	.996 [-.366, .367]	-.31	.093 [-.605, .054]
Peds-QL – Physical Functioning	75.83	16.99	-.04	.773 [-.307, .232]	.13	.349 [-.146, .391]	.45	<.001* [.207, .643]
Peds-QL – Psychological Functioning	67.14	16.04	-.02	.893 [-.288, .253]	-.05	.723 [-.319, .226]	.36	.007* [.104, .578]
Peds-QL Total Score	70.16	15.20	-.03	.838 [-.297, .243]	.01	.920 [-.260, .286]	.43	.001* [.176, .625]
PUTS Total Score[b]	23.80	7.89	.22	.253 [-.157, .534]	.06	.764 [-.315, .416]	-.15	.423 [-.486, .220]

			1. HCT Accuracy Scores		2. Respiratory Task Accuracy Scores		3. IAS-C Total Score	
Variables	***M***	***SD***	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**
RCADS – Separation Anxiety	55.85	15.00	-.09	.540 [-.348, .189]	.12	.381 [-.154, .384]	-.12	.383 [-.380, .153]
RCADS – Generalised Anxiety Disorder	51.87	14.76	.09	.540 [-.189, .348]	.08	.576 [-.198, .345]	-.07	.644 [-.329, .209]
RCADS - Panic	59.42	18.22	-.01	.943 [-.280, .261]	.19	.174 [-.086, .441]	.00	.984 [-.268, .273]
RCADS – Social Phobia	53.00	12.79	.01	.934 [-.259, .281]	-.03	.809 [-.304, .241]	-.04	.782 [-.306, .234]
RCADS – OCD	48.45	12.79	.04	.792 [-.236, .304]	.10	.482 [-.178, .363]	-.01	.933 [-.281, .259]
RCADS – Depression	58.83	17.47	.05	.724 [-.224, .316]	.03	.846 [-.247, .298]	-.28	.043* [-.511, -.010]
RCADS – Total Anxiety	54.94	16.19	.02	.912 [-.236, .760]	.12	.446 [-.170, .370]	-.05	.702 [-.319, .220]

			1. HCT Accuracy Scores		2. Respiratory Task Accuracy Scores		3. IAS-C Total Score	
Variables	***M***	***SD***	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**	***r***	***P* [95% CI]**
RCADS – Total Internalising	56.28	16.92	.10	.485 [-.179, .362]	-.07	.617 [-.337, .206]	-.11	.449 [-.366, .169]
SNAP-IV – Inattention	10.06	7.37	.02	.908 [-.255, .285]	.04	.803 [.240, .305]	-.22	.111 [-.464, .052]
SNAP-IV – Hyperactivity	7.98	6.79	.01	.970 [-.265, .275]	.15	.306 [-.134, .402]	-.26	.061 [-.495, .012]
SNAP-IV – Oppositional	5.19	4.90	-.05	.723 [-.316, .224]	.15	.274 [-.124, .410]	-.03	.823 [-.299, .241]
SNAP-IV – Combined ADHD	18.04	13.20	.01	.993 [-.259, .281]	.10	.505 [-.183, .358]	-.26	.063 [-.493, .014]
YGTSS – Impairment[b]	20.13	10.69	.27	.151 [-.101, .574]	.03	.862 [-.337, .395]	-.42	.022* [-.676, -.068]

Abbreivations: HCT Heartbeat Counting Task, ACS-C Attentional Control Scale for Children, IAS-C Interoceptive Accuracy Scale for Children, MOVES Motor tic, Obsessions and compulsions, Vocal tic Evaluation Survey, Peds-QL Paediatric Quality of Life Inventory, PUTS Premonitory Urge for Tics Scale, RCADS Revised

Child Anxiety and Depression Scale, OCD Obsessive Compulsive Disorder, SNAP-IV Swanson, Nolan, and Pelham Version IV Rating Scale, YGTSS Yale Global Tic Severity Scale, CI Confidence Intervals

[a] Pearson Correlation based on *n* = 52 participants only, as *n* = 1 participant in the FTLB group failed to complete the respiratory interoceptive accuracy task

[b] Pearson Correlation based on *n* = 30 participants (only those in CTD and FTLB groups) as controls were not provided this self-report measure

* *p* = 0.05

2.4.7 Regression Analysis

A multiple linear regression analysis was conducted to explore the influence of interoceptive accuracy, interoceptive beliefs, premonitory urges, and psychiatric symptoms on predicting quality of life in young people with CTDs and FTLB (n = 30). Quality of life was the outcome variable for this regression analysis. HCT accuracy scores, total internalising scores, combined ADHD scores, IAS-C, and PUTS scores were predictor variables. Results are shown in Table 10. The overall model was significant (R^2 = .58, F (5, 24) = 6.80, p < .001), with the predictor variables accounting for 58% of the variance in quality of life. However, only the IAS-C and internalising scale of the RCADS were significant predictors of the quality of life, accounting for 8% and 28% of the model, respectively.

Table 10 Linear regression model showing predictors of quality of life in participants with CTDs and FTLB

Variables	*β*	*SE β*	*t*	*p*	sr^2	95% CI
HCT Accuracy Scores	.62	6.84	0.09	.929	0.00	[-13.50, 14.74]
RCADS – Total Internalising	-.58	0.14	-4.05	<.001	0.28	[-0.88, -0.29]
SNAP-IV – Combined ADHD	-.15	0.19	-0.75	.459	0.01	[-0.55, 0.25]
IAS-C	.42	0.20	2.11	.045	0.08	[0.01, 0.83]
PUTS	-.16	0.33	-0.47	.644	0.00	[-0.84, 0.53]

Abbreviations: HCT Heartbeat Counting Task, IAS-C Interoceptive Accuracy Scale for Children, PUTS Premonitory Urge for Tics Scale, RCADS Revised Child Anxiety and Depression Scale, SNAP-IV Swanson, Nolan, and Pelham Version IV Rating Scale, CI Confidence Intervals

2.5 Discussion

This study aimed to investigate whether interoceptive processes differed in young people aged 10 to 17 years old with CTDs to young people with FTLB, and controls. Moreover, the study also aimed to explore whether interoceptive ability was associated with severity of tics and premonitory urges, alongside attentional control, psychiatric comorbidities, and quality of life.

In this sample, young people with CTDs did not have reduced interoceptive accuracy on both cardiovascular and respiratory performance tasks compared to controls. This finding was consistent with the two previous studies investigating interoception in young people with CTDs, which found no significant differences in both cardiovascular and muscle tension tasks [34, 35]. Moreover, our study found the two interoceptive accuracy tasks were uncorrelated, supporting the view that this ability may not be a stable trait across bodily domains [25, 35]. Thus, our findings indicate young people with CTDs do not have impaired interoceptive accuracy in either domain. One explanation for this may be that altered interoceptive accuracy develops in adulthood following chronic exposure to prediction errors in interpreting and responding to sensory stimuli (due to the extra movements), which may lead to structural changes in the brain [35]. This may explain why impaired interoceptive accuracy has been in found in adults with CTDs only. However, more research is needed to determine whether brain structures involved in interoception (e.g. insular cortex) are abnormal prior to tic onset or develop because of chronic tics.

Our second hypothesis that young people with FTLB would have reduced interoceptive accuracy in comparison to those with CTDs and controls, was also disproved. Results revealed young people with FTLB had higher scores of cardiovascular interoceptive accuracy compared to those with CTDs and controls. Results also showed young people with FTLB had higher scores of respiratory interoceptive accuracy compared to those with CTDs, and similar scores when compared to controls. However, these findings were non-significant. Whilst this provides support for Millman et al. [33] it contradicts previous studies which found adults with FMD have reduced interoceptive accuracy compared to controls [30-32]. Furthermore, a recent study conducted brain scans on adults with FMD whilst undertaking interoceptive accuracy tasks across multiple bodily domains, and found evidence that FMD is associated with abnormal interoceptive processes in the brain [54]. However, these studies were conducted on adult samples and participants had mixed subtypes of FMD, whereas our study solely investigated young people with a specific sub-type of FMD, possibly explaining the differences in findings.

Our study revealed mixed findings when investigating differences in interoceptive beliefs between young people with CTDs, young people with FTLB, and controls. To the researcher's knowledge, this was the first study on young people with CTDs to include confidence ratings on the HCT [26] as a measure of interoceptive beliefs. Whilst young people with CTDs had the highest confidence in perceived accuracy, the findings were not significant. This supports existing research which found no differences in confidence ratings in adults with CTD [29] or adults with FMD when compared to controls [31, 33]. Researchers propose confidence ratings are a measure of participant's beliefs into their perceived accuracy at that moment [25, 50]. However, our study found confidence ratings and HCT [26] accuracy were uncorrelated,

indicating this may not be a valid measure of interoceptive beliefs on cardiovascular tasks, and instead may be measuring awareness of heartbeats [55].

In contrast to confidence ratings, the IAS-C [48] is theorised to be a global measure of interoceptive beliefs, and not specific to bodily domains [25]. This study found young people with FTLB had significantly lower scores on the IAS-C [48] in comparison to young people with CTDs and controls, indicating they believe themselves to be poor at accurately detecting interoceptive cues. However, interoceptive beliefs were not significantly different between young people with CTDs and controls. This supports previous research in which adults with FMD were found to have reduced interoceptive beliefs compared to controls [30-32] and Pile et al. [34] found no significant differences in perceived accuracy in children with CTDs versus controls. One explanation for our findings is that FTLB, and other sub-types of FMD, are associated with atypical bottom-up and top-down processes in the brain which are moderated by attention and increased focus on specific body parts [56]. This, alongside chronic prediction errors, can result in individuals perceiving movements as involuntary and outside of their control, which in turn leads to a belief that they are incapable of correctly identifying internal cues [13, 56]. However, our study found no differences between the three groups in their ability to shift, control, and focus attention, but did find that increased abilities to shift and control attention were associated with greater interoceptive beliefs. This indicates a possible relationship between attentional and interoceptive processes, but implies it is not specific to individuals with FTLB only.

Previous studies on children and adults with CTDs have found premonitory urges are associated with interoceptive accuracy [27-29, 35] and the PUTS [42] has been proposed as a possible measure of interoceptive beliefs, as it judges perceptions of these sensations [35]. However, our study found premonitory urges were not associated with either interoceptive accuracy tasks or the IAS-C [48], suggesting the PUTS [42] may not be a valid measure of interoceptive beliefs, and thus more research is required. It also implies premonitory urges may not be linked to interoceptive processes in young people with CTDs or FTLB. This is supported by Pile et al. [34] who found no relationship between premonitory urges and interoceptive accuracy in children with CTDs. Our findings may differ from adult studies due to the fact that premonitory urges tend to increase with age, and thus interactions with interoceptive processes may not develop until adulthood [6]. Moreover, our study found no differences in premonitory urges between young people with FTLB and those with CTDs, which contradicts the literature which often reports FTLB are associated with fewer or no premonitory sensations in comparison to CTDs [17, 18].

Our third hypothesis that young people with FTLB would have reduced interoceptive insight compared to young people with CTDs and controls was falsified as results revealed no significant differences in this metacognitive process between the three groups. This supports previous research into adults with FMD [31] and adults with CTDs [29] which also found no differences in interoceptive insight when comparing to controls. Whilst our findings suggest interoceptive insight is not atypical in young people with CTDs and FTLB, they may also indicate that individuals with tics have an awareness of their reduced ability to accurately detect interoceptive signals, resulting in no mismatch between subjective and objective ability [31]. Thus, young people with CTDs and FTLB appear to have unimpaired metacognition [31]. More research is required to explore this further.

Our regression analyses showed increased interoceptive beliefs and fewer symptoms of anxiety and depression were significant predictors of improved quality of life in young people with CTDs and FTLB. This partially supports our fourth hypothesis that young people with CTDs would demonstrate a positive relationship between impaired interoceptive processes, quality of life, tic related factors, and anxiety symptoms. However, due to the small sample size we were unable to conduct a sub-group analysis to assess whether this was specific to young people with CTDs only. This contrasts with an existing study which found increased awareness of heartbeats were associated with increased anxiety and reduced quality of life [34]. Our findings suggest that by increasing confidence in abilities to accurately perceive interoceptive signals, young people with CTDs and FTLB may experience better physical and psychosocial functioning in life. However, further research is required to understand the mechanisms involved in these processes, and whether other dimensions of tics, such as severity and frequency, are predictive of this relationship, particularly as premonitory urges were non-significant.

2.5.1 Clinical Implications

The findings of this study suggest that interoceptive beliefs likely play a role in the development or maintenance of FTLB in young people. Thus, it may be beneficial for clinicians to explore young people's beliefs around their ability to notice FTLB symptoms, and additional interoceptive cues, to gain an insight into their perceived accuracy. The positive association between attention shifting/control and interoceptive beliefs suggests interventions focused on increasing abilities to shift and focus attention to and away from internal signals may improve confidence in detecting stimuli and reduce symptoms of FTLB. This is supported by Robinson and Hedderly [20] who found symptoms of patients with FMD improved following interventions that increased external attention and reduced internal focus of bodily sensations. A recent case series also found evidence that externalised attention strategies can be beneficial in reducing

tic frequency and severity in adults with CTDs [57], indicating attentional strategies may be useful in treating both CTDs and FTLB.

Moreover, our findings suggest that improved quality of life in young people with CTDs and FTLB is associated with fewer symptoms of comorbid anxiety and depression. This highlights the need for clinicians to identify and treat co-occurring psychiatric symptoms to improve the wellbeing of patients with CTDs and FTLB. Whilst behavioural therapies for tics have been found to improve quality of life in patients with CTDs, poorer outcomes have been found for patients with comorbid psychiatric conditions [58]. Thus, this suggests the importance of treating comorbid psychiatric symptoms prior to treating tics to improve quality of life and treatment efficacy [58].

The findings of this study revealed young people with CTDs and FTLB have similar interoceptive accuracy to controls, suggesting impairments in interoceptive accuracy may develop with age. This is supported by Brand et al [49] who found interoceptive accuracy on the HCT [26] was not associated with psychopathology or somatising symptoms in children, despite having been evidenced in adult populations. Similarly, Braet et al [59] found no associations between interoceptive accuracy on the HCT [26] and emotion regulation difficulties. Whilst some authors argue interoception remains stable throughout development [60], Braet et al [59] argues children have not fully developed higher order cognitive processes involved in interoception and emotional expression, proposing that impairments in these abilities are not noticed until adulthood when the brain is fully developed. However, Nicholson et al [61] found children with ASD had poor interoceptive accuracy, but adults with ASD did not, in comparison to typically developing controls, and proposes instead that impairments in interoception resolve with age. Thus, more research is required to understand the developmental aspect of interoception and how these processes may differ between children and adults, to then compare with our own findings.

2.5.2 Strengths and Limitations

One strength of our study was the inclusion of the respiratory task designed by Murphy et al. [52]. This domain of interoceptive accuracy had not yet been investigated in either adults or young people with CTDs, and thus our findings extend previous research by suggesting atypical interoceptive accuracy is not present in either the cardiovascular or respiratory domain. The findings also support the view that interoceptive accuracy varies within individuals and highlights the importance of future studies including multiple measures of interoceptive accuracy across separate bodily domains to improve the reliability and validity of the results.

Our study also provides the first attempt to investigate interoceptive processes on a sample of young people with FTLB. Our findings provide support for proposed criteria in distinguishing between FTLB and CTDs, as we found the former to be significantly older at age of symptom onset and have higher rates of comorbid disorders, such as anxiety [16]. However, the size of the sample of young people with FTLB is a major limitation of this study. As the sample was extremely small and underpowered, all findings must be interpreted with caution and cannot be generalised. Moreover, most participants in the FTLB group had a comorbid diagnosis of CTDs, suggesting a possible functional overlay between the two. The presence of comorbid CTDs may be a possible confounding variable in this group. However, due to the small sample size sub-group analysis could not be conducted. Future studies should look to include larger samples of both young people with FTLB and comorbid CTDs and FTLB only, to investigate whether differences exist between the two.

Another limitation of our study is that the IAS-C [48] and confidence ratings were both found to be uncorrelated with the cardiovascular interoceptive accuracy tasks. This indicates that they are not measuring participants' perceived ability to accurately detect heartbeats, suggesting neither are a valid measure of interoceptive beliefs. However, the IAS-C [48] was significantly correlated with the accuracy scores on the respiratory task [52]. These findings imply issues with construct validity and thus the results of this study may not be valid or reliable. Future research should focus on validating the IAS-C [48] in child populations and conducting exploratory factor analysis on this measure to assess whether it measures interoceptive beliefs.

2.5.3 Conclusion

Overall, our study found young people with FTLB had poorer interoceptive beliefs compared to young people with CTDs and controls. However, no differences in either domain of interoceptive accuracy or interoceptive insight were found between the three groups. Interoceptive beliefs were associated with attentional shifting and control and were a predictor of quality of life, alongside reduced anxiety and depression. Severity of premonitory urges did not differ between young people with CTDs and young people with FTLB, and was not associated with interoceptive processes. This study is limited by the small sample size of young people with FTLB and thus the validity of the results is questionable. Future research should aim to investigate interoceptive processes in young people with FTLB using larger samples in order to compare to young people with CTDs.

2.6 References

1. American Psychiatric Association (2013) Neurodevelopmental disorders. In: Diagnostic and statistical manual of mental disorders, 5th edn. https://psychiatryonline.org/doi/book/10.1176/appi.books.9780890425596
2. Freeman RD, Fast DK, Burd L, Kerbeshian J, Robertson MM, Sandor P (2000) An international perspective on Tourette syndrome: selected findings from 3500 individuals in 22 countries. Dev Med Child Neurol 42:436-447. https://doi.org/10.1017/s0012162200000839
3. Hirschtritt ME, Lee PC, Pauls DL, Dion Y, Grados MA, Illmann C, King RA, Sandor P, McMahon WM, Lyon GJ, Cath DC, Kurlan R, Robertson MM, Osiecki L, Scharf JM, Mathews CA (2015) Lifetime prevalence, age of risk, and genetic relationships of comorbid psychiatric disorders in Tourette syndrome. JAMA Psychiatr 72:325-333. https://doi.org/10.1001/jamapsychiatry.2014.2650
4. Eapen V, Cavanna AE, Robertson MM (2016) Comorbidities, Social Impact, and Quality of Life in Tourette Syndrome. Front Psychiatr 7:97. https://doi.org/10.3389/fpsyt.2016.00097
5. Johnson KA, Worbe Y, Foote KD, Butson CR, Gunduz A, Okun MS (2023) Tourette syndrome: clinical features, pathophysiology, and treatment. The Lancet Neurol 22:147-158. https://doi.org/10.1016/S1474-4422(22)00303-9
6. Banaschewski T, Woerner W, Rothenberger A (2003) Premonitory sensory phenomena and suppressibility of tics in Tourette syndrome: developmental aspects in children and adolescents. Dev Med Child Neurol 45:700-703. https://doi.org/10.1017/s0012162203001294
7. Cohen SC, Leckman JF, Bloch MH (2013) Clinical assessment of Tourette syndrome and tic disorders. Neurosci & Biobehav Rev 37:997-1007. https://doi.org/10.1016/j.neubiorev.2012.11.013
8. Brandt VC, Beck C, Sajin V, Baaske MK, Bäumer T, Beste C, Anders S, Münchau A (2016) Temporal relationship between premonitory urges and tics in Gilles de la Tourette syndrome. Cortex 77:24-37. https://doi.org/10.1016/j.cortex.2016.01.008
9. Leckman JF, Bloch MH, Scahill L, King RA (2006) Tourette syndrome: the self under siege. J Child Neurol 21:642-649. https://doi.org/10.1177/08830738060210081001
10. Cavanna AE, Black KJ, Hallett M, Voon V (2017) Neurobiology of the Premonitory Urge in Tourette's Syndrome: Pathophysiology and Treatment Implications. J Neuropsychiatr Clin Neurosci 29:95-104. https://doi.org/10.1176/appi.neuropsych.16070141

11. Brandt V, Essing J, Jakubovski E, Müller-Vahl K (2023) Premonitory urge and tic severity, comorbidities, and quality of life in chronic tic disorders. Mov Disord Clinic Pract 10:922-932. https://doi.org/10.1002/mdc3.13742
12. Edwards MJ, Koens LH, Liepert J, Nonnekes J, Schwingenschuh P, van de Stouwe AMM, Morgante F (2024) Clinical neurophysiology of functional motor disorders: IFCN Handbook Chapter. Clinic Neurophysiol Pract 9:69-77. https://doi.org/10.1016/j.cnp.2023.12.006
13. Cavanna A, Purpura G, Nacinovich R (2022) Neurodevelopmental versus functional tics: The state of the art. Arch Med Health Sci 10:239-246. DOI: 10.4103/amhs.amhs_246_22
14. Heyman I, Liang H, Hedderly T (2021) COVID-19 related increase in childhood tics and tic-like attacks. Arch Dis Child 106:420. https://doi.org/10.1136/archdischild-2021-321748
15. Jack RH, Joseph RM, Coupland CAC, Hall CL, Hollis C (2023) Impact of the COVID-19 pandemic on incidence of tics in children and young people: a population-based cohort study. Eur Clinic Med 57: 101857. https://doi.org/10.1016/j.eclinm.2023.101857
16. Martino D, Hedderly T, Murphy T, Müller-Vahl KR, Dale RC, Gilbert DL, Rizzo R, Hartmann A, Nagy P, Anheim M, Owen T, Malik O, Duncan M, Heyman I, Liang H, McWilliams A, O'Dwyer S, Fremer C, Szejko N, Han VX, Kozlowska K, Pringsheim TM (2023) The spectrum of functional tic-like behaviours: Data from an international registry. Eur J Neurol 30:334-343. https://dx.doi.org/10.1111/ene.15611
17. Ganos C, Martino D, Espay AJ, Lang AE, Bhatia KP, Edwards MJ (2019) Tics and functional tic-like movements: Can we tell them apart? Neurol 93:750-758. https://doi.org/10.1212/wnl.0000000000008372
18. Demartini B, Ricciardi L, Parees I, Ganos C, Bhatia KP, Edwards MJ (2015) A positive diagnosis of functional (psychogenic) tics. Eur J Neurol 22:527-e536. https://doi.org/10.1111/ene.12609
19. Horner O, Hedderly T, Malik O (2022) The changing landscape of childhood tic disorders following COVID-19. Paediatr Child Health 32:363-367. https://doi.org/10.1016/j.paed.2022.07.007
20. Robinson S, Hedderly T (2016) Novel Psychological Formulation and Treatment of "Tic Attacks" in Tourette Syndrome. Front Pediatr 4:46. https://doi.org/10.3389/fped.2016.00046
21. Andersen K, Jensen I, Okkels KB, Skov L, Debes NM (2023) Clarifying the Differences between Patients with Organic Tics and Functional Tic-Like Behaviors. Healthc 11:1481. https://dx.doi.org/10.3390/healthcare11101481

22. Ludlow AK, Anderson S, Robinson S, Owen T, Hedderly T (2024) An investigation into mothers' experiences of their children's functional tic-like behaviour and tic attacks. PLoS One, 19: 0292742. https://doi.org/10.1371/journal.pone.0292742

23. Khalsa SS, Adolphs R, Cameron OG, Critchley HD, Davenport PW, Feinstein JS, Feusner JD, Garfinkel SN, Lane RD, Mehling WE, Meuret AE, Nemeroff CB, Oppenheimer S, Petzschner FH, Pollatos O, Rhudy JL, Schramm LP, Simmons WK, Stein MB, Stephan KE, Van den Bergh O, Van Diest I, von Leupoldt A, Paulus MP, Ainley V, Al Zoubi O, Aupperle R, Avery J, Baxter L, Benke C, Berner L, Bodurka J, Breese E, Brown T, Burrows K, Cha Y-H, Clausen A, Cosgrove K, Deville D, Duncan L, Duquette P, Ekhtiari H, Fine T, Ford B, Garcia Cordero I, Gleghorn D, Guereca Y, Harrison NA, Hassanpour M, Hechler T, Heller A, Hellman N, Herbert B, Jarrahi B, Kerr K, Kirlic N, Klabunde M, Kraynak T, Kriegsman M, Kroll J, Kuplicki R, Lapidus R, Le T, Hagen KL, Mayeli A, Morris A, Naqvi N, Oldroyd K, Pané-Farré C, Phillips R, Poppa T, Potter W, Puhl M, Safron A, Sala M, Savitz J, Saxon H, Schoenhals W, Stanwell-Smith C, Teed A, Terasawa Y, Thompson K, Toups M, Umeda S, Upshaw V, Victor T, Wierenga C, Wohlrab C, Yeh H-w, Yoris A, Zeidan F, Zotev V, Zucker N (2018) Interoception and Mental Health: A Roadmap. Biological Psychiatry: Cogn Neurosci Neuroimaging 3:501-513. https://doi.org/10.1016/j.bpsc.2017.12.004

24. Chen WG, Schloesser D, Arensdorf AM, Simmons JM, Cui C, Valentino R, Gnadt JW, Nielsen L, Hillaire-Clarke CS, Spruance V, Horowitz TS, Vallejo YF, Langevin HM (2021) The Emerging Science of Interoception: Sensing, Integrating, Interpreting, and Regulating Signals within the Self. Trends in Neuroscienc 44:3-16. https://doi.org/10.1016/j.tins.2020.10.007

25. Suksasilp C, Garfinkel SN (2022) Towards a comprehensive assessment of interoception in a multi-dimensional framework. Biol Psychol168:108262. https://doi.org/10.1016/j.biopsycho.2022.108262

26. Schandry R (1981) Heart Beat Perception and Emotional Experience. Psychophysiol 18:483-488. https://doi.org/10.1111/j.1469-8986.1981.tb02486.x

27. Ganos C, Garrido A, Navalpotro-Gómez I, Ricciardi L, Martino D, Edwards MJ, Tsakiris M, Haggard P, Bhatia KP (2015) Premonitory urge to tic in tourette's is associated with interoceptive awareness. Mov Disord 30:1198-1202. https://dx.doi.org/10.1002/mds.26228

28. Narapareddy A, Eckland MR, Riordan HR, Cascio CJ, Isaacs DA (2022) Altered interoceptive sensibility in adults with chronic tic disorder. Front Psychiatr 13:914897. https://doi.org/10.3389/fpsyt.2022.914897

29. Rae CL, Critchley HD, Seth AK (2019) A Bayesian Account of the Sensory-Motor Interactions Underlying Symptoms of Tourette Syndrome. Front Psychiatr 10:29. https://doi.org/10.3389/fpsyt.2019.00029

30. Ricciardi L, Demartini B, Crucianelli L, Krahé C, Edwards MJ, Fotopoulou A (2016) Interoceptive awareness in patients with functional neurological symptoms. Biol Psychol 113:68-74.
31. Ricciardi L, Nisticò V, Andrenelli E, Cunha JM, Demartini B, Kirsch LP, Crucianelli L, Yogarajah M, Morgante F, Fotopoulou A, Edwards MJ (2021) Exploring three levels of interoception in people with functional motor disorders. Parkinsonism & Related Disorders 86:15-18. https://doi.org/10.1016/j.biopsycho.2015.10.009
32. Demartini B, Volpe R, Mattavelli G, Goeta D, D'Agostino A, Gambini O (2019) The neuromodulatory effect of tDCS in patients affected by functional motor symptoms: an exploratory study. Neurol Sci 40:1821-1827. https://doi.org/10.1007/s10072-019-03912-5
33. Millman LSM, Short E, Stanton B, Winston JS, Nicholson TR, Mehta MA, Reinders AATS, Edwards MJ, Goldstein LH, David AS, Hotopf M, Chalder T, Pick S (2023) Interoception in functional motor symptoms and functional seizures: Preliminary evidence of intact accuracy alongside reduced insight and altered sensibility. Behav Res Ther 168:104379. https://doi.org/10.1016/j.brat.2023.104379
34. Pile V, Lau JYF, Topor M, Hedderly T, Robinson S (2018) Interoceptive Accuracy in Youth with Tic Disorders: Exploring Links with Premonitory Urge, Anxiety and Quality of Life. J Autism Dev Disord 48:3474-3482. https://doi.org/10.1007/s10803-018-3608-8
35. Schütteler C, Woitecki K, Döpfner M, Gerlach AL (2023) Interoception and premonitory urges in children and adolescents with tic disorders. Clin Psychol Eur 5:8185. https://doi.org/10.32872/cpe.8185
36. Faul F, Erdfelder E, Lang A-G, Buchner A (2007) G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. Behav Res Methods 39:175-191. https://doi.org/10.3758/bf03193146
37. Wechsler D (2011) Wechsler Abbreviated Scale of Intelligence (WASI-II). Pearson, San Antonio.
38. Derryberry D, Reed MA (2002) Anxiety-related attentional biases and their regulation by attentional control. J Abnorm Psychol 111:225-236. https://psycnet.apa.org/doi/10.1037/0021-843X.111.2.225
39. Muris P, de Jong PJ, Engelen S (2004) Relationships between neuroticism, attentional control, and anxiety disorders symptoms in non-clinical children. Pers Individ Differ 37:789-797. https://psycnet.apa.org/doi/10.1016/j.paid.2003.10.007
40. Melendez R, Bechor M, Rey Y, Pettit JW, Silverman WK (2017) Attentional control scale for children: Factor structure and concurrent validity among children and adolescents referred for anxiety disorders. J Clinil Psychol 73:489-499. https://doi.org/10.1002/jclp.22346

41. Gaffney G, Sieg K, Hellings J (1994) The MOVES: A self-rating scale for Tourette's syndrome. J Chlid Adolesc Psychopharm 4:269-280. https://doi.org/10.1089/cap.1994.4.269
42. Woods DW, Piacentini J, Himle MB, Chang S (2005) Premonitory Urge for Tics Scale (PUTS): initial psychometric results and examination of the premonitory urge phenomenon in youths with Tic disorders. J Dev Behav Pediatr 26:397-403. https://doi.org/10.1097/00004703-200512000-00001
43. Leckman JF, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, Cohen DJ (1989) The Yale Global Tic Severity Scale: Initial Testing of a Clinician-Rated Scale of Tic Severity. J Am Acad Child Adolesc Psychiatr 28:566-573. https://doi.org/10.1097/00004583-198907000-00015
44. Chorpita BF, Moffitt CE, Gray J (2005) Psychometric properties of the Revised Child Anxiety and Depression Scale in a clinical sample. Behav Res Ther 43:309-322. https://doi.org/10.1016/j.brat.2004.02.004
45. Swanson JM (1981) The SNAP Rating Scale for the Diagnosis of the Attention Deficit Disorder.
46. Varni JW, Seid M, Rode CA (1999) The PedsQL: measurement model for the pediatric quality of life inventory. Med Care 37:126-139. https://doi.org/10.1097/00005650-199902000-00003
47. Upton P, Eiser C, Cheung I, Hutchings HA, Jenney M, Maddocks A, Russell IT, Williams JG (2005) Measurement properties of the UK-English version of the Pediatric Quality of Life Inventory™ 4.0 (PedsQL™) generic core scales. Health Qual Life Outcomes 3:1-7. https://doi.org/10.1186/1477-7525-3-22
48. Murphy J, Brewer R, Plans D, Khalsa SS, Catmur C, Bird G (2020) Testing the independence of self-reported interoceptive accuracy and attention. Quart J Exp Psychol 73:115-133. https://doi.org/10.1177/1747021819879826
49. Brand S, Lang K, Bellinghausen C, Witthöft M, Jungmann SM (2024) Interoceptive accuracy in children aged 8 to 13 and their parents: implications for mental health. Cogent Psychol 11:2348043. https://doi.org/10.1080/23311908.2024.2348043
50. Garfinkel SN, Seth AK, Barrett AB, Suzuki K, Critchley HD (2015) Knowing your own heart: distinguishing interoceptive accuracy from interoceptive awareness. Biol Psychol 104:65-74. https://doi.org/10.1016/j.biopsycho.2014.11.004
51. Hart N, McGowan J, Minati L, Critchley HD (2013) Emotional regulation and bodily sensation: interoceptive awareness is intact in borderline personality disorder. J Personal Disord 27:506-518. https://doi.org/10.1521/pedi_2012_26_049

52. Murphy J, Catmur C, Bird G (2018) Alexithymia is associated with a multidomain, multidimensional failure of interoception: Evidence from novel tests. J Exp Psychol Gen 147:398-408. https://doi.org/10.1037/xge0000366

53. Field A (2018) Discovering Statistics Using IMB SPSS Statistics. SAGE, London.

54. Spagnolo PA, Parker JA, Hallett M, Horovitz S (2024) Functional Movement Disorder Is Associated with Abnormal Interoceptive Brain Activity: A Task-based Functional MRI Study. MedRxiv:2024. https://doi.org/10.1101/2024.07.23.24310881

55. Todd J, Swami V, Aspell JE, Furnham A, Horne G, Stieger S (2022) Are some interoceptive sensibility components more central than others? Using item pool visualisation to understand the psychometric representation of interoception. Plos One 17:e0277894. https://doi.org/10.1371/journal.pone.0277894

56. Edwards MJ, Adams RA, Brown H, Pareés I, Friston KJ (2012) A Bayesian account of 'hysteria'. Brain 135:3495-3512. https://doi.org/10.1093/brain/aws129

57. Schaich A, Brandt V, Senft A, Schiemenz C, Klein J-P, Faßbinder E, Münchau A, Alvarez-Fischer D (2020) Treatment of Tourette Syndrome With Attention Training Technique—A Case Series. Front Psychiatr 11:519931. https://doi.org/10.3389/fpsyt.2020.5199312020

58. Sanderson C, Verdellen C, Debes N, Tárnok Z, van de Griendt J, Zimmerman-Brenner S, Murphy T (2024) Addressing co-occurring conditions in behavioural therapy for tic disorders: a review and guideline. Eur Child & Adolesc Psychiatr 33:2111-2127. https://doi.org/10.1007/s00787-022-02097-1

59. Braet J, Giletta M, Desmedt O, Wante L, Rapp L, Pollatos O, Braet C (2024) Should Adolescents Listen to Their Hearts? A Closer Look at the Associations Between Interoception, Emotional Awareness and Emotion Regulation in Adolescents. Psychol Rep:332941241286435.

60. Murphy J, Cheesman R, Gregory AM, Lau J, Ehlers A, Catmur C, Bird G, Eley TC (2019) Estimating the stability of heartbeat counting in middle childhood: A twin study. Biol Psychol 148:107764.

61. Nicholson T, Williams D, Carpenter K, Kallitsounaki A (2019) Interoception is Impaired in Children, But Not Adults, with Autism Spectrum Disorder. J Autism Dev Disord 49:3625-3637.

Chapter 3 Bridging Chapter

Overview of Chronic Tic Disorders and Difficulties in Recruiting Participants with Functional Tic-Like Behaviours

This chapter is formatted to the standards of the school of psychology at the University of Southampton and thus follows APA 7th Referencing.

Word Count: 2500

(excluding references)

3.1 Introduction

Both chapters 1 and 2 of this thesis have been formatted in preparation to undergo peer-review for the Journal of Neurology. This journal frequently publishes articles featuring novel research into chronic tic disorders (CTDs) including Tourette syndrome. Typically, authors do not provide detailed descriptions of CTDs within their articles. This may be because numerous reviews have been published providing detailed backgrounds of CTDs and thus readers are expected to have a certain level of knowledge about these disorders if seeking out these articles. Therefore, I have only included a brief overview of CTDs in chapters 1 and 2 which will be sufficient to set the context of my research and appropriate for readers of the Journal of Neurology. However, it is possible that the examiners for my thesis do not have such an extensive knowledge of CTDs and may not be familiar with terms commonly used to describe specific tics that usually require no explanation within a journal article. Therefore, this chapter aims to provide a more in-depth overview of CTDs including descriptions of commonly reported tics and associated features of the disorders. Moreover, this chapter will also discuss the challenges faced when recruiting for the study described in chapter 2. Possible explanations for recruitment difficulties will be discussed alongside plans for dissemination.

3.2 Overview of Chronic Tic Disorders

3.2.1 Diagnosing Tic Disorders

As described in previous chapters, tics are defined as sudden, rapid, recurrent, and non-rhythmic movements and vocalisations (American Psychiatric Association [APA], 2013). Tics have been documented in patients since the late 19th century, although up until the late 1960s they were considered a symptom of hysteria and often referred to as psychogenic (Jankovic & Kurlan, 2011; Ueda & Black, 2021). In contrast, tics are now considered to have a neurobiological underpinning and are regarded as the most common childhood movement disorder (Ueda & Black, 2021). The diagnostic and statistical manual of mental disorders (DSM-V) recognises five types of tic disorders: Tourette syndrome, persistent (chronic) motor or vocal tic disorders, provisional tic disorder, other specified tic disorder, and unspecified tic disorder (APA, 2013). Tourette syndrome is diagnosed by the presence of multiple motor and vocal tics which persist for at least one year following first tic onset, and develop prior to the age of 18 (APA, 2013). Persistent (chronic) motor or vocal tic disorders require that tic onset occurs before age 18 and persist for at least one year, but only require the presence of either motor or vocal tics, not both, to be diagnosed (APA, 2013). In contrast, provisional tic disorder is diagnosed when tics have been present for less than one year (APA, 2013) and thus is not

considered a CTD. Both other specified and unspecified tic disorders refer to tics that do not reach the above criteria and may be diagnosed when tics have an onset later than age 18 (APA, 2013).

Tics typically develop between the ages of 3 and 8 and reach peak severity at around 11 years old before improving during late adolescence (Freeman et al., 2000; Gill & Kompoliti, 2020). Whilst some patients report reduced frequency or remission of tics in adulthood, a number of longitudinal studies have found evidence that tics persist in 50-80% of cases and will be of greater severity and highly debilitating (Leckman & Bloch, 2015; Reagan et al., 2022). However, it is unclear whether specific factors predict the likelihood of tics continuing with age, and research suggests this may relate to individual differences in brain chemistry and structure (Leckman & Bloch, 2015).

3.2.2 Characteristics of Tics

As aforementioned in chapter 1 and 2, tics are known to wax and wane and are described as suggestible, suppressible, and often accompanied by the presence of a premonitory urge (APA, 2013; Cohen et al., 2013; Johnson et al., 2023; Ueda & Black, 2021), Emotional states such as anxiety and fatigue have been shown to increase tic expression alongside environmental factors that increase stress, such as playing video games and completing schoolwork (Caurín et al., 2014; Conelea & Woods, 2008; Ruhrman et al., 2023). In contrast, participating in sports, concentrating on creative activities, and relaxed states have been found to reduce tics, highlighting the role of contextual factors in tic expression (Caurín et al., 2014).

Tics can be classed as either simple or complex. Simple motor tics are brief movements which require the use of isolated muscle groups, such as eye-blinking (APA, 2013; Johnson et al., 2023). Similarly, simple phonic tics are vocalisations containing syllables or non-words, such as grunting and sniffing (APA, 2013; Johnson et al., 2023). Typically, children first develop simple motor tics in the face which then follow a rostro-caudal progression towards the lower extremities (Leckman & Bloch, 2015). Simple vocal and complex tics often develop a couple of years following first tic onset (Ueda & Black, 2021). Complex motor tics use multiple muscle groups and are often several simple motor tics or coordinated patterns of movement, such as a facial grimace followed by a shoulder shrug, or jumping and spinning (APA, 2013; Johnson et al., 2023). Complex phonic tics are often vocalisations consisting of phrases and words, or a combination of sounds (APA, 2013; Johnson et al., 2023). Moreover, complex tics can include repeating others' actions (echopraxia) or vocalisations (echolalia) alongside mimicking one's own movements (palipraxia) or sounds (palilalia) (APA, 2013; Johnson et al., 2023). Coprophenomena is another variety of complex tics which includes shouting obscene words or

phrases (coprolalia) or making obscene gestures (copropraxia) (APA, 2013; Johnson et al., 2023). Whilst coprolalia is presented as a common feature of Tourette syndrome in the media, studies have found this to be a rare symptom and reportedly affects only 20-25% of people with CTDs (Freeman et al., 2009; Kobierska et al., 2014). Non-obscene socially inappropriate behaviours (NOSIBs) are also considered a subtype of complex tics in which individuals shout insulting or derogatory remarks based on people's physical appearance (e.g. ugly) or display disruptive, unacceptable behaviours such as shouting "bomb" in an airport (Eddy & Cavanna, 2013; Grycz & Janik, 2024). The prevalence of NOSIBs in CTDs is currently unknown as few studies have sought to investigate this phenomenon. In addition, researchers are unclear as to whether NOSIBs are a specific feature of CTDs or are associated with psychiatric comorbidities that feature disruptive behaviours, such as oppositional defiant disorder (Eddy & Cavanna, 2013; Grycz & Janik, 2024). However, Grycz and Janik (2024) found NOSIBs occurred in nearly 25% of a large sample of adults with Tourette syndrome, and onset of NOSIBs occurred at a similar time to tics, suggesting a relationship between the two. Tics can also be referred to as self-injurious and describes repetitive harming behaviours towards oneself, such as biting or hitting (Fischer et al., 2020). A limited number of studies have investigated the prevalence of self-injurious tics in CTDs, but it has been estimated to occur in 14%-17% of patients and in some cases can result in visits to the emergency department (Baizabal-Carvallo et al., 2022; Fischer et al., 2020).

Whilst CTDs are associated with high rates of comorbid obsessive compulsive disorder (OCD), it has been documented that patients with CTDs have specific obsessive compulsive behaviours which are thought to be part of the spectrum of tics (Eddy & Cavanna, 2014). Patients report the need for things to be "just right" and may engage in repetitive forced touching of objects (Eddy & Cavanna, 2014; Ganos et al., 2015). Blocking tics, in which behaviours interfere temporarily with voluntary movement, are thought to be linked with obsessive compulsive behaviours and have been observed in some patients with CTDs, although this is not overly reported (Ganos et al., 2015). It is clear that more research is required to understand how specific tics may link to psychiatric comorbidities to aid our understanding of the prevalence of them within CTDs.

3.2.3 Treatment

Behavioural therapies such as exposure response prevention and habit reversal therapy are commonly used as a treatment for reducing the severity, intensity, and frequency of tics in children with CTDs (Cuenca et al., 2015). Exposure response prevention involves gradually increasing individuals' exposure to premonitory urges whilst resisting the urge to tic, with the aim of habituating to the uncomfortable sensations resulting in reduced tic expression

(Hoogduin et al., 1997; van de Griendt et al., 2013). Alternatively, habit reversal therapy targets specific tics by developing a competing response which aims to block the tic until the premonitory urge that preceded the tic lessens (Azrin & Nunn, 1973; van de Griendt et al., 2013). Whilst research suggests both these therapies can be an effective intervention for children with CTDs, they do not work for all individuals (Frank & Cavanna, 2013; Whittington et al., 2016). This may be due to both therapies requiring an awareness of premonitory urges which, as aforementioned, are not present in all children with tics, particularly those under the age of 10 (Johnson et al., 2023). This highlights the importance of my research investigating interoceptive processes in young people with CTDs, and functional tic-like behaviours (FTLB), to help identify alternative therapies which may be more accessible for all patients with these disorders (Liu et al., 2020).

3.3 Reflecting on the Empirical Study

3.3.1 Challenges with Recruitment

Originally, the plan for the empirical study was to recruit four groups of participants, rather than the three described in Chapter 2. We sought to have the following groups: young people with CTDs only, young people with FTLB only, young people with both a CTD and FTLB, and controls with no diagnosis or history of tics. We aimed to separate participants with FTLB and comorbid CTDs from participants with FTLB only. This was because the literature is still uncertain as to whether FTLB are entirely separate from CTDs, especially as a functional overlay between the two has been identified (Cavanna et al., 2022). Thus, separating the groups would have enabled us to control for comorbid CTDs as a possible confound and investigate whether differences in interoceptive abilities were observed in participants with FTLB only or with comorbid CTDs. However, it became clear early in the recruitment process that we may struggle to find enough participants for both of the proposed FTLB groups, as young people with this diagnosis expressed little interest. Thus, we decided to merge the two FTLB groups to include participants with and without comorbid CTDs in the hopes of improving our chances of recruiting more young people for our study. Unfortunately, despite recruitment being open for six months, we were still unable to recruit an adequate sample size of young people with a diagnosis of FTLB, which seriously impacted the validity and reliability of our results.

One explanation for the lack of responses received from young people with a diagnosis of FTLB may be due to the age range specified within our study. Research has found onset of FTLB is higher in comparison to CTDs, and FTLB usually occur in adolescence (Pringsheim et al., 2023). This, coupled with findings that report young people are having to wait lengthy times

before receiving an assessment, indicates that by the time young people receive a diagnosis of FTLB they may be in late adolescence or early adulthood (Burn et al., 2025). This highlights the importance of timely diagnosis and the need for research to consider extending age ranges to account for the delays in patients receiving diagnoses of FTLB. Thus, it is possible more people were interested in participating in our study, but due to 17 years old being the maximum age we were recruiting, individuals who received a diagnosis later than this would have been unable to participate and would not respond to advertisements. Future research may have to consider including young adults with FTLB when investigating young people with this disorder to help recruit larger sample sizes.

Another explanation for the difficulties in recruiting participants with FTLB may be due to the various terminologies used to describe this phenomenon (Demartini et al., 2015). The advertisement for our study stated participants must have a diagnosis of FTLB and did not include any other terminologies associated with this diagnosis, such as functional tics or TikTok tics (Demartini et al., 2015; Müller-Vahl et al., 2022). Thus, it is possible that young people with FTLB may have been unaware they could participate as they could have received a different term to describe their symptoms at diagnosis. This indicates the importance of clinicians agreeing a specific terminology and consistently using this when diagnosing individuals to provide more clarity to patients, as well as improving researcher's ability to investigate this phenomenon.

Moreover, the reported difficulties in distinguishing between FTLB and CTDs suggests it is possible that patients have been misdiagnosed by professionals (Amorelli et al., 2022). This was observed within our study as $n = 5$ of $N = 7$ participants in the FTLB group had responded to the study's advertisement as a potential participant for the CTD group. Whilst the researchers could not diagnose these participants with FTLB, they did inform participants that it is possible that some of their symptoms are congruent with features of FTLB. The research team felt it was the ethical choice to share this information with participants (and their guardian if the participant was under 16 years old) for multiple reasons. Firstly, evidence suggests that the earlier functional symptoms are identified, the better the chances of recovery are for patients as they are able to access psychoeducation and individualised interventions to improve symptoms (Malaty et al., 2022; Vassilopoulos et al. 2022). Secondly, by informing them of this possibility, participants could make an informed choice to explore this further with their medical professional if they wished. This may enable them to consider different treatment options, which would be important as interventions for CTDs have been found to be ineffective in treating FTLB (Amorelli et al., 2022; Malaty et al., 2022). Despite these participants not having a confirmed diagnosis of FTLB by their medical professional, we chose to include them in the FTLB group to ensure they did not confound the CTD group. It is possible that young people who

saw the study advertised may have FTLB but they are unaware and undiagnosed, further highlighting that interest and recruitment for this study could have been higher if professionals were more certain of the differences between CTDs and FTLB.

3.3.2 Plans For Dissemination

In order to write up the study findings in time for thesis submission, we had to end recruitment for the study. Despite chapter 2 being written to meet the standards of peer-review, we do not intend to submit to the journal immediately due to the small sample size and underpowered findings. Instead, we plan to continue recruiting participants for the mixed FTLB group until we acquire enough for our sample to be adequately powered. This will enable us to draw firmer conclusions about our results, and aid future research and clinical practice more clearly. Once data is analysed, we will edit the paper with the updated sample and then submit for peer-review to the Journal of Neurology.

3.4 References

1. American Psychiatric Association (2013) Somatic Sympton and Related Disorders. In: Diagnostic and Statistical Manual of Mental Disorders.
2. Kanaan RAA (2016) Chapter 4 - Freud's hysteria and its legacy. In: Hallett M, Stone J, Carson A (eds) Handbook of Clinical Neurology. Elsevier, pp 37-44
3. Park JE (2024) Functional Movement Disorders: Updates and Clinical Overview. J Mov Disord 17:251-261.
4. Espay AJ, Aybek S, Carson A, Edwards MJ, Goldstein LH, Hallett M, LaFaver K, LaFrance WC, Jr., Lang AE, Nicholson T, Nielsen G, Reuber M, Voon V, Stone J, Morgante F (2018) Current Concepts in Diagnosis and Treatment of Functional Neurological Disorders. JAMA Neurol 75:1132-1141.
5. Ludwig L, Pasman JA, Nicholson T, Aybek S, David AS, Tuck S, Kanaan RA, Roelofs K, Carson A, Stone J (2018) Stressful life events and maltreatment in conversion (functional neurological) disorder: systematic review and meta-analysis of case-control studies. Lancet Psychiatry 5:307-320.
6. American Psychiatric Association (2013) Neurodevelopmental disorders. In: Diagnostic and statistical manual of mental disorders.
7. Leckman JF, Peterson BS, Pauls DL, Cohen DJ (1997) TIC DISORDERS* *This article is adapted from Leckman JF, Cohen DJ: Tic Disorders. In Rutter M, Taylor E, Hersov L (eds): Child and Adolescent Psychiatry: Modern Approaches, ed 3. London, Blackwell Scientific, 1994, pp 455–466; with permission. Psychiatric Clinics of North America 20:839-861. https://www.sciencedirect.com/science/article/pii/S0193953X05703488
8. Freeman RD, Fast DK, Burd L, Kerbeshian J, Robertson MM, Sandor P (2000) An international perspective on Tourette syndrome: selected findings from 3,500 individuals in 22 countries. Dev Med Child Neurol 42:436-447.
9. Ueda K, Black KJ (2021) A Comprehensive Review of Tic Disorders in Children. Journal of Clinical Medicine 10:2479. https://www.mdpi.com/2077-0383/10/11/2479
10. Eapen V, Cavanna AE, Robertson MM (2016) Comorbidities, Social Impact, and Quality of Life in Tourette Syndrome. Front Psychiatry 7:97.
11. Gill CE, Kompoliti K (2020) Clinical Features of Tourette Syndrome. J Child Neurol 35:166-174.
12. Leckman JF, Bloch MH, Scahill L, King RA (2006) Tourette syndrome: the self under siege. J Child Neurol 21:642-649.
13. Banaschewski T, Woerner W, Rothenberger A (2003) Premonitory sensory phenomena and suppressibility of tics in Tourette syndrome: developmental aspects in children and adolescents. Developmental Medicine & Child Neurology 45:700-703. https://onlinelibrary.wiley.com/doi/abs/10.1111/j.1469-8749.2003.tb00873.x
14. Brandt VC, Beck C, Sajin V, Baaske MK, Bäumer T, Beste C, Anders S, Münchau A (2016) Temporal relationship between premonitory urges and tics in Gilles de la Tourette syndrome. Cortex 77:24-37. https://www.sciencedirect.com/science/article/pii/S0010945216000186
15. Hirschtritt ME, Lee PC, Pauls DL, Dion Y, Grados MA, Illmann C, King RA, Sandor P, McMahon WM, Lyon GJ, Cath DC, Kurlan R, Robertson MM, Osiecki L, Scharf JM, Mathews CA (2015) Lifetime prevalence, age of risk, and genetic relationships of comorbid psychiatric disorders in Tourette syndrome. JAMA Psychiatry 72:325-333.
16. Pringsheim T, Ganos C, Nilles C, Cavanna AE, Gilbert DL, Greenberg E, Hartmann A, Hedderly T, Heyman I, Liang H, Malaty I, Malik O, Debes NM, Vahl KM, Munchau A, Murphy T, Nagy P, Owen T, Rizzo R, Skov L, Stern J, Szejko N, Worbe Y, Martino D (2023) European Society for the Study of Tourette Syndrome 2022 criteria for clinical diagnosis of functional tic-like behaviours: International consensus from experts in tic disorders. European Journal of Neurology 30:902-910. https://onlinelibrary.wiley.com/doi/abs/10.1111/ene.15672

17. Demartini B, Ricciardi L, Parees I, Ganos C, Bhatia KP, Edwards MJ (2015) A positive diagnosis of functional (psychogenic) tics. European journal of neurology 22:527. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=25487253&site=ehost-live
18. Müller-Vahl KR, Pisarenko A, Jakubovski E, Fremer C (2022) Stop that! It's not Tourette's but a new type of mass sociogenic illness. Brain : a journal of neurology 145:476-480. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34424292&site=ehost-live
19. Robinson S, Hedderly T (2016) Novel Psychological Formulation and Treatment of "Tic Attacks" in Tourette Syndrome. Front Pediatr 4:46.
20. Heyman I, Liang H, Hedderly T (2021) COVID-19 related increase in childhood tics and tic-like attacks. Arch Dis Child 106:420-421.
21. Amorelli G, Martino D, Pringsheim T (2022) Rapid Onset Functional Tic-Like Disorder Outbreak: A Challenging Differential Diagnosis in the COVID-19 Pandemic. Journal of the Canadian Academy of Child & Adolescent Psychiatry 31:144-181. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cul&AN=157905036&site=ehost-live
22. Owen T, Silva J, Grose C, Bailey A, Robinson S, Anderson S, Ludlow A, Sharp S, Toghill L, Hedderly T (2022) Case report: Advice for schools on managing functional tic-like behaviours. Frontiers in Psychiatry Volume 13 - 2022https://www.frontiersin.org/journals/psychiatry/articles/10.3389/fpsyt.2022.1001459
23. Rigas A, Mainka T, Pringsheim T, Münchau A, Malaty I, Worbe Y, Cavanna AE, Lees AJ, Lang AE, Martino D, Ganos C (2023) Distinguishing functional from primary tics: a study of expert video assessments. J Neurol Neurosurg Psychiatry 94:751-756.
24. Ganos C, Martino D, Espay AJ, Lang AE, Bhatia KP, Edwards MJ (2019) Tics and functional tic-like movements: Can we tell them apart? Neurology 93:750-758.
25. Cavanna AE, Damodaran L, Purpura G, Nacinovich R (2022) Tourette Syndrome with Functional Overlay: A Case Series. Archives of Medicine and Health Sciences 10:312-316. https://journals.lww.com/armh/fulltext/2022/10020/tourette_syndrome_with_functional_overlay__a_case.33.aspx
26. Andersen K, Cavanna AE, Szejko N, Müller-Vahl KR, Hedderly T, Skov L, Mol Debes N (2024) A Critical Examination of the Clinical Diagnosis of Functional Tic-like Behaviors. Mov Disord Clin Pract 11:1065-1071.
27. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, Shamseer L, Tetzlaff JM, Akl EA, Brennan SE, Chou R, Glanville J, Grimshaw JM, Hróbjartsson A, Lalu MM, Li T, Loder EW, Mayo-Wilson E, McDonald S, McGuinness LA, Stewart LA, Thomas J, Tricco AC, Welch VA, Whiting P, Moher D (2021) The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 372:n71. https://www.bmj.com/content/bmj/372/bmj.n71.full.pdf
28. Kmet LM, Cook LS, Lee RC (2004) Standard quality assessment criteria for evaluating primary research papers from a variety of fields.
29. Mayo-Wilson E, Li T, Fusco N, Dickersin K (2018) Practical guidance for using multiple data sources in systematic reviews and meta-analyses (with examples from the MUDS study). Res Synth Methods 9:2-12.
30. Andersen K, Jensen I, Okkels KB, Skov L, Debes NM (2023) Clarifying the Differences between Patients with Organic Tics and Functional Tic-Like Behaviors. Healthcare (Basel, Switzerland) 11https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37239767&site=ehost-live
31. Berg L, Martino D, L'Erario ZP, Pringsheim T (2024) Symptom Severity and Health Impacts of Functional Tic-Like Behaviors in Youth. Pediatric neurology 155:68-75. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38603984&site=ehost-live

32. Berg L, Pringsheim TM, Lerario M, Martino D (2024) Psychological Factors Associated with Functional Tic-like Behaviours during the COVID-19 Pandemic. Research on child and adolescent psychopathology 52:1157-1172. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38427218&site=ehost-live
33. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) Functional tics: Expanding the phenotypes of functional movement disorders? European journal of neurology 30:3353-3356. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37410535&site=ehost-live
34. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) New-onset functional tics during the COVID-19 pandemic: Clinical characteristics of 105 cases from a single centre. European journal of neurology 30:2411-2417. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37183565&site=ehost-live
35. Cavanna AE, Purpura G, Riva A, Nacinovich R, Seri S (2023) Neurodevelopmental versus functional tics: A controlled study. Journal of the neurological sciences 451:120725. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37421881&site=ehost-live
36. Fremer C, Szejko N, Pisarenko A, Haas M, Laudenbach L, Wegener C, Müller-Vahl KR (2022) Mass social media-induced illness presenting with Tourette-like behavior. Frontiers in psychiatry 13:963769. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36203825&site=ehost-live
37. Fremer C, Szejko N, Pisarenko A, Haas M, Laudenbach L, Wegener C, Müller-Vahl KR (2024) What distinguishes patients with mass social media-induced illness presenting with Tourette-like behavior from those with Tourette syndrome? Results of a prospective cohort study. European archives of psychiatry and clinical neuroscience 274:227-233. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37209130&site=ehost-live
38. Ganos C, Edwards MJ, Müller-Vahl K (2016) "I swear it is Tourette's!": On functional coprolalia and other tic-like vocalizations. Psychiatry research 246:821-826. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=27825787&site=ehost-live
39. Howlett M, Martino D, Nilles C, Pringsheim T (2022) Prognosis of rapid onset functional tic-like behaviors: Prospective follow-up over 6 months. Brain and behavior 12:e2606. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=35593445&site=ehost-live
40. Pringsheim T, Ganos C, McGuire JF, Hedderly T, Woods D, Gilbert DL, Piacentini J, Dale RC, Martino D (2021) Rapid Onset Functional Tic-Like Behaviors in Young Females During the COVID-19 Pandemic. Movement disorders : official journal of the Movement Disorder Society 36:2707-2713. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34387394&site=ehost-live
41. Pringsheim T, Martino D (2021) Rapid onset of functional tic-like behaviours in young adults during the COVID-19 pandemic. European journal of neurology 28:3805-3808. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34293224&site=ehost-live
42. Janik P, Milanowski Ł, Szejko N (2014) Psychogenic tics: Clinical characteristics and prevalence. Psychiatria Polska 48:835-845. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=psyh&AN=2014-44507-015&site=ehost-live
43. Martino D, Hedderly T, Murphy T, Müller-Vahl KR, Dale RC, Gilbert DL, Rizzo R, Hartmann A, Nagy P, Anheim M, Owen T, Malik O, Duncan M, Heyman I, Liang H, McWilliams A, O'Dwyer S, Fremer C, Szejko N, Han VX, Kozlowska K, Pringsheim TM (2023) The

spectrum of functional tic-like behaviours: Data from an international registry. European journal of neurology 30:334-343. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36282623&site=ehost-live

44. Maxwell A, Zouki J-J, Eapen V (2023) Integrated cognitive behavioral intervention for functional tics (I-CBiT): case reports and treatment formulation. Frontiers in pediatrics 11:1265123. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38034832&site=ehost-live
45. Müller-Vahl KR, Pisarenko A, Fremer C, Haas M, Jakubovski E, Szejko N (2024) Functional Tic-Like Behaviors: A Common Comorbidity in Patients with Tourette Syndrome. Movement disorders clinical practice 11:227-237. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38468554&site=ehost-live
46. Nilles C, Martino D, Berg L, Fletcher J, Pringsheim T (2024) What are the Key Phenomenological Clues to Diagnose Functional Tic-Like Behaviors in the Pandemic Era? Movement disorders clinical practice 11:398-402. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38269641&site=ehost-live
47. Nilles C, Szejko N, Martino D, Pringsheim T (2024) Prospective follow-up study of youth and adults with onset of functional tic-like behaviours during the COVID-19 pandemic. European journal of neurology 31:e16051. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37644767&site=ehost-live
48. Paulus T, Bäumer T, Verrel J, Weissbach A, Roessner V, Beste C, Münchau A (2021) Pandemic Tic-like Behaviors Following Social Media Consumption. Movement disorders : official journal of the Movement Disorder Society 36:2932-2935. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34558735&site=ehost-live
49. Armstrong-Javors A, Realbuto E, Dy-Hollins ME, Scharf JM (2024) Increase in Functional Tic Presentations in Sexual Orientation and Gender Identity Minority Youth During Coronavirus Disease 2019. Pediatric neurology 155:182-186. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38677240&site=ehost-live
50. Buts S, Duncan M, Owen T, Martino D, Pringsheim T, Byrne S, McWilliams A, Murphy T, Malik O, Liang H, Heyman I, Hedderly T (2022) Paediatric tic-like presentations during the COVID-19 pandemic. Archives of disease in childhood 107:e17. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34824091&site=ehost-live
51. Duncan M, Pearman Z, Harrold K, Warren A, Evans S, McAllister E, Heyman I, Shavel-Jessop S, Murphy T, Liang H (2024) Evaluation of a psychoeducation group for children presenting with functional tic-like behaviours. Clinical child psychology and psychiatry 29:1011-1025. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38476087&site=ehost-live
52. Ducroizet A, Eccles C, Lancaster R, Kowalczyk A, Owen T, Sopena S, Malik O, Hedderly T (2025) Outcomes of functional tics in adolescents: a single-centre tertiary study. Archives of disease in childhoodhttps://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=39880594&site=ehost-live
53. Firestone MJ, Holzbauer S, Conelea C, Danila R, Smith K, Bitsko RH, Klammer SM, Gingerich S, Lynfield R (2023) Rapid onset of functional tic-like behaviors among adolescent girls-Minnesota, September-November 2021. Frontiers in neurology 13:1063261.

https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36742058&site=ehost-live
54. Han VX, Kozlowska K, Kothur K, Lorentzos M, Wong WK, Mohammad SS, Savage B, Chudleigh C, Dale RC (2022) Rapid onset functional tic-like behaviours in children and adolescents during COVID-19: Clinical features, assessment and biopsychosocial treatment approach. Journal of paediatrics and child health 58:1181-1187. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=35247213&site=ehost-live
55. Hull M, Parnes M (2021) Tics and TikTok: Functional Tics Spread Through Social Media. Movement disorders clinical practice 8:1248-1252. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=34765689&site=ehost-live
56. Larsh TR, Wu SW, Gilbert DL (2022) Comparison of Impairment in Functional Tic Disorders Versus Tourette Syndrome. Pediatric neurology 134:83-84. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=35843129&site=ehost-live
57. Mathew A, Abu Libdeh A, Patrie J, Garris J (2023) Outcome in Pediatric Functional Tic Disorders Diagnosed During the COVID-19 Pandemic. The Journal of neuropsychiatry and clinical neurosciences 35:393-397. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37259545&site=ehost-live
58. Okkels KB, Skov L, Klansø S, Aaslet L, Grejsen J, Reenberg A, Sørensen CB, Debes NMMM (2023) Increased Number of Functional Tics Seen in Danish Adolescents during the COVID-19 Pandemic. Neuropediatrics 54:113-119. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36417931&site=ehost-live
59. Prato A, Saia F, Milana MC, Scerbo M, Barone R, Rizzo R (2023) Functional tic-like behaviours during the COVID-19 pandemic: Follow-up over 12 months. Frontiers in pediatrics 10:1003825. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36699317&site=ehost-live
60. Tomczak KK, Worhach J, Rich M, Swearingen Ludolph O, Eppling S, Sideridis G, Katz TC (2024) Time is ticking for TikTok tics: A retrospective follow-up study in the post-COVID-19 isolation era. Brain and behavior 14:e3451. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38468457&site=ehost-live
61. Trau SP, Quehl L, Tsujimoto THM, Lin F-C, Singer HS (2022) Creating a Patient-Based Diagnostic Checklist for Functional Tics During the COVID-19 Pandemic. Neurology Clinical practice 12:365-376. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=36380888&site=ehost-live
62. Baizabal-Carvallo JF, Alonso-Juarez M, Jankovic J (2023) Contrasting features between Tourette syndrome and secondary tic disorders. Journal of neural transmission (Vienna, Austria : 1996) 130:931-936. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=37117738&site=ehost-live
63. Baizabal-Carvallo JF, Jankovic J (2014) The clinical features of psychogenic movement disorders resembling tics. Journal of Neurology, Neurosurgery & Psychiatry 85:573-575. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cul&AN=103928096&site=ehost-live
64. Szejko N, Fletcher J, Martino D, Pringsheim T (2024) Premonitory Urge in Patients with Tics and Functional Tic-like Behaviors. Movement disorders clinical practice 11:276-281. https://soton.idm.oclc.org/login?url=https://search.ebscohost.com/login.aspx?direct=true&db=cmedm&AN=38468546&site=ehost-live
65. Fahn S, Williams DT (1988) Psychogenic dystonia. Adv Neurol 50:431-455.

66. Robertson MM, Eapen V (1996) The National Hospital Interview Schedule for the assessment of Gilles de la Tourette Syndrome. International Journal of Methods in Psychiatric Research 6:203-226.
67. Shaffer D, Gould MS, Brasic J, Ambrosini P, Fisher P, Bird H, Aluwahlia S (1983) A children's global assessment scale (CGAS). Archives of General psychiatry 40:1228-1231.
68. Leckman JF, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, Cohen DJ (1989) The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity. J Am Acad Child Adolesc Psychiatry 28:566-573.
69. Graham P (2023) Transgender children and young people: how the evidence can point the way forward. BJPsych Bull 47:98-104.
70. Lerario MP, Fusunyan M, Stave CD, Roldán V, Keuroghlian AS, Turban J, Perez DL, Maschi T, Rosendale N (2023) Functional neurological disorder and functional somatic syndromes among sexual and gender minority people: A scoping review. Journal of Psychosomatic Research 174:111491. https://www.sciencedirect.com/science/article/pii/S0022399923003483
71. Vindegaard N, Benros ME (2020) COVID-19 pandemic and mental health consequences: Systematic review of the current evidence. Brain, Behavior, and Immunity 89:531-542. https://www.sciencedirect.com/science/article/pii/S0889159120309545
72. Janssen-Aguilar R, Galíndez-de la Portilla JP, Gómez-Alcorta R, Hernández-Palestina M, Mendez-Dominguez N, Ruiz-Chow A, Crail-Meléndez D (2022) Impact of the COVID-19 pandemic on the incidence of patients with functional neurological disorder seen in a neurological emergency department. Psychiatry Clin Neurosci 76:595-596.
73. Berg L, Pringsheim T, Martino D (2022) Sudden Onset Tic and Tic-Like Presentations in Older Adolescents and Adults. Current Developmental Disorders Reports 9:1-10.
74. Kauhanen L, Wan Mohd Yunus WMA, Lempinen L, Peltonen K, Gyllenberg D, Mishina K, Gilbert S, Bastola K, Brown JS, Sourander A (2023) A systematic review of the mental health changes of children and young people before and during the COVID-19 pandemic. European child & adolescent psychiatry 32:995-1013.
75. Cavanna AE, Spini L, Ferrari S, Purpura G, Riva A, Nacinovich R, Seri S (2024) Functional Tic-like Behaviors: From the COVID-19 Pandemic to the Post-Pandemic Era. Healthcare (Basel) 12
76. Malaty IA, Anderson S, Bennett SM, Budman CL, Coffey BJ, Coffman KA, Greenberg E, McGuire JF, Müller-Vahl KR, Okun MS, Quezada J, Robichaux-Viehoever A, Black KJ (2022) Diagnosis and Management of Functional Tic-Like Phenomena. J Clin Med 11
77. Brandt V, Essing J, Jakubovski E, Müller-Vahl K (2023) Premonitory Urge and Tic Severity, Comorbidities, and Quality of Life in Chronic Tic Disorders. Movement Disorders Clinical Practice 10:922-932. https://movementdisorders.onlinelibrary.wiley.com/doi/abs/10.1002/mdc3.13742
78. Srour M, Lespérance P, Richer F, Chouinard S (2008) Psychopharmacology of tic disorders. J Can Acad Child Adolesc Psychiatry 17:150-159.
79. Nilles C, Pringsheim TM, Martino D (2022) The recent surge of functional movement disorders: social distress or greater awareness? Curr Opin Neurol 35:485-493.
80. Vassilopoulos A, Mohammad S, Dure L, Kozlowska K, Fobian AD (2022) Treatment Approaches for Functional Neurological Disorders in Children. Curr Treat Options Neurol 24:77-97.

Appendix A Author Guidelines for the Journal of Neurology

Types of manuscripts and formal requirements

- **Original Communication** must not exceed 9,000 words (excluding abstract and keywords, figures, tables, captions and references). Exceptions can be made only with the agreement of the responsible Chief Editor.
- **Review Articles** must not exceed 12,000 words (excluding abstract and keywords, figures, tables, captions and references). Exceptions can be made only with the agreement of the responsible Editor.

Manuscript Submission

Submission of a manuscript implies: that the work described has not been published before; that it is not under consideration for publication anywhere else; that its publication has been approved by all co-authors, if any, as well as by the responsible authorities – tacitly or explicitly – at the institute where the work has been carried out. The publisher will not be held legally responsible should there be any claims for compensation.

Permissions

Authors wishing to include figures, tables, or text passages that have already been published elsewhere are required to obtain permission from the copyright owner(s) for both the print and online format and to include evidence that such permission has been granted when submitting their papers. Any material received without such evidence will be assumed to originate from the authors.

Online Submission

Please follow the hyperlink “Submit manuscript” and upload all of your manuscript files following the instructions given on the screen.

Source Files

Please ensure you provide all relevant editable source files at every submission and revision. Failing to submit a complete set of editable source files will result in your article not being considered for review. For your manuscript text please always submit in common word processing formats such as .docx or LaTeX.

Title Page

Please make sure your title page contains the following information.

Title

The title should be concise and informative.

Author information

- The name(s) of the author(s)
- The affiliation(s) of the author(s), i.e. institution, (department), city, (state), country
- A clear indication and an active e-mail address of the corresponding author
- If available, the 16-digit ORCID of the author(s)

If address information is provided with the affiliation(s) it will also be published.

Abstract

Please provide an abstract of 150 to 250 words. The abstract should not contain any undefined abbreviations or unspecified references.

For life science journals only (when applicable)

- Trial registration number and date of registration for prospectively registered trials
- Trial registration number and date of registration, followed by "retrospectively registered", for retrospectively registered trials

Keywords

Please provide 4 to 6 keywords which can be used for indexing purposes.

Text Formatting

Manuscripts should be submitted in Word.

- Use a normal, plain font (e.g., 10-point Times Roman) for text.
- Use italics for emphasis.
- Use the automatic page numbering function to number the pages.
- Do not use field functions.
- Use tab stops or other commands for indents, not the space bar.
- Use the table function, not spreadsheets, to make tables.
- Use the equation editor or MathType for equations.
- Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).

Headings

Please use no more than three levels of displayed headings.

Abbreviations

Abbreviations should be defined at first mention and used consistently thereafter.

Footnotes

Footnotes can be used to give additional information, which may include the citation of a reference included in the reference list. They should not consist solely of a reference citation, and they should never include the bibliographic details of a reference. They should also not contain any figures or tables.

Footnotes to the text are numbered consecutively; those to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data). Footnotes to the title or the authors of the article are not given reference symbols.

Always use footnotes instead of endnotes.

Acknowledgments

Acknowledgments of people, grants, funds, etc. should be placed in a separate section on the title page. The names of funding organizations should be written in full.

Scientific style

Generic names of drugs and pesticides are preferred; if trade names are used, the generic name should be given at first mention.

References

Citation

Reference citations in the text should be identified by numbers in square brackets.

Reference list

The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text.

The entries in the list should be numbered consecutively.

If available, please always include DOIs as full DOI links in your reference list (e.g. “https://doi.org/abc”).

- Journal article

Gamelin FX, Baquet G, Berthoin S, Thevenet D, Nourry C, Nottin S, Bosquet L (2009) Effect of high intensity intermittent training on heart rate variability in prepubescent children. Eur J Appl Physiol 105:731-738. https://doi.org/10.1007/s00421-008-0955-8

Ideally, the names of all authors should be provided, but the usage of “et al” in long author lists will also be accepted:

Smith J, Jones M Jr, Houghton L et al (1999) Future of health insurance. N Engl J Med 965:325–329

- Article by DOI

Slifka MK, Whitton JL (2000) Clinical implications of dysregulated cytokine production. J Mol Med. https://doi.org/10.1007/s001090000086

- Book

South J, Blass B (2001) The future of modern genomics. Blackwell, London

- Book chapter

Brown B, Aaron M (2001) The politics of nature. In: Smith J (ed) The rise of modern genomics, 3rd edn. Wiley, New York, pp 230-257

- Online document

Cartwright J (2007) Big stars have weather too. IOP Publishing PhysicsWeb. http://physicsweb.org/articles/news/11/6/16/1. Accessed 26 June 2007

- Dissertation

Trent JW (1975) Experimental acute renal failure. Dissertation, University of California

Always use the standard abbreviation of a journal’s name according to the ISSN List of Title Word Abbreviations

Tables

- All tables are to be numbered using Arabic numerals.
- Tables should always be cited in text in consecutive numerical order.
- For each table, please supply a table caption (title) explaining the components of the table.

- Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.
- Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.

Figure Lettering

- To add lettering, it is best to use Helvetica or Arial (sans serif fonts).
- Keep lettering consistently sized throughout your final-sized artwork, usually about 2–3 mm (8–12 pt).
- Variance of type size within an illustration should be minimal, e.g., do not use 8-pt type on an axis and 20-pt type for the axis label.
- Avoid effects such as shading, outline letters, etc.
- Do not include titles or captions within your illustrations.

Figure Numbering

- All figures are to be numbered using Arabic numerals.
- Figures should always be cited in text in consecutive numerical order.
- Figure parts should be denoted by lowercase letters (a, b, c, etc.).
- If an appendix appears in your article and it contains one or more figures, continue the consecutive numbering of the main text. Do not number the appendix figures,"A1, A2, A3, etc." Figures in online appendices [Supplementary Information (SI)] should, however, be numbered separately.

Figure Captions

- Each figure should have a concise caption describing accurately what the figure depicts. Include the captions in the text file of the manuscript, not in the figure file.
- Figure captions begin with the term Fig. in bold type, followed by the figure number, also in bold type.
- No punctuation is to be included after the number, nor is any punctuation to be placed at the end of the caption.
- Identify all elements found in the figure in the figure caption; and use boxes, circles, etc., as coordinate points in graphs.
- Identify previously published material by giving the original source in the form of a reference citation at the end of the figure caption.

Figure Placement and Size

- Figures should be submitted within the body of the text. Only if the file size of the manuscript causes problems in uploading it, the large figures should be submitted separately from the text.
- When preparing your figures, size figures to fit in the column width.
- For large-sized journals the figures should be 84 mm (for double-column text areas), or 174 mm (for single-column text areas) wide and not higher than 234 mm.
- For small-sized journals, the figures should be 119 mm wide and not higher than 195 mm.

Supplementary Information (SI)

Springer accepts electronic multimedia files (animations, movies, audio, etc.) and other supplementary files to be published online along with an article or a book chapter. This feature can add dimension to the author's article, as certain information cannot be printed or is more convenient in electronic form.

Before submitting research datasets as Supplementary Information, authors should read the journal's Research data policy. We encourage research data to be archived in data repositories wherever possible.

Submission

- Supply all supplementary material in standard file formats.
- Please include in each file the following information: article title, journal name, author names; affiliation and e-mail address of the corresponding author.
- To accommodate user downloads, please keep in mind that larger-sized files may require very long download times and that some users may experience other problems during downloading.

Numbering

- If supplying any supplementary material, the text must make specific mention of the material as a citation, similar to that of figures and tables.
- Refer to the supplementary files as "Online Resource", e.g., "... as shown in the animation (Online Resource 3)", "... additional data are given in Online Resource 4".

Appendix B Ethics Application Form

ERGO II Ethics application form – Psychology Committee

1. Applicant Details

1.1 Applicant name	Kayleigh Maclellan
1.2 Supervisor	**Dr Valerie Brandt** V.C.Brandt@soton.ac.uk
1.3 Other researchers / collaborators (if applicable): *Name, address, email*	**Dr Tammy Hedderly** tammy.hedderly@gstt.nhs.uk **Dr Tamsin Owen** Tamsin.owen@gstt.nhs.uk

2. Study Details

2.1 Title of study	The association between interoception, tics, anxiety, and quality of life in young people with Tourette Syndrome/Chronic Tic Disorders (TS/CTD) and functional tic-like behaviours (FTLB)
2.2 Type of project (e.g. undergraduate, Masters, Doctorate, staff)	Doctorate

2.3 Briefly describe the rationale for carrying out this project and its specific aims and objectives.
Tourette Syndrome/Chronic Tic Disorders (TS/CTD) are neuropsychiatric developmental disorders characterised by repetitive involuntary movements and vocalisations, called vocal and motor tics (Ganos & Martino, 2015). Individuals with TS/CTD describe an uncomfortable physical sensation preceding tics (premonitory urge) which reduces following tic expression. TS/CTD is more common in males that in females (3-4:1). First symptoms typically occur around the age of 4-6, and typically affect the face first, such as

simple eye blinking tics. TS/CTD is associated with anxiety (Frank et al., 2011), reduced quality of life (Eapen et al., 2016), and can impact on social and school functioning. Current management includes pharmacological and behavioural interventions.

During the COVID-19 pandemic, specialist tic clinics saw a significant increase in the number of children presenting with functional tic-like behaviours (FTLB) in their services (Martino et al.). “Functional tics” are not considered to have neurological correlates like tics, and may be an expression of high stress. In contrast to the tics observed in Tourette syndrome, the ‘tics’ observed in functional tic-like behaviours appear more rhythmic and severe, develop rapidly, affect more females than males, and occur without a premonitory urge, or very unusual urges (e.g. “feels like lightening”). Tourette syndrome and functional tic-like behaviours can co-occur in paediatric patients and both conditions are associated with poor quality of life and higher rates of comorbid neurodevelopmental and psychiatric disorders (Eapon et al., 2015; Martino et al., 2023).

Interoception refers to the perception of internal bodily states, for example heart rate (Craig, 2009). Atypical interoceptive processing has been reported to contribute to higher-order cognitive functioning, and a range of psychiatric and neurological disorders (Murphy et al., 2018). Recent models of interoception have suggested both accuracy (i.e. how accurately an individual perceives their internal states) and awareness (i.e. propensity to be aware of interoceptive information and be focused internally) components exist (Garfinkel et al., 2015) and that measurement of these should include both behavioural and self-report measures (Murphy et al., 2018).

Recently, interoceptive processes have been implicated as a contributing factor in TS/CTD. Reduced interoceptive accuracy in cardiac domains has been found in adults and young people with TS/CTD and has been found to be associated with tic characteristics, anxiety and quality of life (Ganos et al., 2015; Pile et al., 2018). There is also evidence that reduced interoceptive accuracy is present in adults with functional motor disorders and is associated with higher levels of depression (Ricciardi et al. 2016). However, interoceptive awareness has yet to be explored in TS/CTD or FTLB in young people using both behavioural and self-report measures.

. This study therefore contributes to the literature by:

1) Expanding on the understanding of interoceptive processes in terms of awareness and accuracy components in young people with TS/CTD and FTLB across behavioural and self-report measures;
2) Exploring interoceptive process in both cardiac and respiratory domains;

The aim of this project is to empirically measure and generate information of relevance to understanding the role of interoception in relation to tic expression and psychiatric comorbidities in TS/CTD and FTLB. Doing so will assist the refinement of existing cognitive based treatment approaches in TS/CTD and FTLB.

The primary objective is to empirically measure interoception in young people with TS/CTD and FTLB and relate it to tic severity, anxiety, and quality of life.

2.4 Provide a brief outline of the basic study design. Outline what approach is being used and why.

This is a 4 x 2 ANOVA design. The first between-subjects independent variable (group) will have four levels:

1) young people aged 10-17 with TS/CTD only
2) young people aged 10-17 with FTLB only
3) young people aged 10-17 with both TS/CTD and FTLB
4) young people aged 10-17 without a diagnosis of TS/CTD or FTLB (matched for age, gender & IQ).

The second between-subjects independent variable (interoception) will have two factors:

1) interoceptive accuracy
2) interoceptive sensitivity

The two main dependent variables are:

1) interoceptive accuracy (performance on heart rate tasks)
2) interoceptive sensibility (performance on respiratory output tasks and outcomes of self-reported questionnaires)

Tic related factors, psychiatric comorbidities, and quality of life will be assessed via self-reported questionnaires. ANOVAs will be performed to investigate group differences on interoception tasks and questionnaires. Regression analyses will be used to examine the relationship between interoception, tics, and psychiatric symptoms. Correlational analyses will be used to explore the relationship between objective and subjective measures.

2.5 What are the key research question(s)? Specify hypotheses if applicable.

The primary research question is to investigate and compare interoceptive processes in young people with a diagnosis of TS/CTD to young people with FTLB, young people with both TS/CTD and FTLB, and to young people without TS/CTD and FTLB.

The secondary research question is to assess the impact of interoception and attentional control on tic expression, psychiatric comorbidities, and quality of life in young people with TS/CTD and FTLB.

The hypotheses are that:

1) Young people with TS/CTD will exhibit *reduced* interoceptive *accuracy* relative to typically developing (TD) controls.
2) Young people with TS/CTD will exhibit a *positive* relationship between atypical interoception awareness and a) tic related factors and b) anxiety symptoms.
3) Young people with FTLB will exhibit *reduced* interoceptive *accuracy* relative to TD controls and young people with TS/CTD.
4) Young people with FTLB will exhibit *reduced* interoceptive *awareness* relative to TD controls and young people with TS/CTD.

3. Sample and setting

3.1 Who are the proposed participants and where are they from (e.g. fellow students, club members)? List inclusion / exclusion criteria if applicable.

Participants will be recruited from the general public, and will be recruited from a list of people who previously participated in research and gave written agreement to take part again. They will NOT be recruited via NHS services. Young people with TS/CTD and/or FTLB will also be recruited via the Tourette's Action charity website and social media. Young people without a diagnosis of TS/CTD (control group) will be recruited through local schools or via social media

Four groups of participants will be recruited to the study:

1. Young people aged 10-17 years old with a self-reported diagnosis of TS/CTD
2. Young people aged 10-17 years old with a self-reported diagnosis of FTLB
3. Young people aged 10-17 years old with a self-reported diagnosis of TS/CTD and FTLB
4. Young people aged 10-17 years old without a self-reported diagnosis of TS/CTD and FTLB (matched for age, gender, and IQ – control group).

The subject inclusion criteria are:

- Participant is willing and able to give informed consent (for over 16yrs old) or fullinformed assent (under 16yrs) assent for participation in the study and their parent/carer are willing and able to give informed consent on their behalf.
- Participant is aged between 10-17 years with a self-reported diagnosis of TS/CTD AND/OR a diagnosis of FTLB.

Exclusion criteria:

- Other diagnosed neurological condition or learning disability

- Presence of breathing tics or gasping as this may influence the performance on the respiratory tasks
- Young people who are not able to understand and complete the consent or assent form and measures by either their parent's or their own judgement
- Non-English speaking (as questionnaires are normed in English)

3.2. How will the participants be identified and approached? Provide an indication of your sample size. If participants are under the responsibility of others (e.g., parents/carers, teachers) state if you have permission or how you will obtain permission from the third party).

Participants will be recruited via charities and organisations (e.g. Tourette Action Charity) that offer support to young people with a diagnosis of TS/CTD and/or FTLB. Organisations will be contacted via email to request to advertise the study on their websites and social media pages using approved recruitment posters. Previous participants who gave written agreement to participate again will also be contacted. Local schools will also be contacted via email to request recruitment posters be displayed to help recruit typically developing children for the control group. Recruitment posters will also be shared on social media to help with recruitment.

Recruitment posters will contain the researchers contact details for interested individuals to express their interest in participating in the study.

If they are willing to participate, the young person and their parent/legal guardian will be contacted by the researcher to arrange a time and date to attend the University of Southampton for the study procedure.

Participants will be provided with £30 to reimburse them for their travel expenses. Participants will be given cash after completing the study and will be asked to sign a form confirming they have received the reimbursement.

Sample size:

A power analysis was conducted based on Pile et al.'s (2018) medium effect size (η_p^2 = .16). G Power was used to calculate the sample size (α = .05, β = 0.8, F = .44). This

suggested a total sample size of 61 participants (16 participants per group) would provide sufficient power to obtain a medium effect size.

3.3 Describe the relationship between researcher and sample. Describe any relationship e.g., teacher, friend, boss, clinician, etc.
It is unlikely the researcher will have a relationship with the sample. However, if the researcher identified a participant they had a relationship with they would inform their supervisor and discuss whether it would be appropriate for them to participate in the study.

3.4 How will you obtain the consent of participants? (*please upload a copy of the consent form if obtaining written consent***) NB A separate consent form is not needed for online surveys where consent can be indicated by ticking/checking a consent box (normally at the end of the PIS). Other online study designs may still require a consent form or alternative procedure (for example, recorded verbal consent for online interviews).**
Written informed assent/ consent will be taken for all young people and their parents participating in the study before any study specific procedures are undertaken. For participants under the age of 16, consent will be taken from parents/legal guardians and assent will be taken from the young person. For participants over the age of 16, consent will be obtained from the young person and their parent/carer will be made aware of their participation. A study information sheet will be e-mailed to the parents and potential participants, outlining the study and its' aims. Assent / consent will be emailed to families to sign and send back prior to study participation. Consent forms will also be available on site for families to sign on the day of the tasks.

3.5 Is there any reason to believe participants may not be able to give full informed consent? If yes, what steps do you propose to take to safeguard their interests?
Yes, this study includes minors. Minors who are not able to provide informed assent/consent, or have parents who are unable to provide informed consent, will not be included in the study.

4. Research procedures, interventions and measurements

4.1 Give a brief account of the procedure as experienced by the participant. Make it clear who does what, how many times and in what order. Make clear the role of all assistants and collaborators. Make clear the total demands made on participants, including time and travel. *Upload copies of questionnaires and interview schedules to ERGO*.

Procedure:

Once participants have agreed to participate in the study, they will be asked whether they would like to arrange a day to visit the University of Southampton to complete the study or if they would prefer the researchers to come to their home to complete the experimental part of the study

Parent/carers of participants will be emailed an online link for them and their child to complete questionnaires prior to visiting the University of Southampton or the reserachers visiting their home. They will also be sent an anonymous participant code for them to enter at the beginning of the questionnaires to allow the researcher to see they have completed them. This online questionnaire set will include 7 child self-reported measures:

- **Interoceptive Accuracy Scale – Child Version (IAS; Murphy et al., 2018)**

This is a measure of self-perceived interoceptive awareness and has good internal consistency (α = .88).

- **Body Awareness Very Short Form (Cabrera et al., 2018)**
  This questionnaire measures children's awareness of bodily states, providing a self-report of interoceptive sensitivity. Cabrera et al. (2018) found this questionnaire has high internal consistency (ω = .83 – .91).
- **Attention Control Scale for Children (ACS; Muris et al., 2004)**
  This measures children's attentional shifting ability and has good internal consistency (α = .76).
- **Premonitory Urge for Tics Scale (PUTS; Woods et al., 2005)**
  This questionnaire assesses the premonitory urgers individuals experience prior to tic onset. The PUTS has good internal consistency of α = .81 and α = .82 (Pile et al., 2018).
- **The Motor tic, Obsessions and compulsions, Vocal tic Evaluation Survey (MOVES**; **Gaffney et al., 1994**)
  This measures tic frequency and severity as well as other tic related difficulties including obsessive compulsive symptoms. Gaffney et al. (1994) found the MOVES has acceptable

internal consistency (α = .69) and good sensitivity (87%) and specificity (94%) for diagnosing tics.

- **Revised Child Anxiety and Depression Scale (RCADS; Chorpita et al., 2005)**
  This questionnaire measures children's levels of anxiety and depression. It has been found to have high internal consistency (α = .78 - .88) across the subscales.
- **Paediatric Quality of Life Inventory (PEDS-QL; version 4.0; Varni et al., 1999)**
  This measures children's quality of life in various domains and studies have found it has good reliability and validity (Upton et al., 2005)

Participants will be informed if they are unable to complete measures online, time will be allocated when they meet with the researches either at the University of Southampton or in their home to complete them via hand.

On the day of the experimental part of the study, the participant and their parent/carer will be greeted by the researcher who will re-explain the purpose of the study and what is required of the participant during the visit. The researcher will check the participant's understanding of this information and answer any questions they have about the study. The researcher will also run through the eligibility criteria for participating and re-confirm the diagnoses of the participant. For participants in the control group, the researcher will ask whether they have ever had any tics and go through a list of common childhood tics. If the participant is found to have a history of childhood tics, the primary researcher will inform the young person that they are unable to participate in the study but still reimburse them for coming to the University of Southampton. They will also check that the participants have completed the online questionnaires. For participants where they have requested researchers complete the experimental part of the study at their home, the researchers will arrive at the participant home at an agreed time and day. The researchers will bring the equipment and questionnaires and ask for a space to set up the tasks. Participants will still be reimbursed £30 for their time and participation.

The researcher will then will complete three clinician-based questionnaires with the participant:

- **Demographic Information –** age, gender, diagnosis and age it was provided, alongside comorbid diagnoses.
- **Weschler Abbreviated scale of intelligence (WASI; Weschler, 1999)**
  Two subtests of the WASI (matrix reasoning and vocabulary) will be undertaken and used to assess specific cognitive functioning and allow us to identify matched controls for our sample.
- **Yale Global Tic Severity Scale – Global Impairment Section (YGTSS; Leckman et al. 1989)**

The global impairment category assesses the impact tics have on a young person's day-to-day life and functioning. This will be used rather than going through the whole questionnaire. The full questionnaire has acceptable internal consistency (ω = .58) and good inter-rater reliability.

The researcher will also ask the parent/carer to complete a parent-rated measure:

- **Swanson, Nolan, and Pelham, Version IV Rating Scale (SNAP-IV; Swanson, 1992).** This is a parent measure assessing children's symptoms of ADHD. It has previously been found to have good internal consistency (α = .93; Pile et al., 2018).

The researcher will provide the participant breaks throughout the day and once they have completed the above questionnaires, they will be asked to participate in two experimental tasks:

Heartbeat Counting Task

The Heartbeat Counting Task (HCT; Schandry, 1981) provides a measure of interoceptive accuracy by participants counting their heartbeat for 30 seconds without using strategies such as pulse taking. The participants' actual heartbeat is recorded using a pulse oximeter to enable calculation of accuracy. This task has been used in children with TS/CTD previously (Pile et al., 2018). The researcher will demonstrate how to wear the pulse oximeter and how to complete the task for the young person and check their understanding before starting the task.

Respiratory Output Task

The respiratory output task (Murphy et al., 2018) will be used to measure participants' interoceptive awareness. Participants are asked to complete a first large exhalation into a peak flow meter, which will be taken as the standard (100%) for that trial. They are then given a target (e.g. 50% of first exhalation) and asked to perform a second exhalation aiming for this percentage. The actual value is recorded by the peak flow meter. The instructions will be amended for young people to show a scale of 1-10 and breaths will be labelled on this (e.g. "I now want you to do a 5/10 breath"). Participants will complete six blocks of three trial targets (3, 5, 7 out of 10). The researcher will demonstrate how to use the peak flow meter to the young person and allow them to practice using it before starting the trials.

We anticipate the duration of the visit will be up to 1.5 hours. Once they have completed the task and questionnaires they will be asked if they have any further questions. They will then be thanked for participating and will receive a £30 in cash for travel expenses to the University of Southampton/participating in the study.

4.2 Will the procedure involve deception of any sort? If yes, what is your justification?

No.

4.3. Detail any possible (psychological or physical) discomfort, inconvenience, or distress that participants may experience, including after the study, and what precautions will be taken to minimise these risks.

Participants will be asked to complete questionnaires about their tics, mood, and quality of life. These measures are not generally experienced as distressing. On the day of testing, all participating young people and their parent/carer will be informed that the researcher will be available to discuss any concerns. It is possible that completion of the questionnaires (e.g. mood questionnaires) reveals information concerning the participant's mental health which requires disclosure to others. If the participant raises concerns about possible risk to self or others, this will be discussed with them directly by a member of the research team with expertise in mental health. Where appropriate, the participant's parent/legal guardian will be included in the discussion and may be advised to seek a referral for local support (e.g. CAMHS). Local Safeguarding Children procedures will be followed.

It is possible that participants will experience minimal physical discomfort when wearing the pulse oximeter and breathing into the peak flow meter. The researcher will inform the participants that the equipment does not hurt but may feel uncomfortable. The researcher will also demonstrate how to use the equipment safely and correctly. Participants will be reminded they can stop participating at any point in the study. If participants become stressed or anxious during testing, the researcher will provide them with an opportunity to relax and have a break before continuing.

It is possible that young people with TS/CTD and FTLB may find their tics are exacerbated during testing. This is unlikely but if this does occur, participants will be given the opportunity to relax and have a break from the study

It is possible that the participants may not be local to the University of Southampton and have to travel far to participate. This may mean young people and their parent/carer having to miss school/work to attend the research study which could be seen as an inconvenience. Therefore, the researcher will aim to schedule appointments for testing in school holidays or at times/days preferred by the family. Participants will also be informed they will receive £30 for their travel expenses. By allowing participants to complete the experimental part of the study within their own home, it will reduce the demands and inconvenience participants may have encountered when travelling to the University of Southampton. This will allow them to feel more comfortable and accommodate those participants that may not have access to transport to participate in the study.

4.4 Detail any possible (psychological or physical) discomfort, inconvenience, or distress that YOU as a researcher may experience, including after the study, and what precautions will be taken to minimise these risks. If the study involves lone working please state the risks and the procedures put in place to minimise these risks (please refer to the lone working policy).

If safeguarding or risk to self/others concerns are raised by a participant, it is possible the researcher may feel distressed depending on the content of the concerns. If this was to occur, the primary researcher would arrange supervision with a member of the research team to discuss their own wellbeing and concerns.

The researcher will have access to the laboratory at the University of Southampton for participants to complete the questionnaires and tasks for this research study. The researcher will meet participants and their parent/carer and go through the questionnaires and experimental tasks on their own, without the presence of another member of the research team in the clinic room. However, the researcher will ensure visits are scheduled on days and specific times where members of staff are in the building and available should they require support. The researcher will ensure no lone working is undertaken during the study.

The researcher will recruit a voluntary research assistant (VRA) through the scheme by the University of Southampton. Both the lead researcher and the VRA will go together to the participants homes if requested. It is possible the researchers could be exposed to risks such as mould or other household hazards (e.g. clutter, pets,) that could cause injury or ill health. There is also risk that they could encounter aggressive behaviour from participants.

To mitigate these risks the researcher and VRA will always be together and will always have a mobile phone with them. They will inform the supervisor the time and day of when the visit is and send a message once they arrive at the participant home and when they leave. If the researchers fail to check in with the supervisor, the supervisor will contact the police if they cannot be reached. Researchers will also only visit participants homes within usual working hours (Monday-Friday, 9am-5pm).

4.5 Explain how you will care for any participants in 'special groups' e.g., those in a dependent relationship, are vulnerable or are lacking mental capacity), if applicable:

The researcher will be working with young people aged 10-17 years old who may be considered part of a "special group". As aforementioned, if they are under 16-years-old their parent/carer will be required to complete the consent form. All participants will be asked to have a parent/carer accompany them to the clinic for the study.

The researcher does not foresee any other individuals in 'special groups' participating in the research.

4.6 Please give details of any payments or incentives being used to recruit participants, if applicable:

Young people will be given £30 as a thank you for participating in the research and to cover their travel expenses to the University of Southampton. They will be given this following completion of the questionnaires and experimental tasks. Participants will be asked to sign a form confirming they received payment.

5. Access and storage of data

5.1 How will participant confidentiality be maintained? Confidentiality is defined as non-disclosure of research information except to another authorised person. Confidential information can be shared with those already party to it and may also be disclosed where the person providing the information provides explicit consent. Consider whether it is truly possible to maintain a participant's

involvement in the study confidential, e.g. can people observe the participant taking part in the study? How will data be anonymised to ensure participants' confidentiality?
All data will be collected and handled in line with Research Data Management Policy, Open Access Policy, Data Protection Act 2018, and GDPR. To ensure the confidentiality of personal information, all the participants will be randomly allocated a unique study ID code. This unique code will be used on all collected data from the experimental tasks and questionnaires and used throughout the data analysis. Therefore, no personally identifiable information will be associated directly with participant's data. The only personal data that will be recorded is the birth date to determine age. ID codes will be stored on electronic data that is password protected to maintain data security. A link between the data collected and the individual study subject will be created only in case of an emergency and only if the data may be relevant to the resolution of this emergency regarding the individual. Study subjects are provided with the right to have their information removed from the database at any time and by their request the data is available to them for review and correction.

5.2 How will personal data and study results be stored securely during and after the study. Who will have access to these data?
All data will be either stored as paper documents stored in a locked drawer at the University of Southampton or as password-protected electronic data stored on the University of Southampton's secure networks and drives. Only named research members with appropriate backgrounds will have access to the patient's personal data. Those with access to person identifiable information will be made aware of their responsibilities. The database will be used solely for the purpose of research. The data will not be transferred to establishments not participating in the study, including other research facilities, schools, health organisation, etc. For the purpose of analysis, data will be entered into SPSS and anonymized. Consent/assent forms, experimental and questionnaire data, and identifying information will be kept in a locked draw at the University of Southampton for 10 years after study completion.

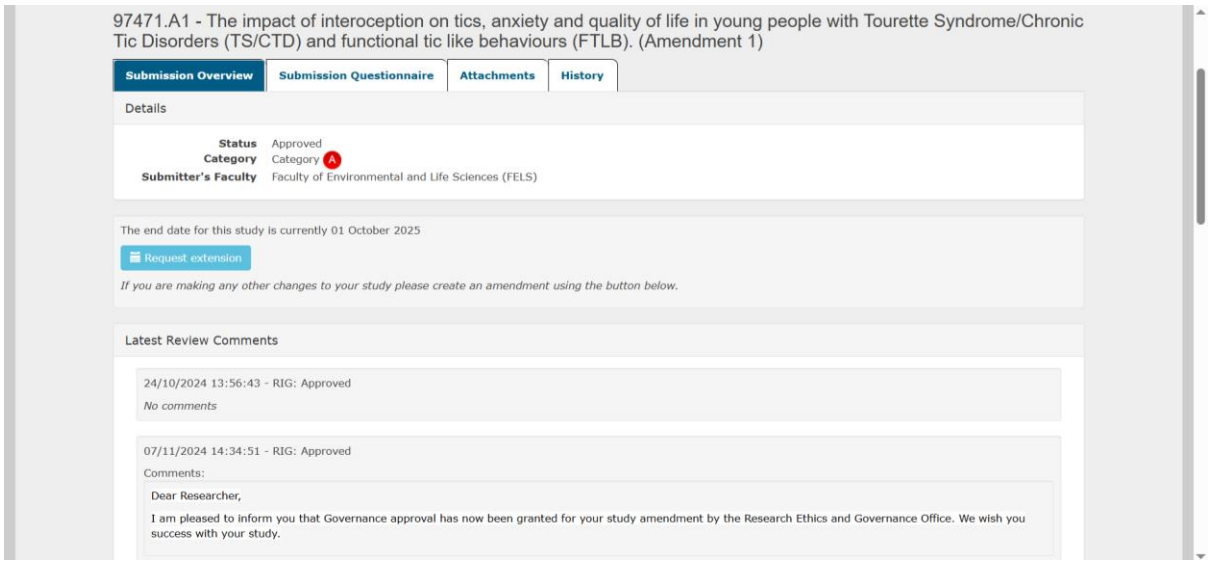
Similarly, any electronic data will be stored securely on the University of Southampton's networks for 10 years following study completion. Data will be destroyed and deleted after 10 years. When travelling to participants homes, all electronic data collected during the visit will be stored on a password protected university laptop and then uploaded to the University of Southampton's secure networks and drives. All equipment and hard copies of confidential data will be stored in a lockable bag when visiting participant homes.

5.3 How will it be made clear to participants that they may withdraw consent to participate? Please note that anonymous data (e.g. anonymous questionnaires) cannot be withdrawn after they have been submitted. If there is a point up to which data can be withdrawn/destroyed e.g., up to interview data being transcribed please state this here.
The participant information sheet will inform participants on how they can withdraw their consent. They will also be reminded on the day of testing of their right to withdraw. Participants can withdraw at any time prior to visiting the University of Southampton for testing and during the day. Participants will be informed that it may not be possible to withdraw six months or more following participating in the study because the data may have already been analysed for the final report.

6. Additional Ethical considerations

6.1 Are there any additional ethical considerations or other information you feel may be relevant to this study?
n/a

Appendix C Ethics Approval



97471.A1 - The impact of interoception on tics, anxiety and quality of life in young people with Tourette Syndrome/Chronic Tic Disorders (TS/CTD) and functional tic like behaviours (FTLB). (Amendment 1)

Submission Overview | Submission Questionnaire | Attachments | History

Details

Status Approved
Category Category A
Submitter's Faculty Faculty of Environmental and Life Sciences (FELS)

The end date for this study is currently 01 October 2025

Request extension

If you are making any other changes to your study please create an amendment using the button below.

Latest Review Comments

24/10/2024 13:56:43 - RIG: Approved

No comments

07/11/2024 14:34:51 - RIG: Approved

Comments:

Dear Researcher,

I am pleased to inform you that Governance approval has now been granted for your study amendment by the Research Ethics and Governance Office. We wish you success with your study.

Appendix D Demographic Questionnaire

Demographic Information:

1. How old are you?

   .....

2. Please select which gender you identify with:

   ☐Male ☐Female ☐Non-Binary

   ☐Other: .....

3. What is your ethnicity?

   ☐Asian, Asian British, or Asian Welsh

   ☐Black, Black British, Black Welsh, Caribbean, or African

   ☐Mixed or Multiple ethnic groups

   ☐White: English, Welsh, Scottish, Northern Irish, or British

   ☐Other ethnic groups: .....

   ☐Prefer not to say

4. How old were you when you first had tics?

   .....

5. Do you have a diagnosis of a tic disorder?

☐ Yes ☐ No

If so, please circle which one best describes your diagnosis:

☐Tourette syndrome (TS) ☐Chronic tic disorder (CTD)

☐Functional tic like behaviours/functional tics (FTLB)

☐Both TS/CTD AND FTLB

☐None of the above

6. How old were you when you were diagnosed?

   .....

7. What professional gave you this diagnosis?
   ☐GP ☐Neurologist ☐Psychiatrist ☐Psychologist

   ☐Other: .....

8. Do you have any other physical health or mental health diagnoses? (e.g. OCD, ADHD, anxiety, autism, epilepsy, learning disability)
   Please list all you have been diagnosed with:

   .....

   ....

   .....

   .....

   .....

   .....

Appendix E Attentional Control Scale for Children

ACS-C (CHILD)

DIRECTIONS: Please read each sentence carefully and circle the answer that best describes how you are most of the time.

	1	2	3	4
1. It's very hard for me to concentrate on a difficult task when there are noises around.	almost	sometime	often	always
2. When I need to concentrate and solve a problem, I have trouble focusing my attention.	almost	sometime	often	always
3. When I am working hard on something, I still get distracted by events around me.	almost	sometime	often	always
4. My concentration is good even if there is music in the room around me.	almost	sometime	often	always
5. When concentrating, I can focus my attention so that I become unaware of what's going on in the room around me.	almost never	sometimes	often	always
6. When I am reading or studying, I am easily distracted if there are people talking in the same room.	almost never	sometimes	often	always
7. When trying to focus my attention on something, I have difficulty blocking out distracting thoughts.	almost	sometime	often	always
8. I have a hard time concentrating when I'm excited about something.	almost	sometime	often	always
9. When concentrating I ignore feelings of hunger or thirst.	almost	sometime	often	always
10. I can quickly switch from one task to another.	almost	sometime	often	always
11. It takes me a while to get really involved in a new task.	almost	sometime	often	always
12. It is difficult for me to coordinate my attention between the listening and writing required in classes.	almost never	sometimes	often	always
13. I can become interested in a new topic very quickly when I need to.	almost	sometime	often	always
14. It is easy for me to read or write while I'm also talking on the phone.	almost	sometime	often	always
15. I have trouble carrying on two conversations at once.	almost	sometime	often	always
16. I have a hard time coming up with new ideas quickly.	almost	sometime	often	always
17. After being interrupted or distracted, I can easily shift my attention back to what I was doing	almost	sometime	often	always
18. When a distracting thought comes to mind, it is easy for me to shift my attention away from it.	almost	sometime	often	always

19. It is easy for me to alternate between two different tasks.	almost	sometime	often	always
20. It is hard for me to break from one way of thinking about something and look at it from another point of view.	almost never	sometimes	often	always

Appendix F Motor tic, Obsessions and compulsions, Vocal tic Evaluation Survey

The MOVES questionnaire

Please answer these questions for the past two weeks.

	NEVER	SOMETIMES	OFTEN	ALWAYS
1. I make noises (like grunts) that I can't stop.				
2. Parts of my body jerk again and again, that I can't control.				
3. I have bad ideas over and over, that I can't stop.				
4. I have to do things in certain order or certain ways (like touching things}.				
5. Words come out that I can't stop or control.				
6. At times I have the same jerk or twitch over and over.				
7. Certain bad words or thoughts keep going through my mind.				
8. I have to do exactly the opposite of what I'm told.				
9. The same unpleasant or silly thought or picture goes through my mind.				
10. I can't control all my movements.				
11. I have to do several movements over and over again, in the same order.				
12. Bad or swear words come out that I don't mean to say.				
13. I feel pressure to talk, shout, or scream.				
14. I have ideas that bother me (like germs or like hurting myself).				
15. I do certain things (like jumping or clapping) over and over.				
16. I have habits or movements that come out more when I'm nervous.				
17. I have to repeat things that I hear other people say.				
18. I have to do things I see other people do.				
19. I have to make bad gestures (like the finger).				
20. I have to repeat words or phrases over and over.				

The motor tic, obsession and compulsion, and vocal tic evaluation scale

Appendix G Premonitory Urge for Tics Scale

Premonitory Urge for Tics Scale (PUTS)

By Douglas Woods, Ph.D.

Journal of Developmental and Behavioral Pediatrics, volume 26, number 6, December 2005 pp397-403

Name _____ Age _____ Place: school clinic home other

Date _____ Diagnosis (if known) _____

How I feel	***Not at all***	***A little***	***Pretty much***	***Very much***
Right before I do a tic I feel like my insides are itchy				
Right before I do a tic I feel pressure inside my brain or body				
Right before I do a tic I feel "wound up" or tense inside				
Right before I do a tic I feel like something is not "just right"				
Right before I do a tic I feel like something isn't complete				
Right before I do a tic I feel like there is energy in my body that needs to get out				
I have these feelings almost all the time before I do a tic				
These feelings happen for every tic I have				
After I do the tic, the itchiness, energy, pressure, tense feelings or feelings that something isn't "just right" or complete go away, at least for a while				
I am able to stop my tics even if only for a short period of time				
Total scores (except item number ten) On a scale of 1-4, from least to most				

Appendix H Yale Global Tic Severity Scale Impairment Rating

IMPAIRMENT

NONE	❑	0
MINIMAL Tics associated with subtle difficulties in self-esteem, family life, social acceptance, or school or job functioning (infrequent upset or concern about tics vis a vis the future, periodic, slight increase in family tensions because of tics, friends or acquaintances may occasionally notice or comment about tics in an upsetting way).	❑	10
MILD Tics associated with minor difficulties in self-esteem, family life, social acceptance, or school or job functioning.	❑	20
MODERATE Tics associated with some clear problems in self-esteem family life, social acceptance, or school or job functioning (episodes of dysphoria, periodic distress and upheaval in the family, frequent teasing by peers or episodic social avoidance, periodic interference in school or job performance because of tics).	❑	30
MARKED Tics associated with major difficulties in self-esteem, family life, social acceptance, or school or job functioning.	❑	40
SEVERE Tics associated with extreme difficulties in self-esteem, family life, social acceptance, or school or job functioning (severe depression with suicidal ideation, disruption of the family (separation/divorce, residential placement), disruption of social tics - severely restricted life because of social stigma and social avoidance, removal from school or loss of job).	❑	50

Appendix I Revised Child Anxiety and Depression Scale

Date: _____ Name/ID: _____

RCADS

Please put a circle around the word that shows how often each of these things happens to you. There are no right or wrong answers.

1. I worry about things	Never	Sometimes	Often	Always
2. I feel sad or empty	Never	Sometimes	Often	Always
3. When I have a problem, I get a funny feeling in my stomach	Never	Sometimes	Often	Always
4. I worry when I think I have done poorly at something	Never	Sometimes	Often	Always
5. I would feel afraid of being on my own at home	Never	Sometimes	Often	Always
6. Nothing is much fun anymore	Never	Sometimes	Often	Always
7. I feel scared when I have to take a test	Never	Sometimes	Often	Always
8. I feel worried when I think someone is angry with me	Never	Sometimes	Often	Always
9. I worry about being away from my parents	Never	Sometimes	Often	Always
10. I get bothered by bad or silly thoughts or pictures in my mind	Never	Sometimes	Often	Always
11. I have trouble sleeping	Never	Sometimes	Often	Always
12. I worry that I will do badly at my school work	Never	Sometimes	Often	Always
13. I worry that something awful will happen to someone in my family	Never	Sometimes	Often	Always
14. I suddenly feel as if I can't breathe when there is no reason for this	Never	Sometimes	Often	Always
15. I have problems with my appetite	Never	Sometimes	Often	Always
16. I have to keep checking that I have done things right (like the switch is off, or the door is locked)	Never	Sometimes	Often	Always
17. I feel scared if I have to sleep on my own	Never	Sometimes	Often	Always
18. I have trouble going to school in the mornings because I feel nervous or afraid	Never	Sometimes	Often	Always
19. I have no energy for things	Never	Sometimes	Often	Always
20. I worry I might look foolish	Never	Sometimes	Often	Always
21. I am tired a lot	Never	Sometimes	Often	Always
22. I worry that bad things will happen to me	Never	Sometimes	Often	Always

Page 1

23. I can't seem to get bad or silly thoughts out of my head	Never	Sometimes	Often	Always
24. When I have a problem, my heart beats really fast	Never	Sometimes	Often	Always
25. I cannot think clearly	Never	Sometimes	Often	Always
26. I suddenly start to tremble or shake when there is no reason for this	Never	Sometimes	Often	Always
27. I worry that something bad will happen to me	Never	Sometimes	Often	Always
28. When I have a problem, I feel shaky	Never	Sometimes	Often	Always
29. I feel worthless	Never	Sometimes	Often	Always
30. I worry about making mistakes	Never	Sometimes	Often	Always
31. I have to think of special thoughts (like numbers or words) to stop bad things from happening	Never	Sometimes	Often	Always
32. I worry what other people think of me	Never	Sometimes	Often	Always
33. I am afraid of being in crowded places (like shopping centers, the movies, buses, busy playgrounds)	Never	Sometimes	Often	Always
34. All of a sudden I feel really scared for no reason at all	Never	Sometimes	Often	Always
35. I worry about what is going to happen	Never	Sometimes	Often	Always
36. I suddenly become dizzy or faint when there is no reason for this	Never	Sometimes	Often	Always
37. I think about death	Never	Sometimes	Often	Always
38. I feel afraid if I have to talk in front of my class	Never	Sometimes	Often	Always
39. My heart suddenly starts to beat too quickly for no reason	Never	Sometimes	Often	Always
40. I feel like I don't want to move	Never	Sometimes	Often	Always
41. I worry that I will suddenly get a scared feeling when there is nothing to be afraid of	Never	Sometimes	Often	Always
42. I have to do some things over and over again (like washing my hands, cleaning or putting things in a certain order)	Never	Sometimes	Often	Always
43. I feel afraid that I will make a fool of myself in front of people	Never	Sometimes	Often	Always
44. I have to do some things in just the right way to stop bad things from happening	Never	Sometimes	Often	Always
45. I worry when I go to bed at night	Never	Sometimes	Often	Always
46. I would feel scared if I had to stay away from home overnight	Never	Sometimes	Often	Always
47. I feel restless	Never	Sometimes	Often	Always

Appendix J Swanson, Nolan and Pelham Questionnaire

SNAP-IV 26-Item Teacher and Parent Rating Scale
James M. Swanson, Ph.D., University of California, Irvine, CA 92715

Patient/Client Name: _____

Date of birth: _____ Gender: _____

Grade: _____ Type of class: _____ Class size: _____

Completed by: _____ Date: _____

Physician Name: _____

For each item, check the column which best describes this child/adolescent:

	Not at all	Just a little	Quite a bit	Very much
1. Often fails to give close attention to details or makes careless mistakes in schoolwork or tasks				
2. Often has difficulty sustaining attention in tasks or play activities				
3. Often does not seem to listen when spoken to directly				
4. Often does not follow through on instructions and fails to finish schoolwork, chores, or duties				
5. Often has difficulty organizing tasks and activities				
6. Often avoids, dislikes, or reluctantly engages in tasks requiring sustained mental effort				
7. Often loses things necessary for activities (e.g., toys, school assignments, pencils or books				
8. Often is distracted by extraneous stimuli				
9. Often is forgetful in daily activities				
10. Often fidgets with hands or feet or squirms in seat				
11. Often leaves seat in classroom or in other situations in which remaining seated is expected				
12. Often runs about or climbs excessively in situations in which it is inappropriate				
13. Often has difficulty playing or engaging in leisure activities quietly				
14. Often is "on the go" or often acts as if "driven by a motor"				
15. Often talks excessively				
16. Often blurts out answers before questions have been completed				
17. Often has difficulty awaiting turn				
18. Often interrupts or intrudes on others (e.g., butts into conversations/ games				
19. Often loses temper				
20. Often argues with adults				
21. Often actively defies or refuses adult requests or rules				
22. Often deliberately does things that annoy other people				
23. Often blames others for his or her mistakes or misbehaviour				
24. Often is touchy or easily annoyed by others				
25. Often is angry and resentful				
26. Often is spiteful or vindictive				

Appendix K Paediatric Quality of Life Scale

PedsQL 2

*In the **PAST MONTH**, how much of a **problem** has this been for you …*

ABOUT MY HEALTH AND ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to walk more than a couple of streets (about 100 metres)	0	1	2	3	4
2. It is hard for me to run	0	1	2	3	4
3. It is hard for me to do sports activities or exercise	0	1	2	3	4
4. It is hard for me to lift heavy things	0	1	2	3	4
5. It is hard for me to have a bath or shower by myself	0	1	2	3	4
6. It is hard for me to do chores around the house	0	1	2	3	4
7. I have aches and pains	0	1	2	3	4
8. I feel tired	0	1	2	3	4

ABOUT MY FEELINGS *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ON WITH OTHERS *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting on with other teenagers	0	1	2	3	4
2. Other teenagers do not want to be my friend	0	1	2	3	4
3. Other teenagers tease me	0	1	2	3	4
4. I cannot do things that other teenagers my age can do	0	1	2	3	4
5. It is hard to keep up with other teenagers my age	0	1	2	3	4

ABOUT SCHOOL / COLLEGE *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my school / college work	0	1	2	3	4
4. I miss school / college because of not feeling well	0	1	2	3	4
5. I miss school / college to go to the doctor or hospital	0	1	2	3	4

PedsQL 4.0 – (13-18)
f:\institut\cultadap\project\pg2161\etude2161\final_versions\format_jlmvarni\uk\pedsql4-core-a-uk.doc
APRIL 2004

Appendix L Interoceptive Accuracy Scale for Children

Written below are some sentences about how well you can feel things **inside your body**. Please read these sentences and use the scale to tell us whether they fit with you. If you think the statement fits you completely, please circle strongly agree. If you don't think it fits you at all, please circle strongly disagree.

It is very important that you only tell us how well you can feel what's **inside** your body without using signs from **outside** of your body. For example, if you can only tell your heart is beating fast by feeling your heartbeat with your hands, this would **not** count as correctly feeling when your heart is beating quickly.

(Scale strongly agree, agree, neither agree nor disagree, disagree, disagree strongly)

I am always correct at feeling…

	Strongly agree	Agree	Neither agree nor disagree	disagree	Disagree strongly
1. When my heart is beating quickly					
2. When I am hungry					
3. When I am breathing quickly					
4. When I am thirsty					
5. When I need to have a wee					
6. When I need to have a poo					
7. When I taste new flavours					
8. When I am going to					

vomit (be sick)					
9. When I am going to sneeze					
10. When I am going to cough					
11. When I am hot or cold					
12. When I am going to fart					
13. When I am going to burp					
14. When my muscles are tired or sore					
15. When I am going to get a bruise					
16. When I am in pain					
17. When I don't have any energy					
18. When someone is touching me in a nice way					
19. When something is going to be ticklish					
20. When something is going to be itchy					

Appendix M instructions for Behavioural Tasks

Heartbeat Counting Task Instructions:

This first task is called the 'count your heartbeats' task.

You will be asked to put your finger into this, its called a pulse oximeter and it measures your heart rate or how fast it is beating. You can see that when I put it in, it shows this number and this is my heart rate. I am going to ask you to wear this pulse oximeter on your non dominant index finger. I will cover the screen with my hand (like this) so you cannot read what it says.

Your job is to try and count how many times you feel your heartbeats from the time you hear the computer "ding" to when it "dings" again. It is important you do not try to measure your heartbeat by taking your pulse or applying pressure on your finger. Instead, I want you to just focus on feeling your heartbeats. After the computer does the second "ding" I want you to stop. I will then ask you to tell me how many times you felt your heartbeat in that time period. We will do this multiple times to help me get enough data to measure it.

Please do not count the number of seconds or minutes, as this is not going to be the number of times your heart has beat. If you do not feel your heartbeats at all during the time, do not say "0" as it is unlikely your heart did not beat, instead I would like you to guess how many times you think your heart beat in that time.

For each trial ask:

How many times did you count your heart beat?

Out of 10, how confident are you that you got that right? (10 being very confident, 0 being not at all)?

And out of interest, how long do you think I was timing for?

Respiratory Task Instructions

This task is called the 'big breath task'.

This will involve a 'peak flow meter' which measures how fast you can push air out of your lungs. You will be asked to breathe into this object multiple times and at different rates to test how good you are at controlling your breath.

To stop you from seeing how hard you are blowing into the peak flow meter, you will be asked to wear a blindfold. You will also be asked to wear these headphones which will be playing some

white noise to distract you from hearing how hard you are breathing. I will let you test these and make sure they are comfortable for you to wear.

I will then ask you to breathe as hard as you can into the peak flow meter (like this) and I will call this your 100% breath (use values between 1 and 10 if participant struggles with %). This will be your maximum breath. I will then ask you to breathe into the peak flow meter again but ask you to aim for a percentage of this breath (e.g. 50% of 100 – or 5 out of 10 – half of your big breath). You will be asked to do this multiple times to allow me to collect enough data and see that you understand. After each round I will ask you to tell me how much of the 10/10 breath you actually did (e.g. if I asked for 50% of 100, did you think it was 50% or did it feel more like a 70% or 40% breath?)

Does all of this make sense? If so, lets have a practice. I will show you how to hold the peak flow meter and will adjust it each time to stop you from knowing how much you did. You will get breaks in between to open your eyes and take the headphones off.

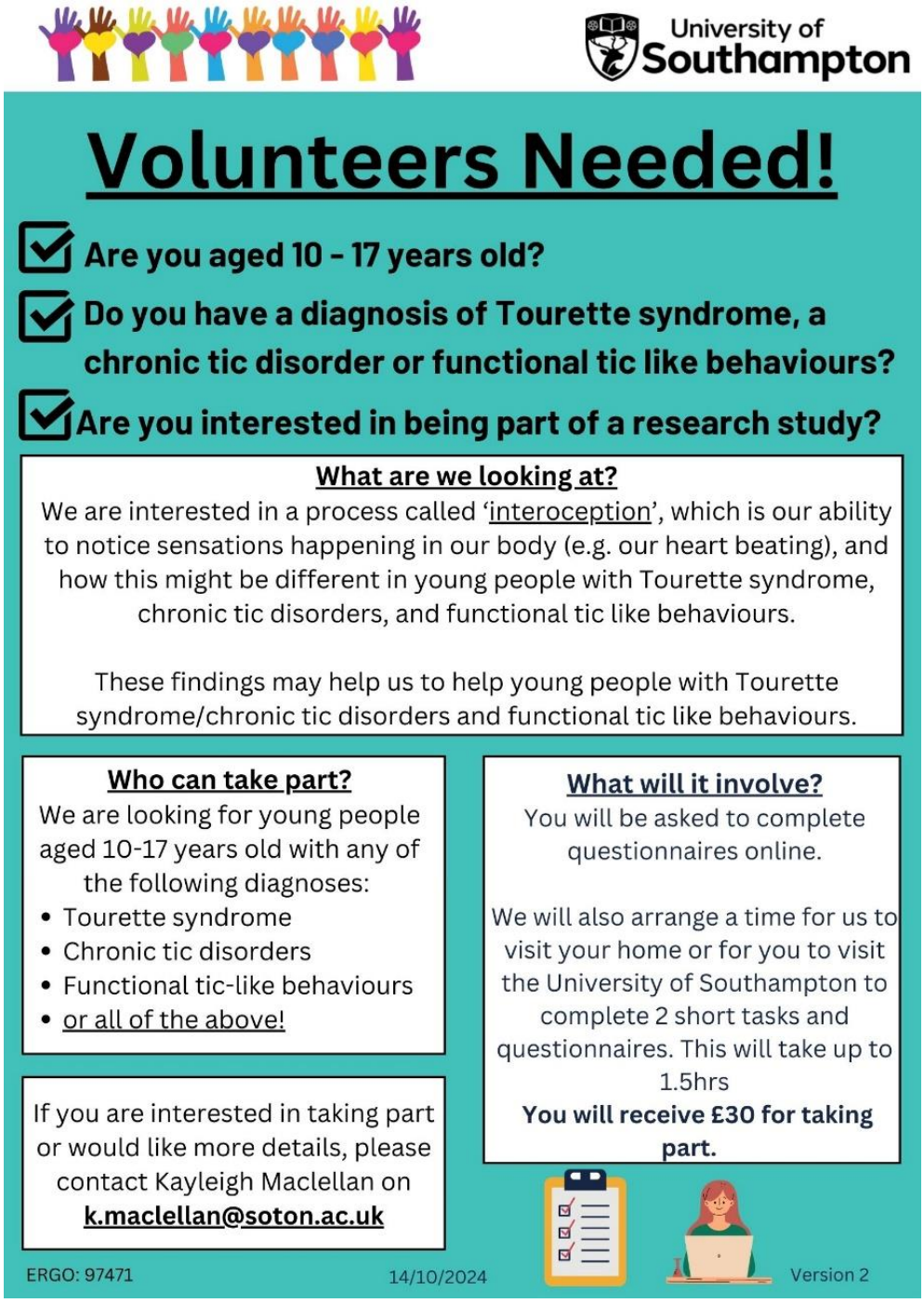
Practice

Please can you give me a 10/10 breath?

Now can you give me a 5/10 breath?

Any issues or are you ready to go?

Appendix N Recruitment Posters



University of Southampton

Volunteers Needed!

Are you aged 10 - 17 years old?

Are you interested in being part of a research study?

What are we looking at?

We are interested in a process called 'interoception', which is our ability to notice sensations happening in our body (e.g. our heart beating), and how this might be different in young people with Tourette syndrome, chronic tic disorders, and functional tic like behaviours.

These findings may help us to help young people with Tourette syndrome/chronic tic disorders and functional tic like behaviours.

Who can take part?

We are looking for young people aged 10-17 years old **without a diagnosis of:**

- Tourette syndrome
- Chronic tic disorders
- Functional tic-like behaviours

You must also not have a diagnosis of any other neurological disorder or a learning disability.

What will it involve?

You will be asked to complete questionnaires online.

We will also arrange a time for us to visit your home or for you to visit the University of Southampton to complete 2 short tasks and questionnaires.
This may take up to 1.5hrs.

You will receive £30 for taking part.

If you are interested in taking part or would like more details, please contact Kayleigh Maclellan on **k.maclellan@soton.ac.uk**

ERGO: 97471 14/10/2024 Version 2

Appendix O Example Participant Information Sheet

PARTICIPANT INFORMATION SHEET

This information sheet is for young people aged 16-17 who have Tourette syndrome, a chronic tic disorder and/or functional tic like behaviours

Study Title: Interoception in Tourette syndrome, chronic tic disorders and functional tic like behaviours

Researcher: Kayleigh Maclellan

Ethics/ERGO number: 97471

We are asking if you would like to take part in a research study with us. Before you decide whether you would like to get involved, we would like you to understand why the research is being done and what will happen.

What is this study about?

We want to see if noticing things happening in your body (e.g. your heart beating) is different in young people with Tourette syndrome/chronic tic disorders (TS/CTD) and/or with functional tic like behaviours (FTLB). This may help us to help young people with TS/CTD and FTLB. This study is being run as part of a Doctorate in Clinical Psychology at the University of Southampton.

Why are you asking me?

We are asking young people aged 10 to 17 with a diagnosis of TS/CTD and/or FTLB to take part in this study. If you don't want to take part, that is absolutely fine. And if you say yes and then decide you don't want to do it anymore, that is absolutely fine too. It is entirely up to you whether you take part or not. We will also share with your parent/carer that you are taking part in this study, but we will not require their consent for you to take part.

Not everyone can take part in this study. It is important that you read these points carefully and make sure that they do not apply to you. You should not take part in this research if:

- You have any other neurological condition or a learning disability
- You have photo-sensitivity epilepsy
- You have a breathing or gasping tic

What is good about taking part in this study?

The study may help us to learn more about what is happening in TS/CTD and FTLB so that we can try to help children and young people with these difficulties. We hope that taking part will be fun and we will give you £30 to say thank you.

What will happen if I say yes?

We will send you and your parent/carer a link to complete questionnaires online. We will then arrange a time for you to visit the University of Southampton or for our researchers to visit your home to complete some tasks and fill out more questionnaires. We will only need to see you once and the session will last around 1.5 hours. We will give you extra time to fill out the questionnaires if you were unable to do this online. You can have a break at any time you like. The tasks you will take part in are described next. You don't need to remember what to do now – we will give you all the instructions on the day.

1 COUNT YOUR HEARTBEATS TASK

We will ask you to count your heartbeat for 30 seconds without using any tools to help you. At the same time, we will record your actual heartbeat by resting your finger on a light that senses your heartbeat. It doesn't involve any other equipment and won't hurt you at all. This will take no longer than 10 minutes.

2 THE BIG BREATH TASK

Next, we will ask you to make a big breath into a tube called a 'peak flow meter' (see photo). This will be your 10 out of 10 breath. We will then give you a target breath to aim for on your next breath, for example a 5 out of 10 breath. This does not involve anything painful or scary. This will take no longer than 10

3 QUESTIONNAIRES

We will ask you to fill in questionnaires both online and at the University of Southampton. Some of these questionnaires will ask you questions about how aware you are of things happening in your body (e.g. your heart beating or your mouth being dry), how good you think you are at being aware of these things and about your mood. Your answers are **private** and will **not** be shared with anyone else. The only time we would have to speak to someone else about your answers is if you were to say you might harm yourself or someone else. In this case, we would help you to speak to your parent/carer or we can speak to them if you would prefer. We would also offer you support and help you to access appropriate help.

Could anything bad happen to me?

We don't think so. However, if you did feel upset or worried by anything, we will make sure that there is someone for you to talk to about it.

If you have a concern about any part of this study, you should speak to the researchers who will do their best to answer your questions. If you, or your parent/carer remain unhappy or

have a complaint about any part of this study, please contact the University of Southampton Head Research Ethics and Governance on 023 8059 5058 or rgoinfo@soton.ac.uk.

How will my information be used?

There are rules in place for keeping information about you and they have been explained in detail to your parent(s)/carer. The University of Southampton is in charge of looking after your information and using it properly. We will keep all information about you safe and secure. The only people from the University of Southampton who will look at this information will be people who need to contact you if they need to review the study once it is finished. Anonymised data with no personally identifiable information will be kept for 10 years in accordance with the University of Southampton's policies. Please ask if you would like help understanding this information or have any more questions.

Who will know that I have taken part?

Only your parent(s)/carer and members of the research team will know you've taken part. Once we have finished the study, we will write our reports in a way that no-one can work out that you took part.

Who can I ask any questions I have about taking part?

If you want to know anything else after reading this sheet, you can speak to your parents/carers or contact me using the contact details below:

Kayleigh Maclellan
Trainee Clinical Psychologist
School of Psychology, Highfield Campus, University of Southampton, SO17 1BJ
K.maclellan@soton.ac.uk

Thank you for reading this information sheet and for considering taking part in our study!

Appendix P Consent Form For Participants Aged 16+

CONSENT FORM FOR PARTICIPANTS AGED 16+

Study Title: Interoception in Tourette syndrome, chronic tic disorders and functional tic like behaviours

Ethics/ERGO Number: 97471

Version 2: 09/08/2024

Thank you for your interest in this study. It is very important to us to conduct our studies in line with ethics principles, and this Consent Form asks you to confirm if you agree to take part in the above study. Please carefully consider the statements below and add your initials and signature only if you agree to participate in this research and understand what this will mean for you. You will be given a copy of this consent form to keep and refer to at any time.

Consent Statements	Participant Initials
I confirm that I understand that by writing my initials in each box below I am consenting to this part of the study. I understand that it will be assumed that any boxes left blank means that I DO NOT consent to that part of the study.	
I confirm that I have read the Participant Information Sheet version 1, dated 26/07/24 explaining the study above and I understand what is expected of me.	
I was given the opportunity to consider the information, ask questions about the study, and all my questions have been answered to my satisfaction.	
I agree to take part in this study and understand that data collected during this research project will be used for the purpose of this study.	
I understand that my participation is voluntary and that I am free to withdraw from this study at any time without giving a reason.	

I understand that all personal information collected about me (e.g., my name and contact details) will be kept confidential (i.e., will not be shared beyond the study team) unless required by law or relevant regulations (e.g., for the purpose of monitoring the safety of this study).	

_____	_____	_____
Name of participant	Signature	Date
_____	_____	_____
Name of Researcher taking consent	Signature	Date

Appendix Q Consent Form For Guardians

CONSENT FORM FOR PARENTS FOR PARTICIPANTS UNDER 16 YEARS OLD

Study Title: Interoception in Tourette syndrome, chronic tic disorders and functional tic like behaviours

Ethics/ERGO Number: 97471

Version 2: 09/08/2024

Thank you for your interest in this study. It is very important to us to conduct our studies in line with ethics principles, and this Consent Form asks you to confirm if you agree for your child to take part in the above study. Please carefully consider the statements below and add your initials and signature only if you agree for your child to participate in this research and understand what this will mean for you and your child. You will be given a copy of this consent form to keep and refer to at any time.

Consent Statements	**Participant Initials**
I confirm that I understand that by writing my initials in each box below I am consenting that my child can participate in this study. I understand that it will be assumed that any boxes left blank means that I DO NOT consent to my child being involved in that part of the study.	
I confirm that I have read the participant information sheet version 1, dated 26/07/2024 explaining the study above and I understand what is expected of my and my child.	
I was given the opportunity to consider the information, ask questions about the study, and all my questions have been answered to my satisfaction.	
I agree for my child to take part in this study and understand that data collected during this research project will be used for the purpose of this study.	
I understand that my child's participation is voluntary and that they are free to withdraw from this study at any time without giving a reason.	

I understand that all personal information collected about my child (e.g., my name and contact details) will be kept confidential (i.e., will not be shared beyond the study team) unless required by law or relevant regulations (e.g., for the purpose of monitoring the safety of this study).	

Name of child:

_____	_____	_____
Name of participant	Signature	Date
_____	_____	_____
Name of Researcher taking consent	Signature	Date

Appendix R Assent Form For Participants Under 16 Years Old

ASSENT FORM FOR PARTICIPANTS UNDER 16YRS OLD

Study Title: Interoception in Tourette syndrome, chronic tic disorders and functional tic like behaviours

Ethics/ERGO Number: 97471

Version 1: 09/08/2024

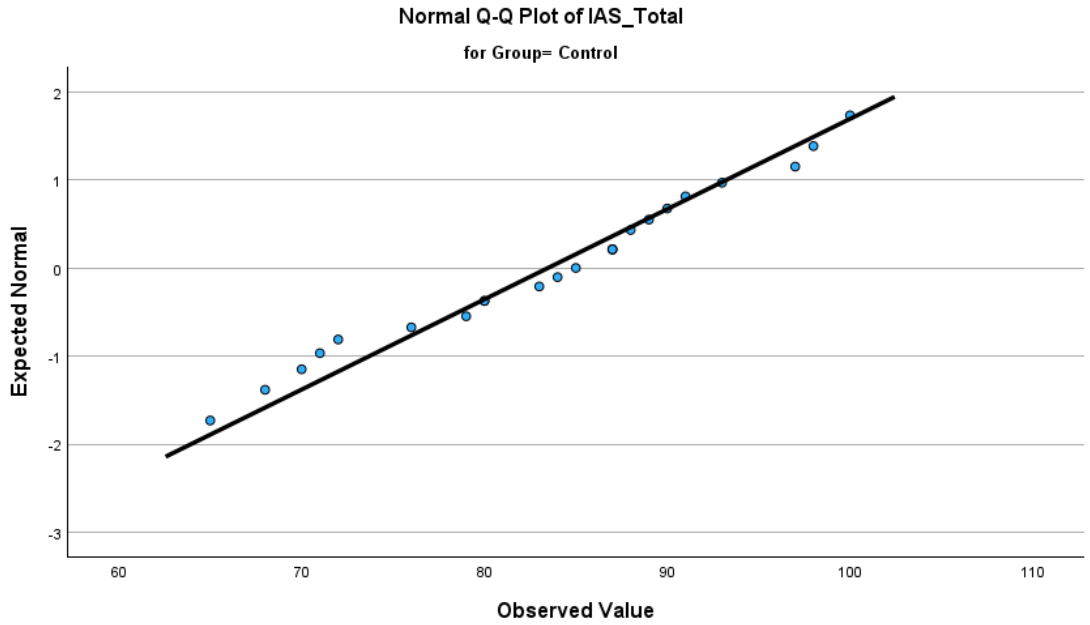
Thank you for your interest in this study. It is important we do research the right way and this form checks that you understand what the study is asking you to do and that you agree to be a part of it. Please read the information below carefully and only add your initials and signature if you agree with what is written and would like to take part in the study. You will be given a copy of this form for you to keep.

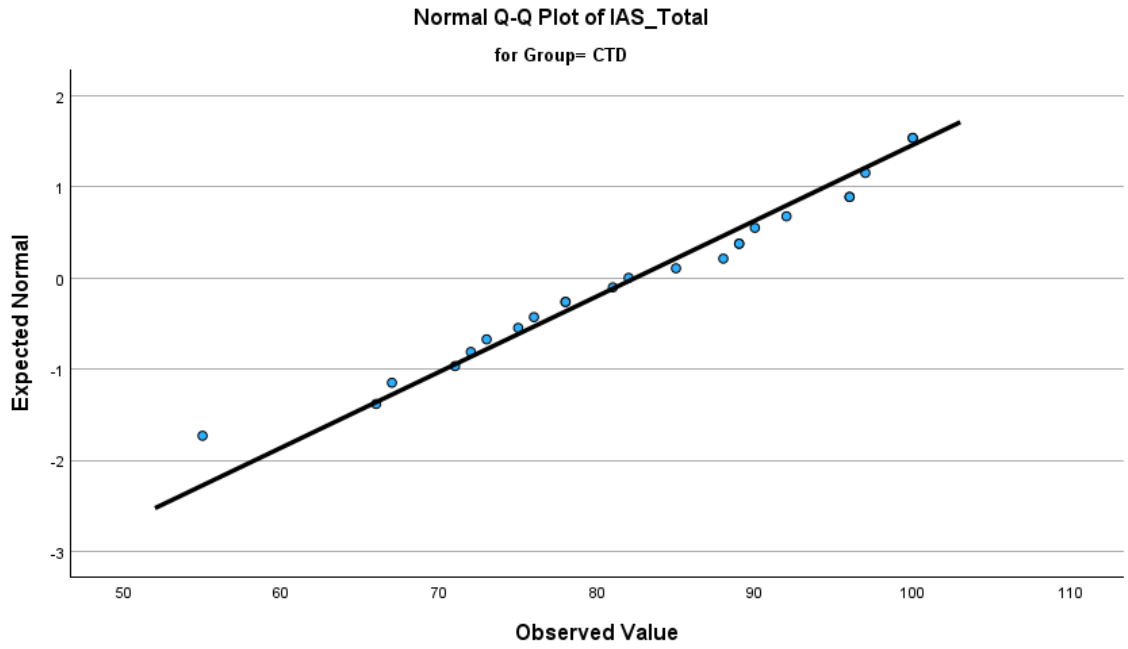
Your parent/carer will also be asked to sign this form and a separate form to say they agree to you taking part in this study.

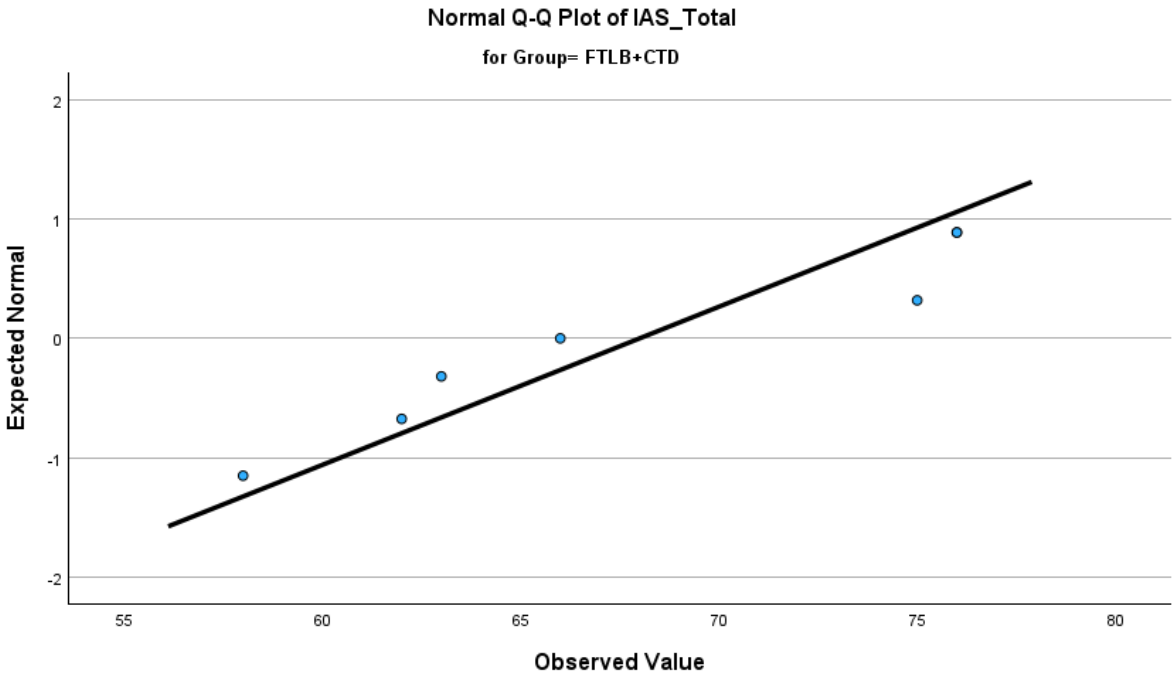
Assent Statements	**Participant Initials**
I understand that by writing my initials in each box below I am agreeing to taking part in the study. I understand if I leave any boxes blank it means I DO NOT consent to that part of the study.	
I agree I have read the Participant Information Sheet version 1, dated 26/07/24 and explaining the study and what I need to do.	
I agree I have had the chance to ask questions about the study and all my questions have been answered.	
I agree to take part in this study and understand my information will be used for this study only.	
I understand I can choose to leave the study whenever I want without having to explain why.	
I understand that any personal information collected about me (e.g., my name and contact details) will not be shared with anyone outside of the study.	

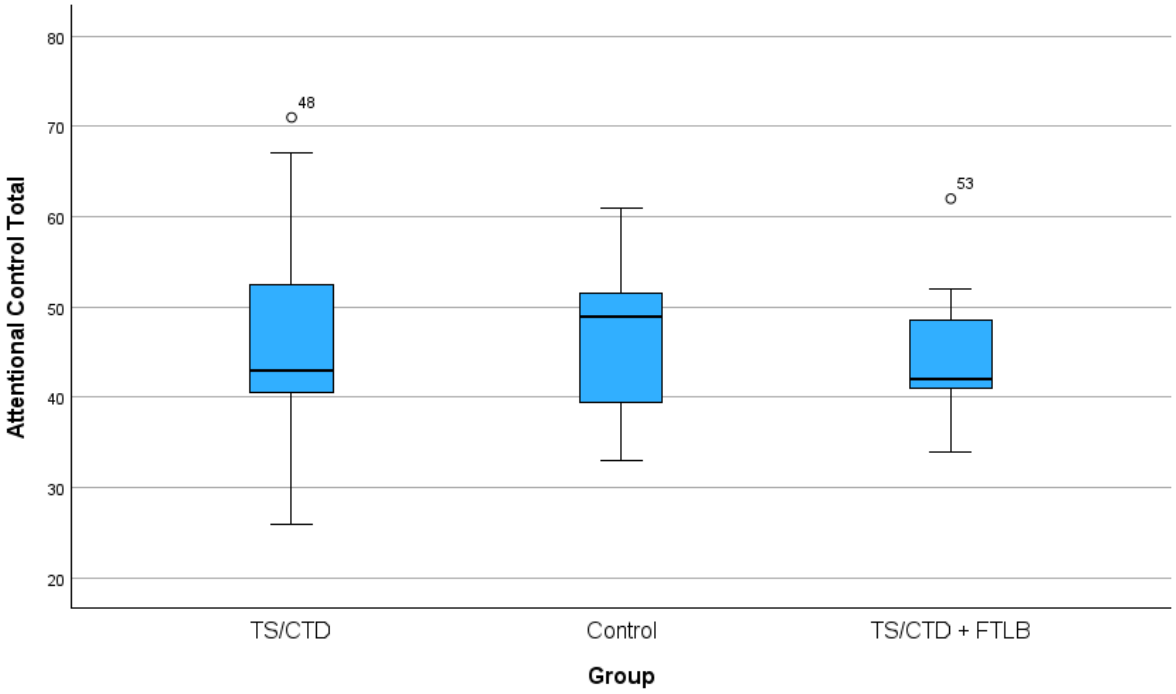
_____	_____	_____
Name of participant	Signature	Date
_____	_____	_____
Name of parent/carer	Signature	Date
_____	_____	_____
Name of Researcher taking consent	Signature	Date

Appendix S Tests for Normality






Normal Q-Q Plot of IAS_Total
for Group= FTLB+CTD
Expected Normal
2
1
0
-1
-2
55
60
65
70
75
80
Observed Value


Attentional Control Total
80
70
60
50
40
30
20
48
53
TS/CTD
Control
TS/CTD + FTLB
Group

Online Resource 1 QualSyst Tool

		Yes (2)	Partial (1)	No (0)	N/A
1	Question / objective sufficiently described?				
2	Study design evident and appropriate?				
3	Method of subject/comparison group selection or source of information/input variables described and appropriate?				
4	Subject (and comparison group, if applicable) characteristics sufficiently described?				
5	If interventional and random allocation was possible, was it described?				
6	If interventional and blinding of investigators was possible, was it reported?				
7	If interventional and blinding of subjects was possible, was it reported?				
8	Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? Means of assessment reported?				
9	Sample size appropriate?				
10	Analytic methods described/justified and appropriate?				
11	Some estimate of variance is reported for the main results?				
12	Controlled for confounding?				
13	Results reported in sufficient detail?				
14	Conclusions supported by the results?				
Total for each category		**x 2 =**	**x 1 =**	**x 0 =**	**x 2 =**
Total sum = (number of "yes" * 2) + (number of partials" * 1)					
Total possible sum = 28 – (number of "N/A" * 2)					
Summary score: total sum / total possible sum					

Online Resource 2 Individual Quality Assessment Scores for Each Included Study

	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject / comparison group selection or source of information / Input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	5. If interventional and random allocation was possible, was it described?	6. If interventional and blinding of investigators was possible, was it reported?	7. If interventional and blinding of subjects was possible, was it reported?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias? Means of assessment reported?	9. Sample size appropriate?	10. Analytic methods described / justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?
Anderson, 2023	1	2	1	2	n/a	n/a	n/a	1	1	2	1	2	2	2
Armstrong-Javor, 2024	2	2	1	1	n/a	n/a	n/a	1	1	2	1	2	0	0
Baizabal-Carvallo, 2023	1	2	1	2	n/a	n/a	n/a	2	1	2	2	1	2	1
Baizabel-Carvallo, 2014	1	2	1	2	n/a	n/a	n/a	0	1	1	1	1	2	1
Berg, 2024[a]	2	2	1	2	n/a	n/a	n/a	2	2	2	2	2	2	2
Buts. 2022	1	2	1	2	n/a	n/a	n/a	1	1	2	1	n/a	2	2

Cavanna, 2022	1	2	1	2	n/a	n/a	n/a	2	0	2	1	1	2	2
Cavanna, 2023	2	2	1	2	n/a	n/a	n/a	1	1	2	1	1	2	1
Cavanna, 2023	1	2	1	2	n/a	n/a	n/a	1	1	2	0	1	2	1
Cavanna,2023	1	2	1	2	n/a	n/a	n/a	1	1	2	1	2	2	2
Demartini, 2015	1	2	1	2	n/a	n/a	n/a	0	0	1	1	n/a	2	2
Ducroizet, 2025	1	2	1	2	n/a	n/a	n/a	1	1	2	1	1	2	2
Duncan, 2024	2	2	1	2	n/a	n/a	n/a	2	2	2	2	1	2	2
Firestone, 2023	1	1	1	2	n/a	n/a	n/a	1	0	1	0	n/a	1	0
Fremer, 2024[a]	1	2	1	2	n/a	n/a	n/a	1	2	2	2	1	2	2
Ganos, 2016	1	2	1	2	n/a	n/a	n/a	0	0	1	1	n/a	2	2
Han, 2022	1	2	1	2	n/a	n/a	n/a	1	1	2	0	1	2	2
Howlett, 2022[a]	2	2	1	2	n/a	n/a	n/a	1	1	2	2	1	2	0
Hull, 2021	1	2	1	2	n/a	n/a	n/a	0	0	1	1	n/a	2	2
Janik, 2021	1	2	1	2	n/a	n/a	n/a	0	0	1	1	1	2	1
Larsh, 2022	2	2	1	2	n/a	n/a	n/a	2	1	2	2	1	2	2
Martino, 2023	1	2	1	2	n/a	n/a	n/a	1	1	2	1	1	2	2
Mathew, 2023	2	2	1	2	n/a	n/a	n/a	1	1	2	1	1	2	1
Maxwell, 2023	1	2	1	1	n/a	n/a	n/a	0	1	2	1	1	1	2

Müller-Vahl, 2024	1	1	1	2	n/a	n/a	n/a	1	1	1	1	1	2	2
Nilles, 2024	2	2	1	2	n/a	n/a	n/a	1	1	2	2	2	2	2
Nilles, 2024	1	2	1	2	n/a	n/a	n/a	1	1	2	2	2	2	2
Okkels, 2023	2	2	1	2	n/a	n/a	n/a	1	1	1	1	n/a	2	2
Paulus, 2021	1	1	1	2	n/a	n/a	n/a	2	2	2	2	2	2	2
Prato, 2023	1	2	1	2	n/a	n/a	n/a	1	1	1	2	1	2	1
Szejko, 2024	2	2	1	2	n/a	n/a	n/a	2	1	2	2	2	2	2
Tomczak, 2024	2	2	1	2	n/a	n/a	n/a	2	2	2	0	1	2	2
Trau, 2022	2	2	1	2	n/a	n/a	n/a	1	1	2	0	2	2	2

[a] Main study quality assessed alongside additional report(s) of same study

2 = Yes, 1 = Partially yes, 0 = No, n/a = not applicable