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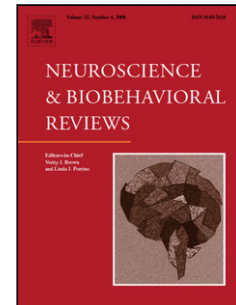
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*Review Article***Prevalence and Clinical Correlates of Self-Injurious Behavior in Tourette Syndrome**

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HIGHLIGHTS

- In Tourette syndrome (TS), tics can be associated with self-injurious behavior (SIB)

- About one third (35%) of patients with TS report SIB of different type and severity
- The overall presence of SIB in TS is associated with obsessive-compulsive behavior
- Subcategorization of SIB would be key to better understand its relationship with TS
- There is a need to develop a standardized assessment/diagnostic tool for SIB in TS

ABSTRACT

Tourette syndrome (TS) is a neurodevelopmental condition characterized by the presence of multiple tics. In addition to common behavioral co-morbidities such as obsessive-compulsive disorder and attention-deficit and hyperactivity disorder, patients with TS can present with self-injurious behavior (SIB): deliberate, non-accidental, repetitive infliction of self-harm without suicidal intent. In order to determine the prevalence and clinical correlates of SIB in TS, we conducted a systematic literature review in accordance to the methodology described in the MOOSE Guidelines for Meta-Analysis and Systematic Reviews of Observational Studies in Epidemiology. The literature search yielded 20 cohort studies that were suitable for quantitative synthesis. Combined results showed that SIB is present in 35% of patients with TS. Obsessive-compulsive behaviors appear to be associated with SIB in patients with TS, and their presence should alert clinicians to the possible co-occurrence of SIB, particularly if the patient has multiple behavioral co-morbidities. Since the reviewed studies were methodologically heterogeneous and were conducted in specialist clinics, our findings cannot be generalized to patients with TS in the wider community.

Keywords: Tourette syndrome; tics; self-injurious behavior; prevalence; obsessive-compulsive behaviors.

1. INTRODUCTION

Tourette syndrome (TS) is a neurodevelopmental disorder characterised by involuntary motor and vocal tics. It has been estimated that TS affects between 0.3% and 1% of the general population, with onset around the age of 6 years and male:female ratio of 3-4:1 (Cavanna and Seri, 2013; Ganos and Martino, 2015; Scharf et al., 2015). Tics, currently defined as sudden, rapid, recurrent, non-rhythmic movements or vocalisations, can be harmful and interfere with the activities of daily life (American Psychiatric Association, 2013; Ganos et al., 2014). The majority of patients with TS present with associated behavioral problems, most commonly obsessive-compulsive disorder (OCD) and attention-deficit and hyperactivity disorder (ADHD) (Martino et al., 2017; Cavanna, 2018). Although tics contribute to the severity of TS, co-morbid behavioral symptoms are core aspects of the assessment of TS-related impairment (Freeman et al., 2000; Cavanna et al., 2012; Eddy et al., 2012; Eapen et al., 2016; Evans et al., 2016).

Self-injurious behavior (SIB) is defined as the deliberate, non-accidental, repetitive infliction of self-harm without suicidal intent (Mathews et al., 2004). SIB is commonly reported in the context of several neuropsychiatric disorders, including Lesch-Nyhan syndrome, autism, and borderline personality disorder (Pies and Popli, 1995). A wide range of SIBs have been reported in the scientific literature on TS, including compulsive skin-picking, self-hitting, self-biting, teeth grinding, and head banging (Mathews et al., 2004). A number of studies have independently estimated the prevalence of SIB amongst patients with TS, but findings have been largely inconsistent (Micheli et al., 1995; Berthier et al., 1996; Freeman et al., 2000; Cath et al., 2001).

In autism, SIB is thought to be related to abnormal sensory processing, whereas the factors associated with SIB in TS are still unknown (Duerden et al., 2012). Although two early studies suggested that SIB is primarily associated with higher tic severity (Robertson et al., 1989; Robertson and Gourdie, 1990), a subsequent large retrospective study reported a positive linear relationship between the number of behavioral co-morbidities and the presence of SIB (Freeman et al., 2000). A more recent study in patients with TS found that the predictors of SIB varied according to the severity of the SIB itself: the presence of obsessive-compulsive symptoms was the main predictor for moderate SIB, whereas impulsivity was a greater predictor than compulsivity for severe SIB (Mathews et al., 2004).

Patients with SIB in the context of TS will not only suffer with the physical pain caused by the behavior itself, but may be distressed by their violent or socially inappropriate movements, which could lead to increased anxiety, particularly if there is a concern about having tics in front of others (Eapen et al., 2016). It is also important to consider the impact of SIB on patients' families and carers: the pathological behavior can be highly distressing and therefore additional support may be beneficial. Since SIB can cause significant morbidity in patients with TS, a better understanding of SIB would improve the health-related quality of life for the patient and others involved in their care. This paper describes the first systematic review of published data on the prevalence and clinical characteristics of SIB in TS. This information is needed to improve the accuracy of diagnosis and aid a better selection of effective interventions as part of a comprehensive management plan for this patient population.

2. METHODS

This systematic review was conducted in accordance to the methodology described in the MOOSE Guidelines for Meta-Analysis and Systematic Reviews of Observational Studies in Epidemiology (Stroup et al., 2000). The literature search was conducted across five electronic databases (PubMed, MEDLINE, EMBASE, PsycINFO, and PsycARTICLES). For each database search, a comprehensive list of synonyms of the search terms was produced and combined using the 'OR' Boolean search operator. The MeSH terms included "Tourette syndrome", "tic disorder", "self injurious behaviour", "self harm", "self mutilation", "comorbidity", "attention deficit hyperactivity disorder", and "obsessive compulsive disorder". The search engine Google Scholar was used to identify grey literature that might not have been detected via the database search. Finally, a manual search of the reference lists of included articles was performed to identify additional relevant studies.

We included original studies on the prevalence of SIB in patients of all ages with a validated diagnosis of TS, as well as studies containing information regarding the possible association of SIB with TS clinical characteristics, such as tic severity, co-morbid OCD and ADHD. Case reports, case series, cross-sectional studies and retrospective cohort studies were deemed eligible for inclusion, whereas qualitative studies were excluded, as they would have

increased the heterogeneity to a level that would not be conducive to quantitative synthesis of the data. Intervention studies were excluded because of their focus on the effectiveness of a treatment intervention, as opposed to the rates and patterns of SIB occurrence.

Studies that met the inclusion criteria were assessed for methodological validity using the Crowe Critical Appraisal Tool (CCAT) (Crowe et al., 2012; Crowe and Sheppard, 2012). The CCAT facilitates the critical appraisal of different study designs by assigning each study a quality score, with the recommended threshold for inclusion set at 30% (Crowe et al., 2011). The use of a validated assessment tool with a high inter-rater reliability score ensured consistency when appraising the quality of the reviewed studies (Crowe et al., 2011).

3. RESULTS

The search strategy retrieved a total of 314 relevant articles. The additional search for grey literature identified a further 837 studies. After reading the full text of the relevant articles, a total of 43 studies were found to meet the inclusion criteria for this systematic review. Of these, 33 articles were deemed suitable for inclusion based on their methodology: 20 cohort studies (Nee et al., 1980; Stefl, 1984; Robertson et al., 1989; Wand et al., 1993; Berthier et al., 1998; Lichter et al., 1999; Freeman et al., 2000; Cath et al., 2001; Robertson et al., 2002; Ohta and Kano, 2003; Eapen et al., 2004; Mathews et al., 2004; Martino et al., 2007; Robertson et al., 2008; Haddad et al., 2009; Kano et al., 2010; Szejko et al., 2019), 1 combined cohort study plus case series (Cheung et al., 2007), 2 case series (Muller-Vahl et al., 2000; Cavanna et al., 2010), and 10 case reports (Robertson et al., 1990; Anandan et al., 2004; Hood et al., 2004; Lim et al., 2004; Leksell and Edvardson, 2005; Bajwa et al., 2007; Banda et al., 2009; Cavanna et al., 2009; Duits et al., 2012; Fornaro et al., 2012). The process leading to the identification, screening, assessment of eligibility, and inclusion of studies is summarised in a PRISMA flow chart (Moher et al., 2009) (**Figure 1**).

[PLEASE INSERT FIGURE 1 HERE]

Following methodology assessment with the CCAT tool, the studies were separated into cohort studies (**Table 1**) and case reports/series (**Table 2**).

[PLEASE INSERT TABLE 1 HERE]

[PLEASE INSERT TABLE 2 HERE]

Data from the 20 cohort studies were quantitatively analysed to produce a pooled estimate of the prevalence of SIB in TS and assess the relationship between SIB and co-morbid behavioral problems. The included studies recruited a total of 7,261 participants: the proportion of males across the studies was 74%, and the average age was 22 years. Both gender and age distribution were similar across the studies.

SIB was found to be present in approximately a third of patients with TS (35%). Prevalence estimates for SIB in TS ranged from 11% in a Japanese study with a cohort of 88 participants (Kano et al., 2010), to 78% in a Dutch study of 32 participants (Cath et al., 2001). Only three studies reported a prevalence of less than 20% (Comings and Comings, 1985; Freeman et al., 2000; Kano et al., 2010) and only one study reported a prevalence higher than 50% (Cath et al., 2001). The pooled prevalence figures of co-morbid OCD and ADHD in the combined TS population were both 48%. When analysing the relationship between SIB and clinical correlates across the reviewed studies, obsessive-compulsive behaviors were found to be associated with the presence of SIB.

The case reports and case series included data from a total of 45 patients, 80% of whom were male, with a mean age of 28. The mean age at TS diagnosis across the case reports was 8 years, and the average tic severity was 55%, as measured on the Yale Global Tic Severity Scale (YGTSS) total scores (Leckman et al., 1989). All 45 patients had at least one psychiatric co-morbidity, with 67% reporting two or more co-morbidities. The prevalence of behavioral co-morbidities was as follows: 16 of the 45 patients reported obsessive-compulsive behaviors, 16 were diagnosed with ADHD, 12 reported anxiety disorder, 12 suffered from depression, 10 had other affective disorders, 8 had oppositional defiant disorder and 7 were diagnosed with co-morbid OCD. The timeline of the development of SIB remains unclear, with a number of case-reports documenting that SIB developed several years after the diagnosis of TS. Two studies reported that SIB developed 5 years after the TS diagnosis (Hood et al., 2004; Duits et al., 2012), and another study found that SIB developed 12 years after the diagnosis of TS (Robertson et al., 1990). However, this was not always the case: one study documented that SIB and TS developed simultaneously when the patient was 4 years old (Leksell and Edvardson, 2005), and another documented the case of a patient who was diagnosed with TS at the age of 13, but did not develop SIB until he was 47 years old (Bajwa et al., 2007). The most frequently reported SIBs were head banging and hitting oneself in the head, followed by the poking of sharp objects into the body, and tongue or lip biting. Facial grimacing, eye blinking, and copropraxia were the most commonly observed motor tics, whereas coprolalia was the most commonly reported vocal tic.

4. DISCUSSION

SIB is a debilitating behavior that is relatively common in the TS population and can have a significant impact on the health-related quality of life of both patients and their families/carers. The present systematic literature review provided the first attempt to quantitatively synthesise prevalence data on SIB in TS. The results of the systematic review indicated that SIB is present in approximately one third (35%) of patients with TS.

The findings of this review suggest a link between SIB and obsessive-compulsive behaviors and are supported by the results of the largest study to date, a multi-centre study by Freeman et al. on 3500 patients with TS (Freeman et al., 2000). This study used subgroup analysis of co-morbidities and reported that

SIB was present in 14% of the patients with TS and co-morbid OCD, 5% of the patients with TS and co-morbid ADHD, and 4% of the patients with TS only. This study also reported that the relative risk of SIB in patients with TS only compared to those with TS and a psychiatric co-morbidity was 3.82 ($p < 0.001$).

This review did not attempt to categorise SIB according to severity. Nevertheless, the heterogeneity of behaviors reported across the reviewed studies could indicate that mild SIB is a different clinical phenomenon to severe SIB. This hypothesis is in line with the results of the study conducted by Mathews et al., who concluded that mild-to-moderate SIB was correlated with obsessive-compulsive behaviors, including compulsive aggression, OCD and overall number of obsessions, whereas severe SIB was correlated with impulse dysregulation, including risk-taking behaviors and episodic rages (Mathews et al., 2004). Interestingly, the results of this study also suggested that both types of SIB correlated with tic severity.

Only a minority of the reviewed studies used the YGTSS, a validated and accessible tool to measure tic severity (Mathews et al., 2004; Martino et al., 2007; Haddad et al., 2009; Szejko et al., 2019). A few studies categorised tic severity as 'mild', 'moderate', or 'severe' (Stefl, 1984; Freeman et al., 2000), and another study defined severity according to whether the tics required treatment and interfered with daily life (Comings and Comings, 1985). With limited data regarding tic severity in patients with SIB in the context of TS, it was not possible to assess the possible relationship between SIB and tic severity.

Furthermore, the varying nature of SIB caused difficulties when comparing the results of the reviewed studies, with severity ranging from mild scratching, tongue biting, and head banging to more severe manifestations such as self-strangulation and eye enucleation. The fact that very few studies included in the present review differentiated between mild and severe SIB could account for the high prevalence estimates and large variability across studies. The lack of distinction between mild and severe SIB across most of the reviewed studies implies that the high variability in the reported prevalence rates do not allow a reliable estimate of the phenomenon among the TS population and prompts further considerations. Although technically clinical manifestations ranging from skin-picking and teeth grinding to stabbing/burning self and violent head jerks could all meet the definition of SIB, the resulting category is arguably too heterogeneous to draw reliable conclusions. For example, there is a degree of overlap between milder body-focused repetitive behaviors (intense urges leading to repetitive biting, picking, and pulling that can cause self-injury) and obsessive-compulsive behaviors. From a clinical perspective, the inclusion of such body-focused repetitive behaviors in the SIB category can be problematic: both patients with TS and their families should be reassured that while these

actions can lead to self-injury, they are not intentionally 'self-injurious' in the way that the SIB terminology is commonly used. One possible solution would be to separate the SIB category into more homogenous subgroups. Favazza and Simeon (1995) have previously characterized four descriptive types of non-suicidal self-injury: major, stereotypic, compulsive, and impulsive. Major non-suicidal self-injury includes infrequent violent acts that destroy significant body tissue, such as eye enucleation and amputation of body parts, and is mainly observed in response to thought abnormalities in patients with psychotic disorders. Stereotypic non-suicidal self-injury acts, most commonly associated with mental retardation (e.g. Lesch-Nyhan syndrome, Cornelia de Lange syndrome), include monotonously repetitive head banging, eye gouging, lip/cheek/tongue/finger biting, and face/head slapping. Compulsive non-suicidal self-injury encompasses repetitive behaviors such as severe skin scratching and nail biting, hair pulling, and skin digging. Finally, impulsive non-suicidal self-injury consists of episodic acts of skin cutting/burning/carving, sticking pins or other objects under the skin and/or into the chest or abdomen, interfering with wound healing, and smashing hand/foot bones (Favazza, 2012). While the heterogeneity of the reviewed cohort studies does not allow to analyze available data from the TS population according to such subcategorization, the findings of the individual case reports/series seem to confirm the possible association between milder SIB (stereotypic; compulsive) and obsessionality. Likewise, the hypothesis of a relationship between more severe forms of SIB (major; impulsive) and impulse dysregulation, as previously suggested by Mathews et al. (2004), cannot be ruled out. It has also been highlighted that the phenomenologic distinction between compulsive and impulsive SIB is mainly heuristic, as in clinical settings there may be significant overlap: for example, patients with certain impulsive behaviors, such as self-cutting, can go on to develop apparently compulsive repetitive SIB (Favazza and Simeon, 1995).

The methodological quality scores of the cohort studies ranged from 35% to 83%, with an average score of 61% according to the employed study quality tool (CCAT). Studies with larger sample sizes tended to score higher on the CCAT. The quality of the case reports ranged between 30% and 70%, with an average score of 51%. Although the quality of the cohort studies was on average better than the quality of the case reports and case series, only three of the 20 cohort studies had primary outcomes that related directly to SIB, response rates were rarely reported, and most studies reported multiple outcomes using varying methods. Moreover, the present review was subject to multiple methodological biases. For example, most studies used retrospective data collection, resulting in recall bias. Both cohort studies and case reports mainly recruited patients from tertiary care centres, resulting in ascertainment and referral bias.

Thus, the findings from this review cannot be generalised to patients with TS in the wider community. The search strategy restricted results to articles published in English language, however it is thought that the extent to which language bias affects validity has reduced in recent years due to a shift towards publication of studies in English (Green and Higgins, 2001). Additionally, a recent study found no evidence of a systematic bias from the use of language restrictions used in systematic reviews (Morrison et al., 2012).

In the present review, strict selection criteria were implemented in an attempt to reduce clinical heterogeneity and to improve the robustness of findings. Specific symptoms belonging to the 'TS spectrum', such as obsessive-compulsive behaviors, were often reported in studies but were difficult to define because of the lack of agreement on lower bounds. Therefore, the association between SIB and obsessive-compulsive behaviors that emerged from the reviewed studies should be interpreted with caution. Similarly, SIB was not defined consistently across the literature, making comparisons between the reviewed studies difficult. Investigations into the sources of variability were restricted by the limited number of studies included in the review. Specifically, subgroup analysis revealed that the prevalence of SIB was higher in studies published after 2000, studies with higher methodological quality, and studies conducted in the United Kingdom.

The present review has identified several key areas that could be the focus of further research into SIB in TS. Firstly, the development of a standardised assessment or diagnostic tool to better define the presence and severity of SIB in TS would greatly improve future research and clinical practice. Specifically, by incorporating the proposed subcategorization of SIB into assessment protocols it would be possible to achieve a better understanding of its relationship with TS. This review has identified a few clinical variables worthy of further investigation to better understand the etiology of SIB. Obsessive-compulsive behaviors were found to associated with SIB in patients with TS, and their presence should alert the physician to the possible co-occurrence of SIB, particularly if the patient has multiple behavioral co-morbidities. Future studies can build upon this evidence to determine the full extent to which psychiatric co-morbidities influence SIB and whether other factors can increase the likelihood of developing SIB in the context of TS. The use of individual participant data in a large prospective cohort study would be necessary to determine the longitudinal time course of SIB in TS and would allow the researchers to better control for confounders.

Several case reports included in this review documented the clinical management of SIB in patients with TS and included information about treatment interventions beyond pharmacological therapy, including behavioral strategies (e.g. fixed interval reinforcement and extinction) and surgical procedures (deep brain stimulation, limbic leucotomies). Other studies have found that medications that are commonly used in patients with TS (first-generation antidopaminergic agents and selective serotonin reuptake inhibitors) are effective in the treatment of SIB in the context of autism (Villalba and Harrington, 2000). Large controlled trials investigating the effectiveness of treatment interventions for SIB in TS could provide clinicians with further information to improve the health-related quality of life of this patient population.

Declarations of interest

None.

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FIGURE

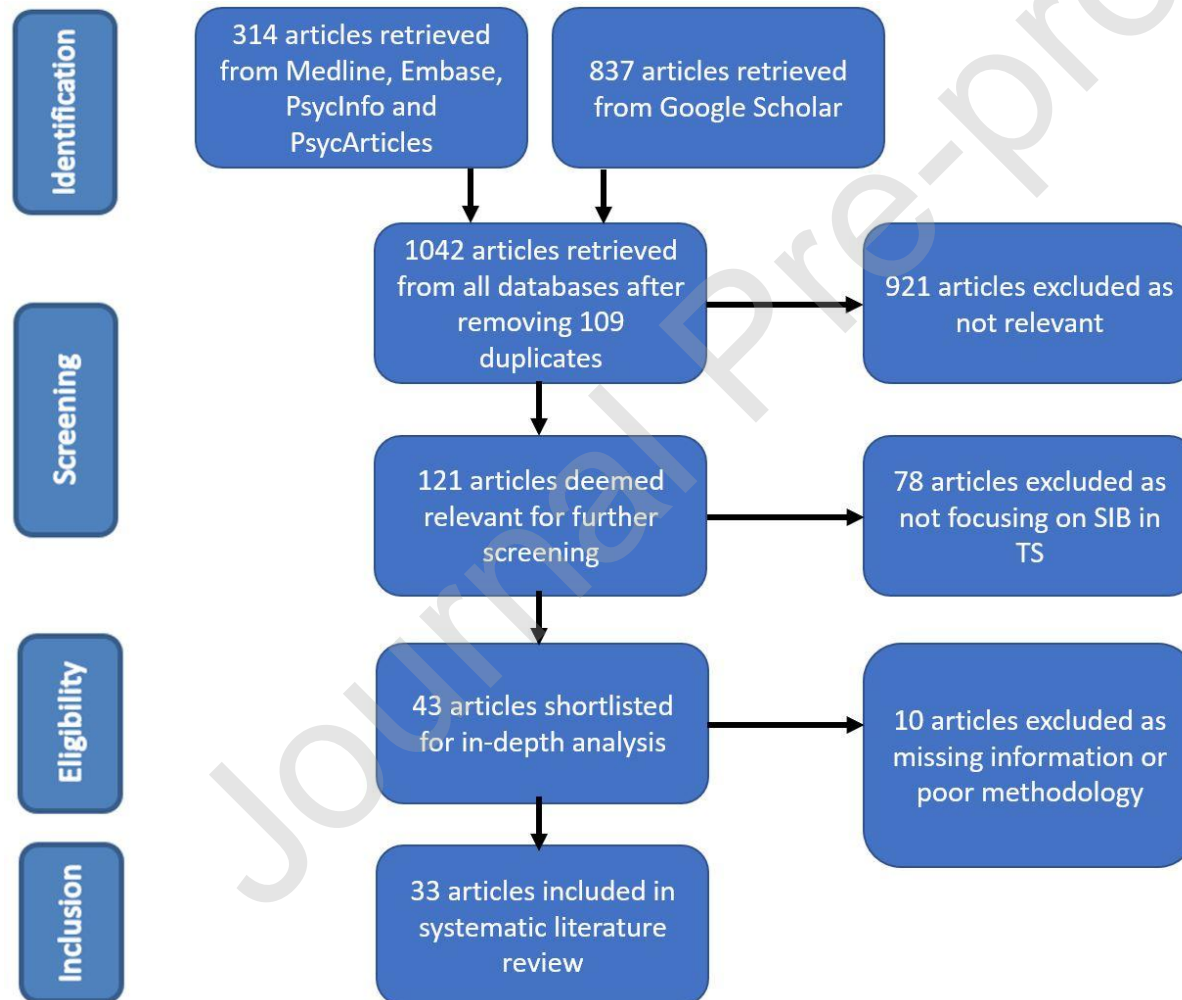


Figure 1. PRISMA flow diagram outlining the identification, screening, assessment for eligibility, and inclusion of studies in the present review.

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TABLES

Table 1. Summary of cohort studies on self-injurious behaviour in patients with Tourette syndrome.

Study	Country	N	Male (%)	Mean age (years)	SIB (%)	OCD (%)	ADHD (%)	Other co-morbidities (%)	Other findings	Quality score (%)
Freeman et al. 2000	22 countries	3500	81	-	14	27	60	OCB 32% LD 23% Affective disorder 20% Anxiety disorder 18% CD/ODD 15% PDD 5%	SIB present in 13% of males, 18% of females ($p<0.005$) SIB present in: <ul style="list-style-type: none"> TS + ADHD + OCD = 13% TS + Anxiety disorder = 11% TS + ADHD = 5% TS + Affective disorder = 5% TS-only = 4% RR of SIB if comorbidity present = 3.82 (95%CI 2.37-6.15) ($p<0.001$)	78
Robertson et al. 2015	UK	578	73	25	30	40	56	Affective disorder 51% Anxiety disorder 43% Aggression 30% CD 15% ASD 10%	-	63
Wand et al. 1993	Canada	446	75	20	31	-	-	Anxiety 67% OCB 65% Hyperactivity 60% Aggression 56%	SIB in 33% of <18 years and in 34% of >18 years (no significance difference)	35
Stefl 1984	USA	431	78	-	34	-	-	Anxiety 80% OCB 74 % Hyperactivity 73% Aggression 64%	-	53

Study	Country	N	Male (%)	Mean age (years)	SIB (%)	OCD (%)	ADHD (%)	Other co-morbidities (%)	Other findings	Quality score (%)
Robertson et al. 2008	UK	410	76	20	43	83	59	Aggression 45%	-	68
Cheung et al. 2007	USA	333	83	20	22	63	65	Affective disorder 29% (suicidal ideation 9.0%) Anxiety disorder 25% ODD 23%	Malignant TS (≥ 1 hospitalisation or ≥ 2 Emergency Department visits) had higher rates of SIB compared to non-malignant 65% vs 20%	53
Mathews et al. 2004	USA and Costa Rica	297	77	23	33	40	40	ADHD + OCD 26%	Mild/moderate SIB correlated with OCB/OCD; severe SIB correlated with impulse dysregulation. Both correlated with tic severity	83
Comings and Comings 1985	USA	250	75	18	13	-	54	Aggression 61% OCB 32%	-	53
Szejko et al. 2019	Poland	165	76	14	39	18	25	Anxiety disorder 48% OCB 45% Aggression 36% Significant social skill problems 16% Affective disorder 15%	ADHD ($p=0.016$) and OCD ($p=0.042$) were determined as risk factors for the development of SIB	68
Haddad et al. 2009	UK	143	72	33	43	41	45	Aggression 26%	No significant difference in prevalence of SIB between patients with TS + ADHD and patients with TS only	68
Martino et al. 2007	UK	128	77	23	44	-	-	-	No significant difference in prevalence of SIB between ANeA+ and ANeA- patients	73
Lichter et al. 1999	USA	111	87	10	23	-	-	-	8% of patients had >1 type of SIB Patients with OCB and SIB had 68 times greater odds of being in the bilineal transmission group as opposed to unilineal or sporadic groups	63
Eapen et al. 2004	UK	91	64	29	44	-	54	Compulsive behaviors 58% Obsessional thoughts 46% Aggression 34%	Positive correlation between OCB and SIB ($r=4.9$, $p=0.023$) SIB correlated with coprophenomena ($r=3.8$, $p=0.037$), echophenomena ($r=4.2$, $p=0.031$), aggression ($r=26.9$, $p=0.000$), and ADHD ($r=12.0$, $p=0.001$) No correlation between SIB and gender	68

Study	Country	N	Male (%)	Mean age (years)	SIB (%)	OCD (%)	ADHD (%)	Other co-morbidities (%)	Other findings	Quality score (%)
Robertson et al. 1989	UK	90	70	24	33	-	-	-	SIB related to cumulative number of motor tics (p=0.006)	58
Kano 2010	Japan	88	76	15	11	-	-	OCB 42% Hyperkinetic disorder 28% Aggression 27% Learning disability 10%	-	50
Robertson et al. 2002	UK	57	79	11	40	-	60	Compulsive behaviors 41% Aggression 30% Obsessional thoughts 25%	-	58
Nee et al. 1980	USA	50	-	-	48	-	-	OCB 68% Learning disability 40% Antisocial behavior 26%	-	63
Cath et al. 2001	Holland	32	53	34	78	-	-	OCB 74%	-	68
Ohta and Kano 2003	Japan	31	90	20	36	39	16	OCB 74% Anxiety disorder 13%	SIB present in 64% of patients with tics+co-morbidity and 20% of patients with tics only	43
Berthier et al. 1998	Spain	30	43	32	40	80	40	Anxiety disorder 73% CD/ODD 37% ASD 3%	-	55

Abbreviations. SIB: Self-injurious behavior, ADHD: Attention-deficit and hyperactivity disorder, OCD: Obsessive-compulsive disorder, OCB: Obsessive-compulsive behaviors, CD: Conduct disorder, LD: Learning disability, PDD: Pervasive development disorder, RR: Relative risk, ASD: Autistic spectrum disorder, ODD: Oppositional defiant disorder, ANeA: Anti-neuronal antibodies.

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Table 2. Summary of case reports and case series on self-injurious behaviour in patients with Tourette syndrome.

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Anandan et al. 2004	USA	14	M	Teeth extraction, ingestion of toxic substances, stabs himself with various objects (e.g. coat hanger tip)	Tongue rolling, nose touching	Grunting	N/A	+	+		55
Bajwa et al. 2007	USA	50	M	Violent head jerks (causing painful cervical spine extension and myelopathy), slamming forearms against forehead, elbowing ribs	Blinking, abdominal tensing, leg tics	Grunting, loud noises, coprolalia	35.5	+	-	Affective disorder	63
Banda et al. 2012	USA	14	M	Head punching (wearing padded helmet and gloves)	Feet touching when walking	Barking noise	N/A	-	-	Affective disorder Autism	70
Duits et al. 2012	Netherlands	20	F	Hitting head and arm, poking sharp objects into body, scratching until drawing blood	Blinking, licking, cheek biting, tapping with right foot	Coughing, barking, uttering sounds, coprolalia	42	-	-	Affective disorder PDD	50
Leksell and Edvardson 2005	Sweden	4	F	Teeth grinding affecting the attachment of teeth	Teeth grinding	-	N/A	-	-	OCB Hyperactive behavior	50
Lim et al. 2004	USA	25	M	Poking finger into eye, punching in periorbital region	Blinking, blepharospasm, clapping	-	N/A	-	-	Affective disorder	33
Robertson et al. 1990	UK	19	M	Head banging, lip biting, burning himself with cigarettes	Winking, facial grimacing	Coprolalia	N/A	-	-	OCB	30

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Fornaro et al. 2011	Italy	54	M	Piercing tongue with pencils and other sharp objects	Blinking, nose twitching, head jerking, shoulder shrugging, mouth touching	Coughing, grunting, palilalia	95	+	-	Fibromyalgia	55
Hood et al. 2004	USA	16	F	Tongue biting	Unspecified	Unspecified	N/A	+	-	-	50
Cavanna et al. 2009	UK	29	M	Head banging	Blinking, frowning, teeth grinding, facial grimacing, spitting, blowing, head nodding, shoulder shrugging, kicking, squatting, copropraxia	Sniffing, noisy breathing, coughing throat clearing, coprolalia	39	+	-	-	35
Müller-Vahl et al. 2000	Germany	31	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Anxiety disorder Severe OCB	53
Müller-Vahl et al. 2000	Germany	25	M	Unspecified	Unspecified	Unspecified	N/A	-	+	Anxiety disorder Affective disorder Mild OCB	53
Müller-Vahl et al. 2000	Germany	64	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Affective disorder Moderate OCB	53
Müller-Vahl et al. 2000	Germany	30	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Anxiety disorder Affective disorder Moderate OCB	53
Müller-Vahl et al. 2000	Germany	52	M	Unspecified	Unspecified	Unspecified	N/A	-	+	Anxiety disorder Affective disorder Moderate OCB	53

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Müller-Vahl et al. 2000	Germany	24	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Anxiety disorder Affective disorder Moderate OCB	53
Müller-Vahl et al. 2000	Germany	28	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Anxiety disorder Affective disorder Severe OCB	53
Müller-Vahl et al. 2000	Germany	38	M	Unspecified	Unspecified	Unspecified	N/A	-	-	Anxiety disorder Affective disorder Severe OCB	53
Cheung et al. 2007	USA	45	F	Unspecified	Unspecified	Complex vocal tics	N/A	+	-	Affective disorder Anxiety disorder	65
Cheung et al. 2007	USA	36	M	Unspecified	Copropaxia	Complex vocal tics, coprolalia	N/A	+	+	Affective disorder Anxiety disorder	65
Cheung et al. 2007	USA	34	M	Unspecified	Unspecified	Unspecified	N/A	+	-	-	65
Cheung et al. 2007	USA	24	M	Unspecified	Unspecified	Unspecified	N/A	+	-	Anxiety disorder	65
Cheung et al. 2007	USA	23	M	Unspecified	Unspecified	Unspecified	N/A	+	-	-	65
Cheung et al. 2007	USA	21	M	Unspecified	Copropaxia	Complex vocal tics	N/A	+	+	Affective disorder (with suicidal ideation) ODD	65
Cheung et al. 2007	USA	19	M	Unspecified	Unspecified	Complex vocal tics	N/A	+	+	-	65
Cheung et al. 2007	USA	17	M	Unspecified	Unspecified	Complex vocal tics	N/A	+	+	-	65

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Cheung et al. 2007	USA	17	F	Unspecified	Copropaxia	Complex vocal tics, coprolalia	N/A	+	-	Affective disorder (with suicidal ideation) ODD	65
Cheung et al. 2007	USA	17	M	Unspecified	Copropaxia	Complex vocal tics, coprolalia	N/A	+	+	Affective disorder Anxiety disorder	65
Cheung et al. 2007	USA	15	M	Unspecified	Unspecified	Complex vocal tics	N/A	+	+	ODD	65
Cheung et al. 2007	USA	11	M	Unspecified	Unspecified	Complex vocal tics, coprolalia	N/A	+	+	Affective disorder (with suicidal ideation) ODD	65
Cheung et al. 2007	USA	38	M	Unspecified	Unspecified	Unspecified	N/A	+	-	Affective disorder	65
Cheung et al. 2007	USA	35	M	Unspecified	Unspecified	Unspecified	N/A	+	+	Affective disorder (with suicidal ideation) Anxiety disorder	65
Cheung et al. 2007	USA	27	F	Unspecified	Unspecified	Complex vocal tics	N/A	+	-	Affective disorder (with suicidal ideation) ODD	65
Cheung et al. 2007	USA	19	M	Unspecified	Unspecified	Complex vocal tics, coprolalia	N/A	+	+	Affective disorder Anxiety disorder ODD	65

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Cavanna et al. 2010	UK and USA	11	M	Unspecified	Facial grimacing, shoulder shrugging, limb movements, echopraxia	Throat clearing, sniffing, snorting coughing, muttering, coprolalia, echolalia, palilalia, pathological laughter	42	-	+	OCB	60
Cavanna et al. 2010	UK and USA	14	M	Unspecified	Facial grimacing, echopraxia	Sniffing, tongue clicking, echolalia, pathological laughter	57	-	+	ODD	60
Cavanna et al. 2010	UK and USA	44	F	Unspecified	Torso twisting and shivering, copropraxia	Yawning, coprolalia, echolalia, palilalia, pathological laughter	80	-	+	ODD NOSIB	60
Cavanna et al. 2010	UK and USA	29	F	Unspecified	Facial grimacing, shoulder shrugging, dystonic movement of upper body, adjusting clothing	Grunting, coprolalia, echolalia, pathological laughter	26	+	-	-	60

Study	Country	Age	Sex	SIB Type	Motor Tics	Vocal Tics	Tic Severity (YGTSS)	OCD	ADHD	Other Psychiatric Comorbidities	Quality Score
Cavanna et al. 2010	UK and USA	57	M	Unspecified	Facial grimacing, Blinking, winking, staring, shoulder shrugging, arm swinging, hands in mouth, echopraxia	Grunting, barking, squeaking, noisy breathing, moaning, coprolalia, palilalia, pathological laughter	41	-	-	OCB Panic disorder	60
Cavanna et al. 2010	UK and USA	26	M	Unspecified	Blinking, finger clicking and tapping, left leg twitching, orchestrated jaw movements and lip puckering	Sniffing, echolalia, palilalia, pathological laughter	85	+	-	Affective disorder	60
Cavanna et al. 2010	UK and USA	13	M	Unspecified	Facial grimacing, shoulder shrugging, pirouetting, abnormal gait, copropraxia, echopraxia	Throat clearing, gasping, wailing, blowing raspberries, echolalia, palilalia, pathological laughter	61	-	+	OCB	60

Abbreviations: SIB: Self-injurious behavior, YGTSS: Yale Global Tic Severity Scale, ADHD: Attention-deficit and hyperactivity disorder, OCD: Obsessive-compulsive disorder, OCB: Obsessive-compulsive behaviors, PDD: Pervasive development disorder, ODD: Oppositional defiant disorder, NOSIB: Non-obscene socially inappropriate behavior, N/A: Not available, +: present, -: not present.

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