



Self-esteem in adults with Tourette syndrome and chronic tic disorders: The roles of tic severity, treatment, and comorbidity

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ABSTRACT

Background: Tourette syndrome (TS) and chronic tic disorders (CTD) are stigmatizing disorders that may significantly impact self-esteem. Alternatively, comorbid psychiatric illnesses may affect self-esteem more than tics themselves. Extant research on self-esteem in TS/CTD is limited, has inconsistently examined the effect of comorbidities on self-esteem, and yields mixed findings.

Method: This study aimed to clarify the roles of tics versus comorbid diagnoses on self-esteem in a large, carefully diagnosed sample of adults with TS/CTD ($N = 122$) receiving 10 weeks of Comprehensive Behavioral Intervention for Tics (CBIT) or Psychoeducation and Supportive Therapy (PST).

Results: Baseline self-esteem did not differ between adults with TS/CTD only and normative means, whereas self-esteem was significantly lower among adults with TS/CTD with a comorbid psychiatric illness. In a multiple regression testing the baseline association between tic severity, presence of comorbid psychiatric illness, and depression severity with self-esteem, comorbidity and depression severity were significantly associated with self-esteem, whereas tic severity was not. Finally, using a generalized linear model, we tested the effects of treatment assignment, comorbidity, and their interaction on changes in self-esteem across treatment, controlling for baseline depression severity. Results showed that for those with a comorbid illness, self-esteem improved significantly more with CBIT than with PST.

Conclusions: Comorbid illnesses appear to affect self-esteem more so than tics among adults with TS/CTD. Therapeutic attention should be paid to treating comorbid diagnoses alongside tics when treating TS/CTD.

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1. Introduction

Tourette syndrome (TS) and chronic tic disorders (CTD) are stigmatizing and often impairing neurodevelopmental disorders, characterized by the presence of tics (sudden, repetitive, rapid movements or vocalizations) for at least 1 year [1]. A diagnosis of TS requires that

both motor and vocal tics have been present, while CTD requires either a motor or vocal tic [1]. TS/CTD onsets in childhood, with tic prevalence reducing dramatically by early adulthood to a rate of approximately 1 per 2000 adults [2].

Due to the visible and audible nature of tics, individuals with TS report stigma, discrimination, and social exclusion [3–5]. For example, one study instructed school-aged children to rank the popularity of their peers [6]. Children with TS were ranked as less popular than classmates, and 35% of children with TS were ranked lowest in their class on at least one factor of the peer ranking questionnaire [6].

These psychosocial problems appear to persist across the lifespan in individuals with TS/CTD. In a sample of 574 adults with CTDs, 68% reported feeling “abnormal” because of tics, and 68% reported experiencing discrimination due to tics (e.g., being asked to leave a public setting)

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[3]. On this survey, 42.8% of the sample avoided group activities and 38.4% avoided being in certain public places. Moreover, 12.6% indicated that they had chosen not to pursue a job promotion due to tics, and 11.9% avoided a job interview due to tics [3].

Given the psychosocial impact of tics, clinical reports of TS/CTD frequently highlight the direct negative effect that tics may have on self-esteem [7–9], one's evaluation of his or her global self-worth [10]. The *scar model* of self-esteem posits that negative experiences such as the discriminatory social interactions experienced by those with TS/CTD lead affected individuals to judge themselves harshly, which erodes self-esteem over time [11].

Despite clinical evidence that TS/CTD may negatively affect one's self-esteem, studies on self-esteem in TS/CTD are limited, utilize small sample sizes, and have yielded mixed results. Among studies of children and adolescents, four studies have documented significantly lower self-esteem or self-concepts in children with TS/CTD than published norms or comparison groups [12–15]. On the other hand, two studies failed to find significantly lower self-esteem in children with TS/CTD compared to normative means or clinical comparisons [6,16]. Of note, self-concept is a closely related construct to self-esteem, and refers to one's view of him or herself based on past success and failure experiences [17]. Three studies also examined the association between self-esteem or self-concept and tic severity in children, adolescents, and emerging adults [12,14,17]; two studies documented non-significant associations between self-esteem or self-concept and tic severity [14,17], whereas the other study documented a significant relationship [12]. Studies in the adult literature are scarce, yet more consistent. Three studies have shown self-esteem scores falling within a normative range [18–20]. Taken together, few studies with small sample sizes have examined self-esteem in TS/CTD, and the results are inconsistent across trials.

One possible explanation for mixed findings in the extant literature is the failure of almost all prior trials to account for the effect of comorbid psychiatric diagnoses on self-esteem [12,15]. Approximately 88% of TS/CTD cases present with comorbid psychiatric illnesses [21]. Most commonly, TS/CTD presents with obsessive compulsive disorder (OCD), attention deficit/hyperactivity disorder (ADHD), anxiety disorders, and depression [4,21]. Psychiatric illnesses are negatively correlated with self-esteem [22,23]. In particular, a strong, consistent relationship has been documented between depression and self-esteem [22]. Thus, it is possible that comorbid psychiatric illnesses have a clearer negative impact on self-esteem than one's tics, and that a failure of past trials to consistently account for comorbidity has contributed to mixed findings, underscoring a major limitation of the current literature.

Comorbidities are examined in only four of the studies described above, and among these studies, findings were somewhat clearer. Among the childhood, adolescent, and emerging adulthood studies, Hanks and colleagues [12] showed that those with CTD and comorbid psychiatric diagnoses had lower self-concepts compared to those with CTD alone. Moreover, self-esteem in this sample was significantly correlated with ADHD, OCD, and depression severity, in addition to tic severity [12]. However, the authors did not examine whether self-concept differed among those with CTD alone and normative means. Likewise, Silvestri and colleagues [17] documented significantly lower self-concepts among adolescents and emerging adults with TS and a comorbid psychiatric diagnosis, compared to those with TS alone. More severe anxiety and depression symptoms were associated with lower self-concept scores [17]. As with Hanks et al. [12], however, the authors did not compare self-concept among those with TS alone to normative means. Finally, Khalifa and colleagues [14] documented significantly lower self-esteem with regard to social relationships among participants with TS and ADHD, compared to those with TS alone. Among the adult studies, Thibert et al. [20] found that adults with TS and comorbid OCD had lower self-concepts than normative means, while self-concepts of those with TS alone did not differ from that of normative means.

Taken together, few prior studies with small samples have empirically examined levels of self-esteem in TS/CTDs. A critical limitation of prior research on self-esteem in TS/CTDs is its inconsistent approach to examining comorbid psychiatric illnesses or concurrent depression, which occur at high rates in tic disorders [4,21] and which also may be associated with lower self-esteem [22,23]. Given the emphasis on self-esteem in clinical conceptualizations of TS/CTD and the mixed findings in the extant literature, the present study's overarching aim was to more systematically clarify the role of tics versus comorbid psychiatric diagnoses on self-esteem in a large, carefully diagnosed sample of adults with TS/CTD ($N = 122$).

Even less is known about the impact of treatment on self-esteem in TS/CTD. One prior waitlist-controlled study of CBT for tics and body-focused repetitive behaviors (BFRBs) (e.g., trichotillomania) based on habit reversal therapy (HRT) examined self-esteem at pre- and post-treatment [19]. In this combined sample of participants with TS/CTD or BFRBs, self-esteem increased significantly in the treatment condition, with small effects [19]. No significant increase in self-esteem was observed in the waitlist group. However, the authors did not test whether increases in self-esteem were significantly greater in the treatment condition compared to the waitlist condition, nor did they examine changes in self-esteem among TS/CTD participants separately from those with BFRBs. Therefore, to comprehensively contribute to the literature on self-esteem in TS/CTD, the present study also aimed to build an initial understanding of the impact of treatment on self-esteem for adults with TS/CTD. Using data from a large-scale, multisite, randomized trial of Comprehensive Behavioral Intervention for Tics (CBIT) versus Psychoeducation and Supportive Therapy (PST), we examined self-esteem at baseline, as well as changes in self-esteem across 10 weeks of treatment. CBIT is the recommended empirically supported psychotherapeutic intervention for TS/CTD [24,25], and PST is one of the most common psychotherapeutic interventions obtained in the community.

The first aim was to evaluate whether baseline self-esteem differs significantly between adults with TS/CTD only, adults with TS/CTD and a current comorbid psychiatric diagnosis, and published normative adult levels of self-esteem. Based on existing literature [12,14,20], we expected those with TS/CTD and a comorbid diagnosis would have significantly lower self-esteem compared to TS/CTD only or adult norms. We hypothesized that self-esteem would not differ significantly between those with TS/CTD only and adult norms [20]. The second aim was to examine correlates of baseline self-esteem in adults with TS/CTD. To this end, we examined whether baseline tic severity, presence (yes/no) of a current comorbid psychiatric illness, and baseline depression severity were significantly associated with baseline levels of self-esteem. We hypothesized that each predictor would be significantly, independently related to self-esteem. Finally, the third aim was to examine the impact of treatment assignment, current comorbid illness, and their interaction on self-esteem, accounting for depression severity at baseline. Given that very little prior research has measured self-esteem across clinical trials for TS/CTD, this aim was exploratory in nature and we did not form a priori hypotheses.

2. Material and methods

For a full description of the RCT methods, including participants, inclusion and exclusion criteria, procedures, and descriptions of treatments, see Wilhelm et al. [25].

2.1. Participants

Participants ($N = 122$) were recruited from three academic clinical research sites across the United States and were randomly assigned to a treatment condition ($n = 59$ PST, $n = 63$ CBIT). Independent evaluators blind to treatment assignment completed assessments.

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