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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.

Keywords

Self-esteem, Tourette syndrome, Chronic tic disorder, Comorbidity, Comprehensive Behavioral Intervention for Tics

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], [4], [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) [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], [8], [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], [13], [14], [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], [19], [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.

Inclusion criteria required that participants were at least 16 years old and met diagnostic criteria for either TS or CTD based on Clinical Global Impression – Severity scores (moderate or higher) and Yale Global Tic Severity Scale scores (>14 for TS, and >10 for CTD). If participants were on medication for tics, their doses were required to be

stable for at least 6 weeks prior to enrollment, without intentions to change dosage during the study. Participants were also required to have English fluency and an IQ > 80. Participants with a current or lifetime comorbid diagnosis (e.g., ADHD, mood disorder, anxiety disorder, OCD) were eligible if their comorbid condition did not require another treatment at the time of screening. Exclusion criteria ruled out participants with a history of pervasive developmental disorders, schizophrenia, or current substance use disorders. Participants who had received four or more prior sessions of behavior therapy for tics were excluded.

2.2. Measures

2.2.1. Structured clinical interview for ***DSM-IV-TR*** axis I disorders, research version, patient edition (SCID-I/P)

The SCID-I/P [26] is the gold-standard structured clinical interview for psychiatric diagnoses. A trained clinician administered the SCID-I/P at the screening visit to evaluate inclusion and exclusion criteria, as well as presence of current and lifetime comorbid psychiatric illnesses. In the present study, a dichotomous (yes/no) variable was created for presence or absence of one or more current DSM-IV psychiatric diagnoses.

2.2.2. Yale Global Tic Severity Scale (YGTSS)

The YGTSS [27] is the gold-standard tool for clinician-assessed tic severity. The YGTSS assesses motor and vocal tics, in terms of frequency, intensity, number, complexity, and interference, rated on Likert scales from 0 to 5. Scores from these subscales are summed to comprise the total tic severity score. Higher scores indicate greater severity. The YGTSS has strong psychometric properties, including internal consistency, convergent and divergent validity, and inter-rater reliability [27]. The YGTSS was used to establish inclusion criteria. In the present study, we used the total tic severity score as our measure of tic severity.

2.2.3. Clinical Global Impression – Severity (CGI-S)

The CGI-S [28] is a well-validated measure of global symptom severity. The scale is rated on a 7-point Likert scale, with higher scores indicating greater severity. The CGI-S was used to establish inclusion criteria.

2.2.4. Rosenberg Self-Esteem Scale (SES)

The SES [29] is a self-report measure of one's self-esteem. It uses a 10-item Likert scale, with scores ranging from 0 to 30. Higher scores indicate greater self-esteem, and the normative mean (*SD*) in a large U.S.-based adult sample was 22.62 (*5.80*) [30]. The SES has strong test-retest reliability (0.82 to 0.88) and strong internal consistency [31]. In the present study, internal consistency was strong at baseline ($\alpha = 0.91$) and week 10 ($\alpha = 0.90$).

2.2.5. Beck Depression Inventory-II (BDI-II)

The BDI-II [32] is a self-report measure of depression symptom severity. The measure consists of 21 Likert scale items, with total scores ranging from 0 to 63. Higher scores indicate greater depressive severity. The BDI-II demonstrates strong internal consistency ($\alpha = 0.91$) in psychiatric outpatients [32]. It also demonstrates strong convergent validity with other measures of depression, and discriminant validity with measures of anxiety [33]. In the present study, internal consistency was strong at baseline ($\alpha = 0.91$).

2.3. Procedures

For detailed procedures of the RCT, see Wilhelm et al. [25]. Informed consent was obtained from adult participants and from parents of minors; assent of adolescent participants was also obtained. The study was approved by the three sites' institutional review boards. Participants were randomly assigned to treatment conditions, both of which consisted of 8 therapy sessions over 10 weeks. Sessions occurred weekly except for the final two sessions, which took place 2 weeks apart.

CBIT [34] involved psychoeducation about tics and TS, awareness training, competing response training (i.e., HRT), relaxation skills, and applying functional analysis to manage the triggers and consequences of tics. PST involved psychoeducation about tics and TS, as well as supportive therapy for tics and related concerns.

Participants completed self-report and clinician-administered assessments at screening (i.e., SCID-I/P, CGI-S), baseline (i.e., YGTSS, SES, BDI-II), and week 10/end-of-treatment (i.e., YGTSS, SES, BDI-II), to establish inclusion and exclusion criteria and to measure change over treatment. Although the original trial also included 3- and 6-month follow-up assessments, the extent of missing data on the SES precluded examination of these time points in the current study.

2.4. Data analyses

We examined normality of the primary study variables with visual examination of distributions, skewness, and kurtosis values. Variables appeared to be normally distributed and were within recommended cutoffs [35].

To examine whether our sample's baseline scores on the SES differed significantly from published norms (Aim 1), we compared SES scores among study participants with TS/CTD only, study participants with TS/CTD and one or more current comorbid psychiatric diagnoses ("current comorbidity"), and the SES mean *(SD)* from a large (*N* = 503), nationally-representative sample of American adults [30], using 3 independent samples *t*-tests. We calculated Cohen's *d* to determine the effect size of any differences in means.

To examine the strength of the association between self-esteem and tic severity at baseline (Aim 2), we used a multiple regression model with baseline self-esteem as the dependent variable and baseline tic severity, current comorbidity, baseline depression severity, and study site as predictors.

To examine the impact of treatment assignment and current comorbidity on changes in self-esteem from baseline to end of treatment (Aim 3), we conducted a generalized linear model (GLM) with self-esteem change scores (week 10 minus baseline) as the dependent variable, treatment assignment (CBIT vs. PST), current comorbidity, study site, the interaction of treatment assignment and current comorbidity, and the interaction of study site and treatment as predictors, and baseline depression severity as a covariate.

For the regression model and GLM (aims 2 and 3, respectively), we calculated Cohen's f^2 as an estimate of the local effect size of specific variables in the context of the multivariate regression models [36], where f^2[predictor] = (r^2 [full model] – r^2 [model without predictor])/(1 – r^2 [full model]), and each r^2 was calculated as r^2 [model] = (var[null model] – var.[model])/var.[null model]. Cohen's $f^2 \geq 0.02$, $f^2 \geq 0.15$, and $f^2 \geq 0.35$ indicate small, medium, and large effects, respectively [37]. All analyses were conducted using SAS software, Version 9.4 of the SAS system for Windows [38].

3. Results

There were no baseline differences in tic severity or self-esteem between treatment groups (*p*s ≥ 0.18). Additionally, as reported in Wilhelm et al. [25], there were no significant differences in occupation, level of education, race, ethnicity, or marital status between treatment groups. Most participants (*n* = 88) had no comorbid psychiatric diagnoses, whereas 34 participants had at least one current comorbid psychiatric disorder. See Wilhelm et al. [25] for a comprehensive list of the sample's comorbid psychiatric diagnoses. Current comorbid diagnoses were not mutually exclusive, and the most common diagnoses in the sample included OCD (*n* = 19), specific phobia (*n* = 12), and generalized anxiety disorder (*n* = 8).

3.1. Baseline levels of self-esteem in TS/CTD alone, TS/CTD with a comorbid psychiatric diagnosis, and normative means

As hypothesized, an independent samples *t*-test revealed that self-esteem differed significantly between those with TS/CTD only (M = 23.00, SD = 5.21) and those with TS/CTD and at least one current comorbid diagnosis (M = 16.76, SD = 6.41), with a large effect [$t(118) = 5.49$, $p < 0.001$, Cohen's $d = 1.07$]. Also as hypothesized, self-esteem in a nationally representative American adult sample (M = 22.62, SD = 5.80) [30] did not differ significantly from self-esteem in participants with TS/CTD only [$t(588) = 0.57$, $p = 0.57$, Cohen's $d = 0.07$], whereas it did differ significantly from participants with TS/CTD and at least one current comorbid diagnosis, with a large effect [$t(534) = 5.59$, $p < 0.001$, Cohen's $d = 0.96$].

3.2. Baseline association between tic severity, depression severity, and comorbid diagnosis with self-esteem

Overall, the regression model was significant [$F(5, 113) = 28.63$, $p < 0.001$] and explained 54% (adjusted r-squared) of the variance in self-esteem at baseline. Correlations among the predictors were very small between tic severity scores and either current comorbidity ($r = 0.14$) or depression severity ($r = 0.17$) and small between current comorbidity and depression severity ($r = 0.32$), so that multicollinearity among predictors was not an issue (variance inflation factors [VIFs] < 2). The presence of at least one current comorbid psychiatric diagnosis ($b = -3.4 \pm 0.93$, $t = -3.70$, $p < 0.001$, $f^2 = 0.12$) and baseline depression severity ($b = -0.53 \pm 0.06$, $t = -9.40$, $p < 0.001$, $f^2 = 0.77$) were significant predictors of lower levels of self-esteem at baseline. Site effects were minimal ($f^2 < 0.01$). Contrary to hypotheses, baseline tic severity ($b = 0.04 \pm 0.06$, $t = 0.58$, $p = 0.57$, $f^2 < 0.01$) was not significantly independently associated with baseline self-esteem, after accounting for current comorbidity, depression severity, and site effects.

3.3. Impact of treatment assignment on self-esteem, accounting for current comorbid diagnoses and depression severity at baseline

Over the course of 10 weeks of treatment, there was a small increase in self-esteem across both treatments from baseline (M = 21.3, SD = 6.2) to week 10 (M = 22.5, SD = 5.7), $d = 0.19$. A small interaction effect of comorbid diagnosis by treatment type on changes in self-esteem [$F(1,92) = 7.66$, $p < 0.01$, $f^2 = 0.07$] indicated that only participants with comorbid psychiatric diagnoses who were randomized to CBIT reported increases in self-esteem by the end of treatment, whereas those with comorbid diagnoses randomized to PST and participants without comorbid diagnoses (regardless of treatment assignment) did not experience increases in self-esteem. The main effect of treatment assignment on change in self-esteem [$F(1,92) = 5.63$, $p = 0.02$, $f^2 < 0.01$] was very small and driven by the interaction. The main effect of the presence (yes/no) of a comorbid psychiatric diagnosis on change in self-esteem was not significant [$F(1,92) = 0.07$, $p = 0.79$, $f^2 < 0.01$]. Baseline depression severity was not a significant covariate [$F(1,96) = 3.09$, $p = 0.08$, $f^2 = 0.01$] in the model, and site effects were non-significant [site: $F(2,92) = 2.67$, $p = 0.08$, $f^2 = 0.01$; site by treatment interaction: $F(2,92) = 0.03$, $p = 0.98$, $f^2 < 0.01$]. Overall, the model explained about 7% of the variance in observed self-esteem changes.

4. Discussion

Self-esteem is a clinically important variable to consider when treating TS/CTDs, as tics are often visible or audible phenomena that can lead to bullying, teasing, and social ostracism [6]. However, existing research on self-esteem in TS/CTDs consists of a handful of trials in small samples, yielding mixed findings. Importantly, extant studies rarely account for the presence of comorbid psychiatric illnesses or concurrent depression severity, both of which co-occur frequently with tics [4,21] and are consistently related to self-esteem in the broader literature [22,23]. In the present study, we aimed to clarify mixed findings from prior research by

examining self-esteem in a large, well-classified sample of adults with TS/CTD. We also aimed to address a limitation of prior work by including the presence of comorbid psychiatric conditions in analyses. To this end, the present study provides the most comprehensive examination of self-esteem among the largest sample of adults with TS/CTD to date.

By accounting for comorbid psychiatric conditions, the present study provides clarification regarding levels of self-esteem in adults with TS/CTD compared to normative adult levels. Specifically, those with TS/CTD alone did not differ from published national norms for self-esteem in adults, whereas adults with TS/CTD who had at least one current comorbid diagnosis had distinctly lower self-esteem compared to published national norms. This finding is consistent with the existing, smaller studies that accounted for comorbidity [12,14,17,20], lending reliability to our understanding that only people with TS/CTD *and* an additional current psychiatric comorbidity have, on average, lower than normative levels of self-esteem.

Multiple regression results examining correlates of self-esteem provide further evidence that one's comorbid psychiatric symptoms, rather than one's tics, contribute to lower self-esteem in adults with TS/CTD. Both depression severity and presence of a current comorbid psychiatric diagnosis at baseline were significantly, independently associated with self-esteem at baseline, whereas tic severity was not related to self-esteem.

Taken together, these findings have clinical implications for the conceptualization and treatment of TS/CTD. Most notably, it appears that tics alone may not be detrimental to one's self-esteem; clinicians should evaluate on an individual basis the impact of tics on patients' self-evaluation, but clinicians should not assume that tics have a damaging effect on self-esteem. On the other hand, clinicians should take care to evaluate and address issues of self-esteem among adults presenting with TS/CTD with a comorbid psychiatric diagnosis. When treating tic patients with comorbid diagnoses, clinicians should discuss with the patient whether incorporating treatment for comorbid diagnoses may be of benefit. Moreover, clinicians may wish to incorporate additional strategies that target self-esteem (e.g., cognitive skills to identify and evaluate negative beliefs about oneself, core belief work) into behavior therapy for TS/CTD when adults present with comorbid diagnoses. Research is needed to evaluate whether these adaptations to standard treatment for TS/CTD benefit patients' self-esteem.

While behavior therapies such as CBIT are the most empirically-supported psychological treatment for TS/CTD, it is largely unknown how such treatment affects the self-esteem of individuals with TS/CTD. In the present study, results showed that individuals with a comorbid illness experienced small improvements in self-esteem across treatment when receiving CBIT, but no changes in self-esteem were reported among participants with comorbid diagnoses receiving PST. It is possible that boosts in self-efficacy from learning to manage tics in CBIT generalize, such that patients feel greater self-efficacy to manage their comorbid psychiatric illnesses as well. This may in turn enhance one's self-esteem. Another possible explanation for this finding is that patients generalize behavioral strategies learned in CBIT to their comorbid psychiatric illnesses, thereby inadvertently learning to manage comorbid psychiatric symptoms. In turn, these patients may experience benefits to their self-esteem. The most common comorbid diagnosis in the sample was OCD, which has similarities to TS/CTD and which may also respond to aspects of behavior therapy such as using a competing response in place of a compulsion. However, more research is needed to understand the mechanisms through which self-esteem improves across CBIT but not across PST for those with comorbid diagnoses.

Findings should be interpreted with both the present study's strengths and limitations in mind. One limitation to the present study is that certain comorbid diagnoses (e.g., schizophrenia, substance use disorders) were excluded as part of the eligibility criteria for the clinical trial; relatedly, those with other comorbid diagnoses were only included if their comorbid conditions did not require additional treatment [25]. Findings may have differed somewhat in a sample that included the full range and severity of comorbid psychiatric illnesses. For example, the overall rate of comorbid diagnoses may have been higher in our sample if participants with

comorbid conditions that required concurrent treatment had been included. Additionally, mean level of self-esteem among those with TS/CTD with a comorbid illness may have been lower than that reported in the present sample. Despite this limitation, the present study utilized data from a large, rigorous RCT, with clinician-administered diagnostic assessments, independent evaluators, and manualized treatments monitored for adherence. Therefore, data offer reliable, clinically valid, and novel information about self-esteem in adults with TS and CTD.

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Trial registration: clinicaltrials.gov Identifier: NCT00231985.

Declaration of interests

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