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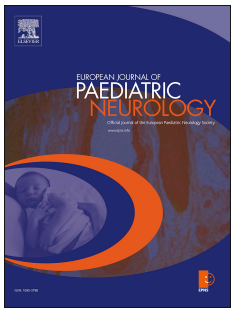

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Self-concept and self-esteem in patients with chronic tic disorders: A systematic literature review

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Abstract

Chronic tic disorders are neurodevelopmental conditions characterized by the presence of motor and/or phonic tics and often accompanied by co-morbid behavioral problems. Chronic tic disorders can negatively affect the level of functioning of young patients across social and family domains, with possible repercussions on their self-perception. We conducted a systematic literature review to assess the clinical correlates of both components of self-perception (self-concept, i.e. what patients think about themselves, and self-esteem, i.e. how they feel about their self-concept) in patients with chronic tic disorders. Reported levels of self-perception varied widely across studies, partly due to the methodological heterogeneity of the reviewed literature. Poor self-concept and self-esteem appeared to be more strongly related to the presence of psychiatric co-morbidities (especially obsessive-compulsive disorder, attention-deficit and hyperactivity disorder, and anxiety disorders) than to tic severity. Poor peer relationship, social difficulties, as well as problems with parents' acceptance were identified as further risk factors for low self-perception. Finally, the reviewed studies highlighted a link between self-perception and quality of life in patients with chronic tic disorders, alongside the protective role of good social adjustment. This information can therefore assist treating clinicians in the choice of tailored therapeutic interventions for this patient population, including behavioral management techniques that can improve self-concept and self-esteem through increased self-efficacy.

Keywords: Tic disorders; Tourette syndrome; tics; self-perception; self-concept; self-esteem.

Introduction

Chronic tic disorders (CTDs) are neurodevelopmental conditions characterized by the presence of motor and/or vocal tics, with onset before the age of 18 years. In patients with Gilles de la Tourette syndrome (GTS) motor and phonic tics coexist, whereas in chronic motor or vocal tic disorders one component is missing. GTS is the most severe chronic tic disorder, affecting up to 1% of school-age children, with a male:female ratio of 3-4:1 [(1,2)]. In the majority of patients (approximately 90%), the clinical picture is complicated by the presence of co-morbid behavioral problems: obsessive-compulsive disorder (OCD), attention-deficit and hyperactivity disorder (ADHD), mood and anxiety disorders, as well as impulsivity, are the most commonly reported psychiatric co-morbidities [(1-3)].

GTS is a potentially disabling disorder with detrimental consequences on young patients' level of functioning in social and family settings. Specifically, children and adolescents with GTS are at high risk for developing poor peer relationships [(4)], especially in the presence of co-morbid disorders [(5)]. These problems are likely to play important roles in the interplay with self-perception of patients with CTDs. According to the psychological literature, self-perception can be divided into two different components: self-concept and self-esteem. The construct of self-concept refers to what persons think about themselves (descriptive account of the self, based on cognitive perceptions), whereas self-esteem reflects how they feel about their self-concept (positive or negative evaluation of the self, based on subjective opinions) [(6)]. Self-concept, in turn, results from the sum of personal identity plus social identity: the integration of the social identity into self-concept begins during childhood through the experience of peer relationships [(7)]. Conversely, self-esteem refers to the person's subjective evaluation of his/her own worth: this concept does not necessarily reflect the objective appreciation of a person's abilities, but encompasses feelings of self-acceptance and self-respect. The developmental trajectory of self-esteem is characterised by progressive increase from adolescence to adulthood and peak around 50-60 years of age, followed by decrease in the old age. High levels of self-esteem are predictive of good outcomes in domains such as academic achievement [(8)], social support, physical and mental health, satisfaction with marriage and job [(9)]. Low self-esteem, on the other hand, implies self-rejection, self-dissatisfaction, and self-contempt, as the individual lacks respect for the self he observes [(10)].

Although knowledge about the correlates of self-perception in patients with tic disorders would be helpful to clinicians to tailor treatment interventions to their patients' needs, relatively little is known on this important topic. We therefore set out to conduct a systematic review of the literature on self-concept and self-esteem in patients with chronic tic disorders in order to better understand the determinants of self-perception in this patient population.

Methods

The methodology applied to our systematic literature review was based on the guidelines described in the PRISMA consensus statement [11]. Two scientific databases (PubMed and PsycInfo) were searched for relevant articles using the following terms: “Tourette”, “tic”, “self-esteem”, “self-concept”, “self-perception”. No restrictions about gender and age of the study samples were applied. We included in our review only original articles published in English language, without time limits. In order to ensure that no relevant studies were missed out, references lists from articles identified were manually screened and the Google Scholar database was searched for grey literature. A summary of the selection process of the reviewed articles is presented in **Figure 1**.

[PLEASE INSERT FIGURE 1 HERE]

Results

Our systematic literature search identified ten articles: all apart from one involved pediatric patients (participants younger than 18 years of age). Five studies were published in the last ten years. Three studies recruited their participants from a population-based setting, whereas the clinical samples in the other studies were recruited from specialist centres (university-based clinics). The reviewed studies were divided into two groups, depending on whether the main topic was self-concept (n=7) or self-esteem (n=3).

With regard to self-concept, three studies did not find significant differences in the perception of self-concept between patients with CTDs and controls [12-14]; three studies found that patients with co-morbid psychiatric disorders (but not patients with pure tic disorders) reported lower self-concept than controls [15-17]; one study [18] found that patients with pure CTDs reported lower self-concept than controls. **Table 1** summarizes the relevant information from the studies focusing on self-concept.

[PLEASE INSERT TABLE 1 HERE]

Edell and Motta [12] investigated the role of parental attitude toward the child and the influence of illness severity on self-concept and psychosocial functioning in young patients with GTS. Self-concept scores in children with GTS were positively correlated to the children's perception of parental accepting behavior and negatively correlated to the children's perception of parental controlling behavior. Parental self-concept, family adjustment, and tic severity did not correlate with the child's self-concept, whereas perceiving their parents as controlling was positively correlated with the children's trait anxiety score.

Edell-Fisher and Motta [13] investigated self-concept in young patients with GTS and their mothers. Children with GTS reported similar scores on self-concept compared to controls, whereas patients' mothers reported lower scores than controls' mothers. Moreover, the patient group scored lower than the control group on measures of behavioral problems. Significant correlations were found between tic severity and children's reports of behavioral and emotional problems, but not self-concept.

Thibert et al. [15] explored self-concept in a sample of adult patients with GTS. The authors found that only patients with GTS and significant obsessive-compulsive symptoms reported significantly lower self-concept and higher social anxiety compared to the general population.

Khalifa et al. [18] explored self-concept in children and adolescents with GTS in a school-based setting. Children with GTS reported lower levels of self-concept than controls. Neither tic severity nor age at tic

onset was associated with children's self-concept. Children with GTS and co-morbid ADHD showed lower self-concept in the domains of physical appearance and social relationships. Lower self-concept scores were associated with higher Full Scale Intelligence Quotient scores.

Gutierrez-Colina et al. [14] explored self-competence, quality of life, and psychosocial functioning in children with GTS. The authors found that children with GTS did not perceive lower levels of self-competence compared with norms of healthy children, although higher levels of behavioral and emotional problems (separation anxiety, depression, OCD and ADHD symptoms) were reported by the patient group. Likewise, patients' parents perceived that their children had more behavioral and emotional problems, and that they had significantly worse quality of life and overall level of functioning in psychosocial, academic, and emotional areas. Children's levels of self-competence inversely correlated with social anxiety, fear of humiliation, and fear of poor performance, whereas social aspects of self-competence inversely correlated with anxiety and affective symptoms. Moreover, social self-competence positively correlated with overall quality of life, as well as its emotional, psychosocial and social domains.

Hanks et al. [16] explored the mediators of self-concept in children and adolescents with CTDs. Only 20% of patients reported low levels of self-concept and patients with co-morbid OCD reported lower self-concept than patients with CTDs only. Patients' overall self-concept was negatively correlated with the severity of co-morbid disorders (OCD, ADHD, depression) and positively correlated with quality of life. Tic severity accounted for 17% of the variance in depressive symptom severity and self-concept appeared to mediate the relationship between tic severity and depressive symptom severity in adolescence but not in childhood.

Silvestri et al. [17] explored self-concept and its determinants in adolescents and young adults with GTS. Levels of self-concept of patients with co-morbid psychiatric disorders were significantly lower than those reported by patients with GTS only, especially in the competence, academic, affect, and social domains. Anxiety symptoms were the main predictors of self-concept (especially trait anxiety, which significantly affected the social and affective dimensions of self-concept). Depressive symptoms also had a negative influence on self-concept (especially on the physical, affective, competence, and social domains), whereas tic severity showed no influence on self-concept.

Only three studies investigated self-esteem in patients with CTDs: two studies [4,5] found that patients with GTS reported similar levels of self-esteem to controls; one study [19] found that patients with CTDs reported lower self-esteem than controls. **Table 2** summarizes the relevant information from the studies focusing on self-concept.

[PLEASE INSERT TABLE 2 HERE]

Stokes et al. [4] examined both self-esteem and peer relationships of children and adolescents with GTS. Patients with GTS reported average levels of self-esteem, their parents rated their social competence as average, and their teachers rated their school performance and adaptive functioning as average. However, there was evidence of difficulties in peer relationships, as young patients with GTS were rated by their classmates as more withdrawn and less popular. Moreover, patients with GTS were more likely to be judged as aggressive and withdrawn than their classmates. Interestingly, neither tic frequency nor tic severity were predictors of these social problems. Patients with co-morbid ADHD were rated as significantly more aggressive than patients without ADHD: these latter were rated as less popular and tended to be more withdrawn than their classmates.

Bawden et al. [5] studied the effects of social problems and family adjustment on self-esteem and peer relationships in children and adolescents with GTS. When compared to children suffering from another chronic condition (diabetes mellitus), young patients with GTS did not report significant differences in any of the scales of Self-Perception Profiles for Children, except for the Athletic Competence scale. No significant differences were found in self-rated social skill between GTS children and the control subjects, however the mothers of the children with GTS reported more difficulties in family problem solving than the mothers of the control subjects. Moreover, the children with GTS were rated as being more aggressive, more withdrawn and less popular than their classmates (measures of peer relationships did not correlate with tic severity or duration). The most relevant risk factors for social problems were identified in the presence of co-morbid ADHD (linked to aggressive behavior) and co-morbid OCD (linked to withdrawal behavior).

Hesapçıoğlu et al. [19] evaluated self-esteem and quality of life in children and adolescents with CTDs. Patients with GTS scored significantly lower than controls on the Rosenberg Self-Esteem Scale, and younger patients (below 12 years of age) scored significantly lower than older patients. Younger age and female gender were identified as the most relevant risk factors for low self-esteem in patients with CTDs. Moreover, lower self-esteem correlated with decreased quality of life in all areas but the academic one. Patients with GTS rated their quality of life as significantly lower than controls in terms of total health and across the physical, school, and psychosocial domains. Likewise, parents' perceptions of their children's quality of life were significantly lower in the patient group compared to the control group across all domains. An inverse correlation was found between quality of life and symptoms of anxiety and depression, whereas there was no correlation between quality of life and tic severity.

Discussion

To the best of our knowledge, this is the first systematic review of the scientific literature on self-perception in patients with CTDs. To date, few studies examined self-perception in patients with CTDs, with considerable variability in methodological aspects (especially assessment strategies) and recruitment settings.

The majority of the reviewed studies (seven out of ten) focused on the assessment of self-concept in patients with CTDs. The results of four studies showed that patients with CTDs reported lower self-concept than controls, whereas the remaining three studies did not find significant differences in self-concept between patients and controls. However, only one study [18] found that patients with pure CTDs reported lower self-concept than controls, whereas three studies [15-17] found that lower self-concept is reported only by the patients with co-morbid psychiatric disorders. Of note, the three studies [12-14] that did not find evidence of lower self-concept in patients with CTDs mentioned the presence of problems in social functioning, rather than poor self-concept itself.

Poorer self-concept in children and adolescents with CTDs appears to be predicted by the presence of co-morbid disorders (especially OCD), whereas it seems to be unrelated to the severity of the tic disorder (or its duration). This finding is in line with the literature on the predictors of quality of life [20-22], as well as global impairment [23], in patients with CTDs. Parents' reactions to the diagnosis and parental style was shown to be a possible contributor to the development of the children's self-concept [12]. This finding corroborates previous observations that parenting style is crucial for the psychological development of children with CTDs: a rejecting and controlling parental style was found to be associated with anxiety and depression in children with GTS, whereas an accepting and autonomy-granting parental attitude was a protective factor against anxiety and depression [24]. Data on self-concept in adult patients with CTDs are limited to one study [15] and do not allow any meaningful conclusions, although the findings of this study are in line with those from the paediatric literature.

Of the three studies investigating self-esteem in children and adolescents with CTDs, two [4,5] found that patients with GTS reported similar levels of self-esteem to controls, whereas one [19] found that younger patients with CTDs reported lower self-esteem than controls. Interestingly, children with CTDs showed problems in peer relationships [5] and were perceived as withdrawn and unpopular by their peers [4], with impaired psychosocial functioning even in the presence of average levels of global self-esteem. Again, the children's level of social functioning did not appear to be related to the severity of their tic disorder, but was affected by the presence of co-morbid disorders such as OCD and ADHD [4,5]. These findings reflect

previous observations on the association between the presence of co-morbid OCD and the tendency towards withdrawal in children with CTDs [25].

Taken together, these findings have clinical implications for the management and support of young patients with CTDs. Importantly, it appears that tics alone may not be detrimental to one's self-concept and self-esteem. Therefore, child neurologists or developmental pediatricians should evaluate the impact of tics on patients' self evaluation on an individual basis, and should not assume that tics have a damaging effect on patients' self-concept or self-esteem. However, child neurologists or developmental pediatricians should take care to assess and address issues of self-concept and/or self-esteem among patients with CTDs and psychiatric co-morbidities in collaboration with child and adolescent psychiatrists and psychologists. Specifically, clinicians may wish to incorporate additional strategies that target self-concept and self-esteem (e.g. core belief work, cognitive skills to recognize and assess negative beliefs about oneself) into standard behavioral treatment strategies for CTDs when patients present with co-morbid psychiatric disorders. Behavioral therapies for tics include the Comprehensive Behavioral Intervention for Tics (CBIT), which integrates Habit Reversal Training with procedures designed to mitigate influences of daily life that worsen tics [26]. Patients who learn to manage their tics using behavioral techniques such as CBIT feel greater self-efficacy, which may in turn enhance their self-concept and self-esteem. Moreover, poorer self-concept in young patients with CTDs appears to be mainly predicted by the presence of co-morbid OCD, which may also respond to behavioral management techniques. Patients who feel greater self-efficacy to manage their co-morbid compulsive behaviors might experience improvements in their self-concept and self-esteem as well.

In summary, reported levels of self-perception varied widely across studies on self-perception in patients with CTDs. Poor self-concept and self-esteem appeared to be more strongly related to the presence of psychiatric co-morbidities (especially OCD, ADHD, and anxiety disorders) than to tic severity. Poor peer relationship, social difficulties, as well as problems with parents' acceptance were identified as further risk factors for low self-perception. Finally, the reviewed studies highlighted a link between self-perception and quality of life in patients with CTDs (possibly mediated by social self-competence), alongside the protective role of good social adjustment.

Our systematic literature search highlighted that the reviewed studies on self-perception in patients with CTDs are low in number (especially in the adult population) and highly heterogeneous in methodology (especially in the recruitment settings and assessment tools) [27]. These problems limit the generalizability of our conclusions and prompt further research into the clinical correlates of self-perception in patients with CTDs, as well as the potential role that treatment interventions aimed at optimizing social adjustment may

have in improving the global outcome of this patient population. Future research should also address possible prognostic implications of self-perception in young patients with CTDs during transition to adult life [28,29].

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Conflict of interest

The authors declare that they have no conflict of interest.

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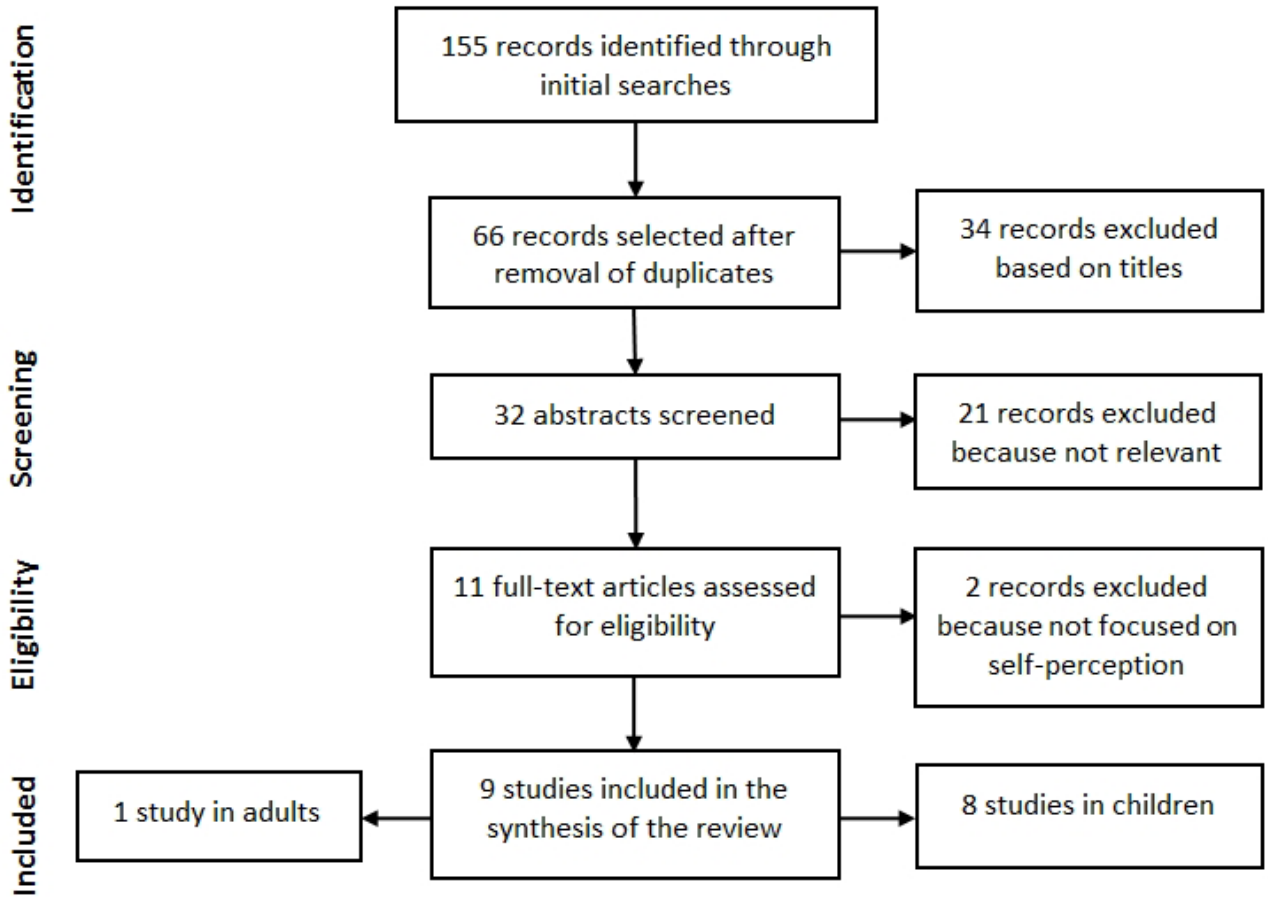
FIGURE

Figure 1. Flow-chart of the selection process of the reviewed articles.



TABLES

Table 1. Studies investigating self-concept in patients with chronic tic disorders.

Study	Year	Country	Setting	n	Mean age (range) / male gender %	Diagnosis (% co-morbidity)	Self-concept assessment tool (type) / subscales	Domain(s) most affected	Domain(s) least affected	Other findings
Edell and Motta	1989	USA	University-based clinic	30	10 (7-15)	GTS	Tennessee Self-Concept scale (parent-rated) + Piers-Harris Children's Self Concept Scale (self-rated) + Child's Report of Parental Behavior Inventory-Revised	-	-	Children's perception of parental behaviour as more willing to accept their diagnosis of GTS was a predictor of better self-concept
Edell-Fisher and Motta	1990	USA	University-based clinic	30	10 (7-15) 9%	GTS (48% OCD)	Tennessee Self-Concept scale (parent-rated) / Behavior, Identity, Physical Self, Self-Criticism, Self-Satisfaction + Piers-Harris Children's Self Concept Scale (self-rated)	Behavior	-	Patients with GTS reported similar self-concept, but more behavioral problems, compared to controls, with behavioral problems and dysphoric mood being related to Illness severity; the patients' mothers reported lower self-concept than the controls' mothers
Thibert et al.	1995	Canada	Population-based sample	98	32 (18-70) / 5	GTS	Tennessee Self-Concept Scale (self-rated) / Behavior, Identity, Physical Self, Self-Criticism, Self-Satisfaction	Behavior, Identity, Physical Self	Self-Criticism, Self-Satisfaction	Patients with GTS and co-morbid OCD (but not patients with GTS alone) reported significantly lower self-concept than the general population

Khalifa et al.	2010	Sweden	Population-based sample	25	10 (7-15) / 88%	GTS (68% ADHD; 16% OCD)	'I think I am' Test (self-rated) / Achievement, Family Relations, Physical Wellbeing, Psychological Wellbeing, Social Relations	Achievement, Family Relations, Social Relations	Physical Wellbeing, Psychological Wellbeing	Patients with GTS reported more negative self-concept than controls (neither tic severity nor age of tic onset correlated with lower self-concept)
Gutierrez-Colina et al.	2015	USA	University-based clinic	39	12 (8-17) / 77%	GTS (21% ADHD; 21% OCD)	Perceived Competence Scale for Children (self-report) / General, Social	-	-	Children with GTS did not report lower self-competence compared with norms of healthy children (self-competence was significantly correlated with social anxiety, fear of humiliation, and fear of poor performance)
Hanks et al.	2015	USA	University-based clinic	97	11 (6-17) / 79%	GTS or CTDs (24% ADHD; 20% OCD; 25% ADHD+OCD)	Piers-Harris Children's Self Concept Scale-Second Edition (self-rated) / Adjustment, Behavior, Freedom from Anxiety, Happiness and Satisfaction, Intellectual and School Status, Physical Appearance and Attributes, Popularity	Freedom from Anxiety, Popularity	Intellectual and School Status, Physical Appearance and Attributes	A minority (20%) of patients with CTDs reported low self-concept (patients with CTDs plus co-morbid disorders reported lower self-concept than patients with CTDs only)
Silvestri et al.	2017	Italy	University-based clinic	22	18 (15-19) / 77%	GTS (36% OCD; 18% ADHD; 9% ADHD+OCD)	Multidimensional Self Concept Scale (self-rated) / Academic, Affect, Competence, Family, Physical, Social domains	-	-	Patients with GTS and co-morbid psychiatric disorders perceived lower self-concept than patients with GTS only (anxiety, but not tic severity, significantly influenced patients' self-concept)

Abbreviations: ADHD, attention-deficit and hyperactivity disorder; CTDs, chronic tic disorders; GTS, Gilles de la Tourette syndrome; OCD, obsessive-compulsive disorder.

Table 2. Studies investigating self-esteem in patients with chronic tic disorders.

Study	Year	Country	Setting	N	Mean age (range) / male gender %	Diagnosis (% co-morbidity)	Self-esteem assessment tool (type) / subscales	Domain(s) most affected	Domain(s) least affected	Other findings
Stokes et al.	1991	Canada	University-based clinic	29	11 (8-15) / 72%	GTS (24% ADHD; 3% OCD)	Piers-Harris Children's Self Concept Scale (self-rated)	-	-	Patients with GTS reported average self-esteem and were significantly more withdrawn, more aggressive and less popular than their classmates (social problems were not predicted by tic frequency or duration)
Bawden et al.	1998	Canada	Population-based sample	26	10 (7-15) / 85%	GTS (12% ADHD; 8% OCD)	Self-Perception Profile for Children (self-rated) / Behavioral Conduct, Global Self-Worth, Physical Appearance, School Competence, Social Acceptance	Athletic Competence	--	Patients with GTS did not report significant self-esteem problems, but showed increased risk for poor peer relationships related to co-morbid ADHD (not to tic severity or family adjustment)
Hesapçıoğlu et al.	2014	Turkey	University-based clinic	57	11 (6-16) / 75%	GTS or CTDs (40% ADHD; 19% OCD)	Rosenberg Self-Esteem Scale (self-rated)	-	-	Patients with GTS reported lower self-esteem than controls, with younger age (<12 years) and female gender (female) as predictors of lower self-esteem

Abbreviations: ADHD, attention-deficit and hyperactivity disorder; CTDs, chronic tic disorders; GTS, Gilles de la Tourette syndrome; OCD, obsessive-compulsive disorder.

Highlights

Little is known about self-perception in patients with chronic tic disorders (CTDs)

We conducted a systematic literature review on self-concept and self-esteem in CTDs

Patients with CTDs report various levels of self-concept and self-esteem

The presence of psychiatric co-morbidities is associated to poorer self-perception