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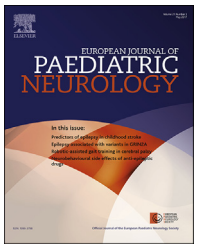

Original article

A preliminary examination of self-concept in older adolescents and young adults with Gilles de la Tourette syndrome

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ABSTRACT

Introduction: Gilles de la Tourette syndrome (GTS) is a childhood-onset neuropsychiatric disorder characterised by multiple tics and often associated with behavioural problems. Although there is evidence of significantly reduced self-esteem in children and adolescents with GTS, little is known about perceived self-concept and its clinical determinants at the transition age between adolescence and adulthood. We therefore set out to investigate self-concept in a clinical sample of young patients with GTS at this crucial age for personal development.

Methods: In addition to standard demographic and clinical data, we collected self-ratings using a standardised battery of psychometric instruments, as well as the Multidimensional Self Concept Scale, a comprehensive questionnaire developed to assess self-concept in subjects aged 9– to 19 years, tapping into the social, competence, affect, academic, family, and physical domains.

Results: We found that patients diagnosed with at least one co-morbid psychiatric disorder ("GTS-plus" phenotype) reported significantly lower self-concept than patients with "pure GTS", whereas tic-related variables had no impact on self-concept. Anxiety symptoms were the main determinants of self-concept, especially trait anxiety with regard to social and affective domains. Affective symptoms could also have a negative impact on the physical, affective, competence, and social domains of self-concept.

Conclusion: Routine screening for anxiety and affective symptoms should be recommended in all patients with GTS seen at transition clinics from paediatric to adult care, in order to implement effective treatment interventions whenever possible.

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1. Introduction

Gilles de la Tourette syndrome (GTS) is a childhood-onset neuropsychiatric disorder characterised by multiple motor tics plus at least one phonic tic lasting longer than 1 year.[1,2] The average age at onset of GTS is around 7 years. The prevalence of GTS in children and adolescents is up to 1%, with a male:female ratio of 3–4:1, and a significant reduction of tic severity after adolescence occurs in about one third of cases.[3,4] Co-morbid psychiatric disorders are reported by the vast majority (90%) of patients with GTS, especially attention-deficit hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD).[2] GTS is also associated with anxiety and affective disorders, rage attacks, impulsivity and self-injurious behaviours.[5–7]

Both tic severity and the presence of psychiatric co-morbidities have been shown to affect patients' health-related quality of life (HR-QoL).[8,9] Tics can impair everyday activities (including job and school attendance) and social relationships.[10] School functioning is often compromised, especially in young patients with co-morbid ADHD.[11] Although co-morbid ADHD is known as the main cause of social and behavioural difficulties in this patient population,[12] affective symptoms have consistently been shown to be a major determinant of patients' wellbeing, as a considerable proportion of patients with GTS receive a formal diagnosis of affective disorder, irrespective of being diagnosed with ADHD, throughout adolescence[13] and adulthood.[14] In particular, there is evidence of significantly reduced self-esteem in children and adolescents with GTS. A study by Khalifa et al.[15] showed that children with GTS have lower self-perception compared to unaffected children. Children with GTS and co-morbid ADHD reported the most significant impairment in the physical appearance and social interactions domains. Moreover, poor self-perception in the physical appearance domain was found to be associated with earlier age at tic onset. Hesapçıoğlu et al.[16] found that female patients younger than 12 years reported lower self-esteem because of their tic disorder. Low self-esteem was found to be associated with impairment in all HR-QoL areas, with the exception of the academic domain. Although the concepts of self-esteem and self-concept have often been used interchangeably, Bracken specifically defined individuals' self-concepts as "learned evaluations of themselves that are based upon their past successes and failures, reinforcement histories, and the ways others react to them and interact with them".[17] Self-esteem and self-concept are components of self-perception which affect the broader concept of HR-QoL. A study by Hanks et al.[18] showed that children and adolescents with a diagnosis of chronic tic disorder have reduced levels of self-concept, and identified a negative correlation between self-concept and the severity of both tics and co-morbid psychiatric disorders. An association was observed between patient's self-concept and overall HR-QoL, and it was noted that younger children are less likely to exhibit depressive symptoms and tend to report higher self-concepts than adolescents.

To date, few data are available about the impact of GTS at the transition between adolescence and adulthood, a crucial age for the development of self-concept. This is a delicate age for patients with tic disorders, as the persistence of symptoms into young adulthood suggests that patients might have to face the possibility that their tic disorder persists throughout life as a potentially stigmatising condition. Self-concept is particularly important amongst older adolescents with tics, who represent a group of young people with GTS at a crucial point in their lives both in terms of adjustment and maturation and also in relation to their being at a watershed for the progression or decline of symptoms. As tics will remit for many children by late adolescence, those who have tics in late adolescence will likely have them for the remainder of their lives. As self-concept is important for long-term functional outcomes (e.g. job, relationships, social factors), the importance of understanding factors that contribute to self-concept during this key developmental period cannot be underestimated. We set out to investigate self-concept and its determinants in a clinical sample of adolescents and young adults with GTS. This was the first study to focus on late adolescence: we hypothesised that patients with GTS in this age group experience problems with self-concept related to co-morbid conditions[19] and assessed their clinical correlates with a comprehensive battery of psychometric instruments.

2. Methods

2.1. *Participants*

Twenty-two patients (5 girls) with a diagnosis of GTS and a mean age of 18 years (age range 15–19 years; standard deviation (SD) 1 year), were recruited from the Child and Adolescent Neuropsychiatry Outpatient Unit of the Sapienza University in Rome between October 2013 and February 2015. The main inclusion criterion was the presence of a validated diagnosis of GTS according to current Diagnostic and Statistical Manual for Mental Disorders (DSM) criteria.[1] Exclusion criteria consisted in the diagnosis of intellectual disability, schizophrenia or other psychotic disorders. Suitable participants were recruited from consecutive attenders to our specialist clinic and included a mix of newly referred patients and follow-up patients. Each participant and his/her parents (for patients younger than 18 years) provided written informed consent before enrollment.

All participants were of white Caucasian ethnic origin with Italian as first language. All but 5 participants (of which 3 with vocational training diploma) were still in education and living with their parents. No patient had educational support needs or physical disorders. Co-morbid diagnoses were established through clinical interview incorporating previous diagnostic records. Eight out of the 22 participants had no co-morbid psychiatric disorder ("pure" GTS), 8 were diagnosed with OCD, 4 with ADHD, and 2 with both OCD and ADHD. The mean age at tic onset was 8 years (SD 3 years). Five participants had at least one first-degree relative with a tic disorder. Eighteen participants were on pharmacotherapy (8 with atypical antipsychotics, 1 with typical antipsychotics, 2 with antidepressants, 7 with other medications). The duration of pharmacotherapy ranged from 1 month to 8 years and there were no previous psychological and pharmacological