COMORBID TOURETTE'S SYNDROME AND ANXIETY DISORDER. CASE REPORT

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ABSTRACT

Gilles de la Tourette syndrome is a childhood-onset neurodevelopmental disorder characterized by motor and phonic tics that debut usually before the age of 10 and exhibit a waxing and waning course. This case report details the diagnosis and treatment of an adolescent patient with Tourette's syndrome and phobic anxiety disorder.

Keywords: Tourette Syndrome, anxiety, phobia.

INTRODUCTION:

Prevalence estimates a general occurrence of around 1% [1,2]. The etiology is complex and multifactorial. The polygenic aspect of the disorder involves common risk variant genes combined with rare mutations [3]. Structural and functional abnormalities of both motor and non-motor basal ganglia-thalamo-cortical circuitries contribute to the heterogeneity of the disorder [4]. Other factors such as infections, prenatal and perinatal difficulties are also involved [5]. Patients diagnosed with Tourette syndrome (TS) have additional comorbid neuropsychiatric disorders, with an indicated comorbidity rate of 88% in a large global study. The most common comorbidities are ADHD and obsessive-compulsive disorder, but increased incidence of mood disorders, anxiety, insomnia, hostility, coprolalia, self-injurious behavior, and personality disorders has also been observed [6]. Although there are various therapeutical options for TS, the complicated physiopathology of the disease, the waxing and waning aspect as well as the comorbid conditions, require a complex management of these cases.

CASE REPORT

A 10-year-old female presented in 2015 with motor and vocal tics that debuted gradually, with increasing intensity over the previous years. Motor tics included eye blinking, nose twitching, sticking out the tongue, touching the shoulder with the chin, jerking of a single shoulder, quickly extending the arms, skipping, and stomping the foot. The first motor tic observed appeared around the age of 2, with a sudden onset and was represented by pulling on her dress. The worst tics reported by the parents were observed when the patient was 8 years old. Phonic tics began after the motor tics were already present.

These tics included throat clearing, animal noises, specific words or phrases and humming. The first phonic tic appeared around the ages of 8 and had a gradual onset, with a peak at the age of 9 and was represented by cat noises. The tics appear in anxiety-generating situations or before bed. On presentation, the tics were assessed using the Yale Global Tic Severity Scale (YGTSS) with a Total Tic Severity Score of 28 and a Total Yale Global Tic Severity Scale Score of 68, due to the functional impairment. The patient also

exhibited symptoms of general anxiety with performance anxiety and marked preoccupation for her health. Numerous phobias were reported such as an intense fear of large-sized animals and fear of darkness. Both YSR and CBCL questionnaires were applied on presentation with anxious/depressed scores at the 81st percentile for both YSR and CBCL. Internalizing T-scores calculated based on these questionnaires were similar (YSR Internalizing T-score = 49 CBCL Internalizing T-score = 53), while externalizing T-scores were slightly diverging (YSR Externalizing T-score = 27 and CBCL Externalizing T-score = 52). Psychological evaluation indicated average intellectual performance (IQ=107).

Following the clinical evaluation of the signs and symptoms, according to the DSM-5 and ICD-10 criteria, the diagnoses of Gilles de la Tourette syndrome and anxiety disorders (social and phobic anxiety) were considered. After discussion with the parents, we decided to initiate pharmacological treatment with haloperidol due to the significant functioning impairment (subjective and objective). The treatment choice was made based on accessibility and the assessment of possible side effects at that time. Progressive doses up to 15 drops of haloperidol per day were prescribed, as well as dietary supplements of magnesium and vitamin B6 and cognitive-behavioral therapy (CBT) sessions. Upon later reevaluation, the dose of haloperidol was increased to 21 drops/day (up to 0.05 mg/kgc).

In 2016, at an ulterior follow-up, it was reported that the patient experienced the exacerbation of the motor and vocal tics, affecting daily activities. Also new tics were described, including abdominal tics with tensing of the abdomen. The pharmacological treatment was switched to progressively decreasing doses of haloperidol associated with progressively increasing doses of aripiprazole, up to 5 mg/day. In the same year, two other YGTSS scales were performed on two different check-ups. At the beginning of the year, the YGTSS registered a Total Yale Global Tic Severity Scale Score of 19, with no functional impairment reported. The motor tics, however, were virtually always present, of mild intensity and with minimal interference of behavior or speech. Meanwhile, there was a single phonic tic reported that appeared occasionally and of mild intensity. Treatment was continued with 5 mg/day aripiprazole and CBT with a later assessment at the end of the same year, when the YGTSS reported an increased score (Total Yale Global Tic Severity Scale Score = 28), due to the fact that the existent tics associated minimal functional impairment in terms of self-esteem and social acceptance.

The goal of the pharmacological treatment was to reduce tics to a level they would no longer produce significant psychosocial functioning disturbance. Over the next period, there were alternating episodes of improvement of overall duration and intensity of tics (especially during school holidays) with episodes of exacerbated symptoms. We initiated the progressive decrease of the medication in the periods when the tics were diminished and did not determine the impairment of the functioning. In the summer of 2017, there was an attempt at stopping pharmacological treatment, but reappearance of high intensity tics determined its resumption in the same year. Furthermore, after an episode of high intensity anxiety and tics in 2018, treatment with sertraline was initiated but shortly stopped 6 months later because of its limited effect. Treatment with aripiprazole 5mg/day and CBT was continued as it proved to be the most efficient option in managing the symptomatology. CBT has shown improvement of reaction in anxiety-generating situations as the 14-year-old learned adaptative techniques in these types of situations. Another attempt at stopping the pharmacological treatment was made in 2019, but tics reappeared shortly

after stopping, thus aripiprazole 5mg/day was reinitiated.

DISCUSSION

This case involves a child who presented with motor and phonic tics, associated with anxiety disorders. Upon clinical evaluation, diagnoses of Tourette's Syndrome (TS) and comorbid anxiety disorders were made. Administration of antipsychotics and CBT sessions has proven to be the most effective therapeutical option, with decrease of tics intensity, number, and overall duration of the episode, as well as improvement of anxiety symptoms. The association between Tourette's syndrome and anxiety has been documented, with an increased risk on anxiety in patients diagnosed with TS [7-10]. The most associated psychiatric conditions are the obsessive-compulsive disorder (OCD) and ADHD, but mood disorders, anxiety, disruptive behavior disorders, eating and substance disorder were also reported. Most of these comorbidities debut early in life and the presence of some might be mediated by comorbid OCD or ADHD [10]. The management of TS involves psychological intervention in the form of behavioral therapy. Existing literature data indicates that the Comprehensive Behavioral Intervention for Tics (CBIT), including Habit Reversal Therapy (HRT), was an efficient form of intervention in order to reduce tics [11]. However, CBIT is hard to access due to the lack of trained professionals, time and compliance demands or poor response to therapy due to patient's age and motivation [12].

Cognitive behavioral therapy is an effective therapeutical option administered either in combination with medication or alone in the management of TS [13]. Other types of therapies that have proven effectiveness in TS included massed negative practice, supportive psychotherapy, exposure with response prevention, self-monitoring, relaxation therapy, assertiveness training, contingency management, a tension-reduction technique, and biofeedback training [14]. Results of a systematic review of the pharmacological treatment used in Tourette's syndrome supported the use of antipsychotics for this disease. These agents proved to be effective in reducing tics in children and young people with Tourette's syndrome. Furthermore, the balance between benefits and adverse reactions favors agents such as risperidone, clonidine, and aripiprazole [15].

Aripiprazole studies have suggested good efficacy in terms of tic reduction and improvement of behavioral symptoms, as well as good tolerability. Most common side effects of aripiprazole include insomnia, fatigue, drowsiness, nausea with fewer side effects reported on weight-gain, QT interval alterations, hyperprolactinemia, extrapyramidal symptoms and tardive dyskinesia [16, 17]. Some of the most effective medications in treating tics are neuroleptics such as haloperidol and pimozide which act through type 2 dopamine receptors blockade. However, they also dysregulate cholinergic, serotonergic, histaminergic and alpha-adrenergic transmission, thus leading to side effects such as weight gain, drowsiness or impaired cognitive performance, as well as hyperprolactinemia and extrapyramidal symptoms (18). Pharmacological treatment for children with chronic tics or Tourette syndrome should be initiated only if psychological interventions are not sufficient and if the symptoms clearly affect the life of the child. Other than the therapeutical options previously mentioned, invasive techniques such as botulinum toxin injections and deep brain stimulation were used in the management of this disorder with satisfactory outcomes in both cases [18-22].

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