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Case Report

A case of severe oral self-injurious Tourette's syndrome alleviated by pregabalin

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Abstract

Self-injurious behavior (SIB) associated with Tourette's syndrome (TS) is a severe neuropsychiatric condition that causes significant distress and can impair social functioning. The current treatment options for the condition include pharmacological, physical and psychosocial interventions. However, given the need for more effective interventions, especially for those patients who are unresponsive and/or intolerant to standard medications, further exploration of novel treatments is imperative. In this report, we present a case of SIB-TS that was successfully treated with pregabalin. The patient received 1-year of follow-up and was noted to have considerable improvement in symptoms. Although rigorous controlled studies are required, based on our case study, pregabalin may be a potential treatment option in some cases of SIB with TS.

Keywords: Tourette's syndrome; Self-injury; Pregabalin

1. Background

In Tourette's syndrome (TS), motor tics generally present as a harmless flinching of the muscles of the face, neck, arms or legs, and can be simple or complex in nature. However, motor tics can result in self-injurious behaviors (SIB), which can cause significant harm to the patient and are observed in as many as 30–48% of TS cases [1]. In addition, SIBs are commonly observed in patients with other psychiatric disorders, including severe depression, autism and obsessive–compulsive disorder (OCD) [2]. However, when SIBs occur in conjunction with tics, they can result in a malignant form of TS, which may often lead to hospitalization, especially in the presence of severe injuries (e.g., injuries of the oral cavity) [3]. With this malignant form of TS, antidepressants, benzodiazepines, antiepileptics or antipsychotics may be used as effective treatment options. However, the treatment for SIB when associated with TS is often complicated by the frequent presence of comorbid conditions. The presence of particular comorbidities may make certain treatment options for tics less likely to be effective. For example, patients with comorbid attention deficit hyperactivity disorder (ADHD) may have difficulties remembering to take medications, and more anxious patients (especially those with severe OCD) may experience an exacerbation of their tics due to the increased focus on them

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during habit reversal training. In cases in which the comorbid condition causes more distress and impairment than the tics and SIB, their treatment is of utmost importance. Consequently, the number of treatment options suggested for SIB associated with TS is high and includes psychotropic medications as well as other treatment modalities, including Botox injections and deep brain stimulation [4,5]. Because the use of these agents is associated with significant side effects and is known to be of only modest efficacy [6], the exploration of novel strategies, including pregabalin, is of critical importance. In this case study, we describe the successful use of pregabalin in a case of SIB-TS.

2. Case

Mr. G.C. was a married 54-year-old Caucasian male with a 10th grade education who worked as a data-entry clerk. He had no contributory family, social or premorbid history, and presented to our outpatient Department of Psychiatry in the absence of any apparent recent stressor or life event requesting a medical evaluation. The patient reported a history of fibromyalgia and OCD, both of which had been characterized by a waxing and waning course for 25 years. His OCD was characterized by obsessions of contamination and a fear of death but not by compulsions. He also had the following features, which presented during childhood: simple motor tics (excessive eye blinking, nose twitching, head jerking and shoulder shrugging), complex motor tics (touching his own mouth and continuous piercing of the tongue using pencils and other sharp objects, which resulted in chronic severe oral injuries that were not preceded by premonitory urges or obsessional thoughts and were not related to concomitant stressors or environmental triggers; Fig. 1), simple vocal tics (coughing and grunting), complex vocal tics (palilalia, which is the repetition of one's own words) and spontaneous changes in the cadence, volume and prosody of speech. The symptoms of OCD and TS were diagnosed according to the *Diagnostic and Statistical Manual for Mental Disorders-Fourth Edition* criteria [7] and were quantified using the Yale-Brown Obsessive-Compulsive Scale (Y-BOCS) [8] (the patient scored a 25, suggesting "severe" symptoms), the Yale Global Tic Severity Scale (YGTSS) (the patient's total score was 95; "always, severe, 2–5 verbal and >5 motor tics") [9] and a Brown Assessment of Beliefs Scale (BABS; the patient scored a 2, suggesting "excellent insight, fully rational") [10]. The patient had previously failed multiple trials (lasting >6 months each) of selective serotonin reuptake inhibitors, which included fluoxetine at up to 60 mg/day; tricyclic antidepressants (TCAs), which included clomipramine at up to 300 mg/day; and a first-generation antipsychotic (FGA), which included haloperidol at up to 12 mg/day or a combination of these medications with benzodiazepines and/or cognitive behavioral therapy (CBT). The patient underwent a 3-week washout from fluoxetine 60 mg/day plus haloperidol 8 mg/day and received an ECG, which showed a QTc of 490 ms. In the setting of QTc prolongation and the persistence of significant oral injury, the patient was prescribed pregabalin at 600 mg/day, which allowed for consistent symptom remission for the following 52 weeks. At 52 weeks, the patient's QTc was recorded to be 390 ms (Fig. 2).

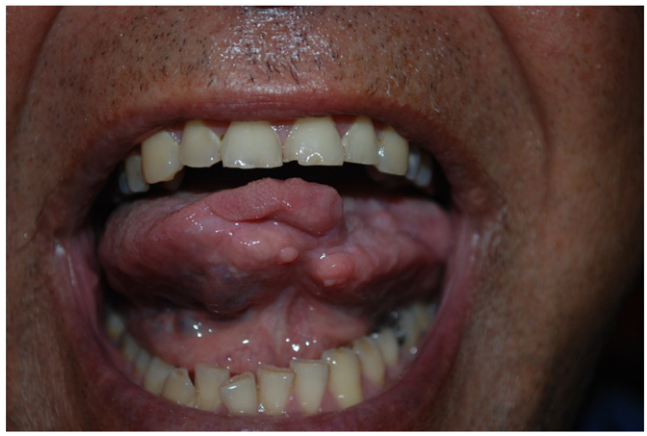

Fig. 1. Loss of substance at the third anterior left edge of the tongue most likely due to chronic piercing in the absence of a history of otherwise relevant lesions of the oral cavity. The cessation of oral traumatic self-injury is documented by the cicatricial outcomes of the tongue.

3. Discussion

Since the recognition and naming of TS by Gilles de la Tourette and Jean-Martin Charcot in the late 19th century, the diagnosis and treatment of TS have followed an unusual trajectory. Indeed, since that time, the syndrome has gone from being considered an idiosyncratic and rare disorder to being recognized as a more common, model neuropsychiatric disorder. In the 1970s, the dramatic results of haloperidol treatment for TS represented a major triumph for the biological view of the syndrome. Since that time, there has been much progress with respect to the medical treatment for TS. Nonetheless, response rates to standard medications for TS, especially for more severe cases complicated by SIB, remain unsatisfactory [11]. Furthermore, SIB-TS has been recognized as a complex phenotype that not only includes tics and SIBs but is also known to co-occur with other disorders, most commonly ADHD as well as other behavioral and anxiety/affective disorders [12]. Indeed, comorbid disorders are often the major focus of treatment in patients with TS complicated by SIB, thus shifting the focus off of finding other novel potential treatments for the SIB associated with TS.

In this case, the patient experienced very severe SIB with oral mutilation due to both TS and/or OCD. The patient was not responsive to standard medications prescribed for either disorder and was transitioned to pregabalin treatment.