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Case Report

Positive clinical effects of gamma knife capsulotomy in a patient with deep brain stimulation-refractory Tourette Syndrome and Obsessive Compulsive Disorder

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ABSTRACT

We report the first case of a patient with severe, intractable Tourette Syndrome with comorbid Obsessive Compulsive disorder, who recovered from both disorders with gamma-knife (GK) stereotactic radiosurgery following deep brain stimulation (DBS). This case highlights the possible role of the internal capsule within the neural circuitries underlying both TS and OCD, and suggests that in cases of treatment-refractory TS and comorbid OCD, bilateral anterior capsulotomy using stereotactic radiosurgery may be a viable treatment option.

1. Introduction

Tourette syndrome (TS) is a complex neuropsychiatric disorder characterized by multiple motor tics and at least one vocal tic. TS is frequently comorbid with obsessive compulsive disorder (OCD), which can complicate treatment, worsen prognosis and further impair quality of life. Despite this, no clinical trial to date has specifically evaluated the treatment of OCD symptoms in patients with TS. Multiple open label trials suggest beneficial effects of neurosurgical interventions in severe, treatment-refractory cases of both OCD and TS [1,2]. We report the case of a patient with severe, intractable TS with comorbid OCD, who recovered from both disorders with gamma-knife (GK) stereotactic radiosurgery following deep brain stimulation (DBS).

2. Case report

The patient was a 47-year-old divorced female from Germany who developed OCD aged 5 years, with obsessions around contamination and a number of compulsive behaviors, such as avoiding physical contact with her parents or "dirty" surfaces in her house. At the age of 10 years, she was diagnosed with TS, with predominant complex-motor tics. In adulthood, self-injurious behaviors emerged alongside a sleep disorder, characterized by impaired initiation and increased motor activity. She first received treatment for TS at the age of 24 years. She received cognitive behavioral therapy, as well as pharmacotherapy including calcium antagonists, tricyclic antidepressants, selective serotonin reuptake inhibitors, and both typical and atypical antipsychotics, with no significant improvement. Trials of the cannabinoid dronabinole and methylphenidate were unsuccessful for TS or OCD symptoms and the patient required multiple admissions to psychiatry units. At the age of 42 years, all medications were discontinued due to a history of adverse effects, including metabolic syndrome, deranged liver function tests and sedation.

The patient underwent implantation of a bilateral DBS device within the posteroventrolateral globus pallidus internus (GPi) for treatment-refractory TS. Prior to surgery, she met DSM-5 criteria for TS and OCD, her Yale Global Tic Severity Scale (YGTSS) score was 39/50 and Yale Brown Obsessive Compulsive scale (Y-BOCS) score was 20/50. Following DBS implant, her TS symptoms remained severe (YGTSS = 39/50) and her OCD symptoms worsened (Y-BOCS = 28/

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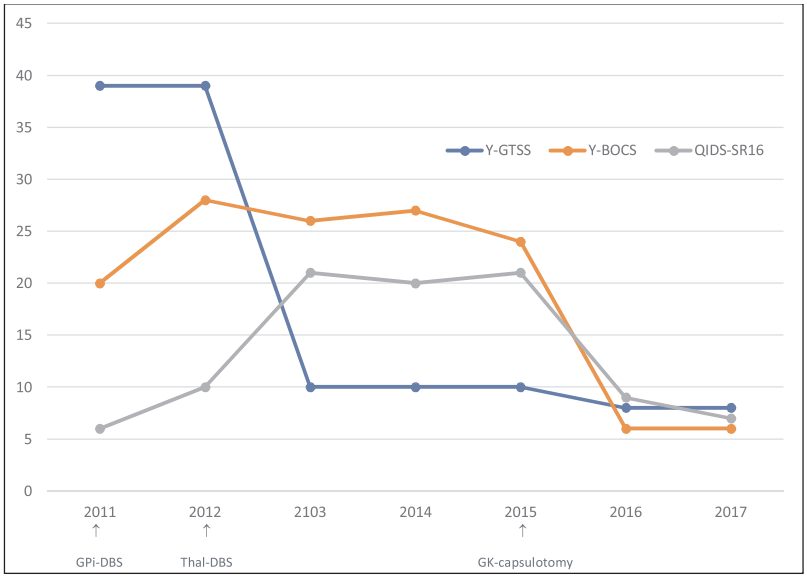

Fig. 1. Changes in Tourette, OCD and depressive scores assessed respectively with Yale-Global Tic Severity Scale (Y-GTSS), Yale-Brown Obsessive Compulsive Scale (Y-BOCS) and Quick Inventory Depression Scale – Self report (QIDS-SR 16).

50). Therefore a year later, a second bilateral DBS device was inserted into the ventral anterior and ventrolateral motor regions of the thalamus. DBS significantly improved both motor and vocal tics (YGTSS = 10/50), which re-emerged immediately upon pausing DBS. Furthermore, self-injurious behavior also improved, however there was no sustained improvement in her OCD symptoms (Y-BOCS = 28/50) (Fig. 1).

At that time, the patient presented severe OCD (Y-BOCS = 24/50) and major depressive disorder (Quick Inventory Depression Scale - Self Report 16(QIDS-SR16) = 21/27); her functioning status was severely impaired, requiring continuous assistance with oral intake and toileting (Global Assessment Functioning (GAF) = 22/100). Depression arose secondary to the failure of the second neurosurgical treatment. She frequently destroyed objects she touched due to unusual destructive obsessions and dressed in a blanket and walked barefoot. Despite obsessional fears around food contamination, she was of normal weight and insight was preserved. Due to the severity of her treatment-refractory OCD symptoms, she was offered bilateral anterior capsulotomy using GK stereotactic radiosurgery.

Defined targets for radiosurgery were the ventral portions of the anterior limb of the internal capsule. Treatment planning was performed with Leksell Gamma Plan software (Elekta, Stockholm, Sweden). Two 4-mm isocenters were used bilaterally separated by 5.5 mm and a radiation dose of 130 Gy was delivered over 306 min (Fig. 2). The patient was discharged the day after the procedure and neuroimaging at five months confirmed ablation of the planned targets (Fig. 3). At nine months her OCD symptoms had partially improved and by one year she was in clinical remission (Y-BOCS = 6/50; QIDS-SR16 = 9/27; GAF = 87/100). Depressive symptoms improved secondary to OCD remission. In addition, TS remained in remission without DBS. No side effects were identified, apart from significant (35 kg) weight gain which was not related to increased appetite.

3. Discussion

To our knowledge, this is the first published case of intractable comorbid TS and OCD fully responsive to bilateral gamma-knife anterior capsulotomy. Due to potential neurological complications, such as hemiplegia and dystonia, lesioning procedures for the treatment of TS symptoms have been largely replaced by DBS since the turn of the millennium. At present, guidelines for functional neurosurgery in TS recommend DBS in treatment-resistant adults who are severely affected by their tic symptoms [1]. Two basal nuclei have emerged as preferred targets for DBS in this patient population: GPi and thalamus. Ablative procedures are not recommended, however thalamotomy, limbic leucotomy, and thalamic lesioning have been associated with positive results in small case-series [1].

In severe OCD, neurosurgical intervention may represent a potentially effective therapeutic options for carefully selected patients, with strong evidence for ablative neurosurgery and moderate evidence for DBS [2]. Anterior cingulotomy, capsulotomy, subcaudate tractotomy, and limbic leucotomy have all been reported to be efficacious [2,3]. The anterior limb of the internal capsule, nucleus accumbens or thalamus/subthalamic nucleus are often used as elective targets for DBS in patients with OCD [2]. It is important to note that for TS and OCD, ablative neurosurgery and DBS have only been studied in uncontrolled small-sized open label studies, with the exception of one randomized controlled trial which had inconclusive results [3].

Remission of TS symptoms following GK capsulotomy, without need for ongoing DBS, was unexpected. A review of the literature has shown some evidence for the effectiveness of this treatment approach in patients with severe and treatment-refractory TS. Although DBS trials for TS have mainly targeted the basal ganglia [1], limbic structures have been implicated in the pathogenesis of TS and positive results have been shown in a previous small thermo- capsulotomy case-series of patients with TS [4]. This case report provides additional support to the concept of shared brain regions underlying both TS and OCD.

Finally, it is notab9e that the patient experienced significant weight gain following radiosurgery. This complication has been occasionally reported in case series of capsulotomy surgery [5] and a plausible explanation for this finding is that ablation of this brain area resulted in disruption of the cortical-striatal-thalamic-cortical circuitries, leading to impaired cognitive control. However, there is no evidence for impaired cognitive control in our patient and she never complained about an increased appetite after GK capsulotomy. Thus, the mechanism underlying weight gain remains unclear.

4. Conclusion

This case highlights the possible role of the internal capsule within the neural circuitries underlying both TS and OCD, and suggests that in cases of treatment-refractory TS and comorbid OCD, bilateral anterior