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A case of non-affective psychosis followed by extended response to non-stimulation in deep brain stimulation for obsessive-compulsive disorder

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Case report

We present a case of acute onset of psychotic symptoms, without signs of hypomania, following DBS for intractable OCD in a patient with no prior history of psychosis or delusional beliefs. The patient was a male in his early 20s with a history of intractable OCD, receiving DBS of the ventral capsule/ventral striatum (VC/VS) implanted as part of a randomized clinical trial of DBS for OCD (NCT00640133). The study sought to investigate effectiveness of DBS as a treatment for intractable OCD, as preliminary data had shown promising therapeutic effects [1,2].

The patient's OCD symptoms began at age 11, marked by exactness, thoroughness and perfectionism. His obsessions were accompanied by near constant doubt and a "need to know", as well as, concerns about offending others, leading to excessive reassurance seeking. Prior to seeking DBS for OCD, he had received numerous conventional treatments without substantial symptom improvement, including trials of at least three serotonin reuptake inhibitors (SRI), one of which was clomipramine; all trials exceeded 6 months in length and were augmented with benzodiazepines (e.g., clonazepam). Dopamine antagonist augmentation was also tried. Cognitive behavior therapy, including exposure and response prevention, was tried over a period of many years, both in traditional weekly outpatient sessions as well as in a specialty residential treatment setting. At the time of his initial assessment for DBS, his OCD severity rated in the extreme range on the Yale-Brown Obsessive Compulsive Scale (Y-BOCS; baseline score = 33). He had a history of depression but was not in a depressive episode at the time of initial evaluation for DBS.

The surgery to implant the DBS device was unremarkable, and placement was verified by imaging (Fig. 1). Active stimulation was delivered to contact 2 (left) and contact 3 (right), both monopolar; contact points were chosen following post operative titration for most acute affective effect and stimulus optimization. At the patient's nine-month follow-up, his OCD symptoms had improved approximately 33% (Y-BOCS = 22), representing a partial response. Approximately 18 months after implantation, the patient presented with an acute onset of psychotic symptoms shortly after DBS amplitude was increased. At the following visit DBS voltage was reduced, but psychotic symptoms persisted. The patient described ideas of reference, magical thinking unrelated to any previous OCD symptoms (e.g., talking to trees), and other delusional beliefs. He felt that something was not right and engaged in delusional behaviors in attempts to "fix" himself (e.g., hugging trees to get them to take the evil out of him). There was no accompanying grandiosity, euphoria, pleasure-seeking, or change in sleep suggesting hypomania. His functioning worsened in tandem. An antipsychotic was started (risperidone 2mg/day). The patient had no prior history of psychotic symptoms, psychotropic medications had been stable, and no recent substance use. Family history was not available.

Psychotic symptoms improved somewhat two weeks after risperidone was started, as did daily functioning. However, ideas of reference persisted. Risperidone was increased to 4mg daily,

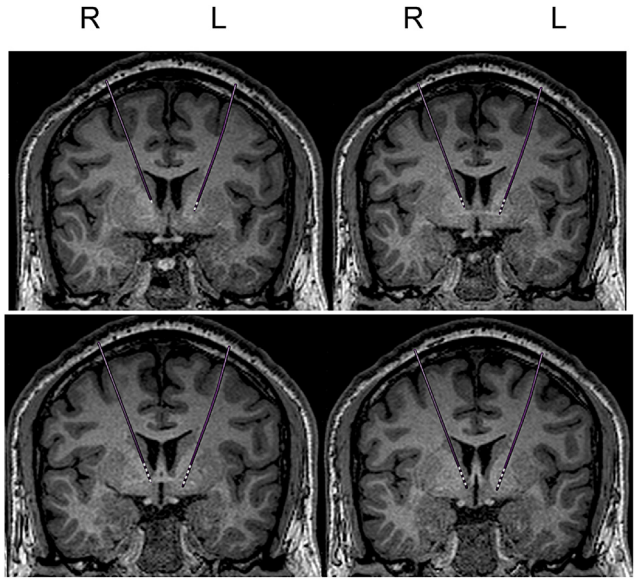

Fig. 1. MRI with deep brain stimulator leads overlaid. The left lead traverses the anterior limb of the internal capsule with the majority of contacts aligned A-P with the anterior commissure. The right lead is more medial but still within capsule, traversing the caudate nucleus before reaching the capsule.

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without further benefit. After discussion with the patient, risperidone dosage was again increased and DBS was stopped on a trial basis. When the patient was re-evaluated two weeks later, delusions were no longer present. It was unclear whether the resolution of the patient's psychotic symptoms was due to additional medication, stopping DBS, or a combination of the two. Due to the unknown cause of onset and resolution of psychotic symptoms, the decision was made to continue risperidone with DBS off. At follow-up two months later, psychotic symptoms had not returned. Approximately 6 months later risperidone was discontinued as psychotic symptoms did not return. OCD symptoms remained improved (Y-BOCS = 18) despite DBS being off and the patient had a significant increase in functioning. He was able to attend a local community college, working his way up to more challenging courses, and gained/remained employed at the same business. He continued to attend therapy during this time.

OCD symptoms returned approximately 2 years later when the patient began to experience a significant life stressor. OCD worsening included increased difficulty in saying or doing anything that might be hurtful to others(Y-BOCS = 24). The DBS was reactivated, with changes in the active contacts, contact 0 (left) and contact 2 (right), based on clinician judgment. To date there has been no return of psychotic symptoms, and the patient remains off antipsychotics. With the reinstatement of DBS, OCD symptoms again improved, reaching a subclinical level (Y-BOCS = 12). The patient continues to live semi-independently, is still employed at the same business, and is engaged in productive life activities.

To our knowledge, this is the first reported case of nonaffective psychotic symptoms following DBS for intractable OCD in the absence of known psychosis vulnerability. Psychosis is a known potential side-effect of DBS of the subthalamic nucleus (STN) for Parkinson's disease (PD). Such cases include transient intraoperative or postoperative psychosis (e.g. Ref. [3,4], psychotic symptoms embedded within a manic or hypomanic episode (e.g. Ref. [5], and symptoms which occur after longer-term stimulation. Due to the high rate of psychosis within the general Parkinson's population, it is often difficult to determine whether DBS is a factor in the onset of such symptoms. Psychosis has also been reported after DBS of the nucleus accumbens (overlapping the VC/VS) for depression [6], and in the context of stimulation-related hypomania during DBS of the ventral striatum [6–8]. As in those reports, the current patient's psychotic symptoms were effectively managed using medication plus DBS cessation. As the decision to turn off the DBS coincided with an antipsychotic dosage increase, it is unknown which intervention was responsible for the resolution of the psychotic symptoms. In addition to improvement in psychosis, the patient's OCD symptoms remained improved despite turning off the DBS device. This is in contrast to the pattern of symptom worsening typically seen when stimulation is interrupted [1,9,10]. One might speculate that the relatively sustained recovery resulted from induced neuroplasticity in cortico-striatal loops through which DBS is thought to act in OCD.

Declaration of competing interest

Abigail A. Testo, Dr. Sarah Garnaat, Dr. Andrew Corse, Dr. Nicole McLaughlin, Dr. Thilo Deckrsbach, Dr. Emad Eskandar and Dr. Benjamin Greenberg have no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

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