


CASE REPORT

Improvement in both severe obsessive–compulsive disorder and refractory tardive dystonia following electroconvulsive therapy: A case report

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Abstract

Background: Obsessive–compulsive disorder (OCD) is often resistant to treatment and may be complicated by tardive dystonia (TDt) with the use of neuroleptics. Furthermore, patients with TDt often have an inadequate response to pharmacotherapy. Although electroconvulsive therapy (ECT) is considered a common treatment option for both TDt and OCD, its efficacy has not been well established for either condition.

Case Presentation: Our case was a 37-year-old Japanese woman who showed improvement in both refractory TDt and severe OCD following ECT. A total of 12 ECT sessions resulted in an improvement in both diseases. To the best of our knowledge, this is the first report of a case in which ECT was effective for both TDt and OCD.

Conclusion: Our report highlights the following two points: when TDt is associated with severe OCD, and the effect of pharmacotherapy is inadequate, ECT may be considered as a treatment option; given the common mechanism of frontal cortex-basal dysfunction reported in both dystonia and OCD, ECT may have an effect on this pathway.

KEYWORDS

electroconvulsive therapy, obsessive–compulsive disorder, tardive dystonia

BACKGROUND

Tardive dystonia (TDt) is persistent dystonia associated with exposure to neuroleptic drugs and is characterized by a variety of involuntary movements and abnormalities of limb position and posture due to muscle construction. It is more likely to occur with the use of first-generation antipsychotics. The prevalence of TDt is reported to be 2.7%, and focal rotatory cervical dystonia is the most common form.¹ Although anticholinergic drugs, benzodiazepines,

dopamine agonists, and botulinum toxins are currently offered treatments, their effects are limited. Electroconvulsive therapy (ECT) is an additional treatment option; however, there is insufficient scientific evidence for its use in TDt patients.

Obsessive–compulsive disorder (OCD) is characterized by obsessions and compulsive behaviors and is treated by selective serotonin reuptake inhibitors (SSRIs), second-generation antipsychotics, and cognitive behavioral therapy (CBT). In general, 40%–60% of OCD patients respond favorably to appropriate pharmacotherapy.² However, even with

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appropriate treatment, 10% of OCD patients are considered treatment-resistant.² In cases of treatment resistance, neuromodulation approaches are alternative treatment options, which include deep brain stimulation (DBS), repetitive transcranial magnetic stimulation (rTMS), and ECT.

Pharmacotherapy for OCD is challenging when response to SSRIs is poor, and antipsychotics cause TDt. Although ECT is considered a common treatment option for both TDt and OCD, its efficacy for these conditions is not well established. We report a case of successful ECT for both refractory TDt and severe OCD.

CASE PRESENTATION

Our patient was a 37-year-old Japanese woman who had a cousin with OCD. When she was in junior high school, there was a period in which she was bullied and did not go to school. At the age of 23 years, she felt that her house was dirty and cleaned compulsively all day. Subsequently, she was diagnosed with OCD by a psychiatrist. Her symptoms improved with 100 mg of fluvoxamine daily. When she was 31 years old, she had agitation that interfered with her daily life. She was started on 15 mg of levomepromazine daily for its calming effect on agitation, which was effective. She developed cervical dystonia 5 years after starting levomepromazine and was diagnosed with TDt by a neurologist. She was offered treatment with a botulinum toxin but refused because she did not like the injection. She was taken off levomepromazine and was treated with 3 mg biperiden, 75 mg promethazine, 3 mg etizolam, 15 mg diazepam, and 1 mg clonazepam daily. However, her TDt did not show any improvement, and her OCD also worsened. Therefore, she was treated with 150 mg fluvoxamine and 50 mg paroxetine daily and CBT. The effect was insufficient, and the antipsychotics were difficult to use because of TDt. She was admitted to our hospital to undergo ECT to treat both her refractory TDt and severe OCD.

On admission, the patient had a Yale–Brown Obsessive–Compulsive Scale (Y-BOCS) score of 25, an Abnormal Involuntary Movement Scale (AIMS) score of 16, and a Hamilton Depression Rating Scale (HAM-D) score of 13 points. Blood examinations, chest x-ray, and brain magnetic resonance imaging were all normal. She received bilateral ECT using the Thymatron System IV (Somatics Inc.). Before ECT, we tapered her benzodiazepines. We used thiopental sodium (4.5 mg/kg) and succinylcholine (1.0 mg/kg). After a total of 12 ECT sessions, her Y-BOCS score improved from 25 to 3, her AIMS score from 16 to 8, and her HAM-D score from 13 to 2 points (Table 1). During ECT, we tapered off fluvoxamine and maintained paroxetine at a dose of 50 mg daily. She continues to visit our hospital and has been able to maintain her improved condition.

DISCUSSION AND CONCLUSION

We report a case of successful ECT for refractory TDt and severe OCD. Although both diseases are challenging to treat with pharmacotherapy, a total of 12 ECT sessions resulted in

TABLE 1 Changes in each evaluation after electroconvulsive therapy

	Before	After
Y-BOCS	25	3
AIMS	16	8
HAM-D	13	2

Abbreviations: Y-BOCS, Yale–Brown Obsessive–Compulsive Scale; AIMS, Abnormal Involuntary Movement Scale; HAM-D, Hamilton Depression Rating Scale.

improvements. To the best of our knowledge, this is the first report of a case in whom ECT was effective for both TDt and OCD.

In our case, when OCD worsened, we increased the dose of SSRIs and combined two drugs (fluvoxamine and paroxetine); however, the patient did not improve. If a patient is refractory to SSRIs, additional antipsychotics may be considered in pharmacotherapy.² However, in our patient, TDt made it difficult to use antipsychotics. When antipsychotic use is difficult because of side-effects, such as TDt and tardive dyskinesia, treatment options are limited. Because ECT may be effective for both OCD and TDt, it may be a promising treatment option for complicated cases.

In our case, ECT alleviated symptoms of TDt, and the patient's AIMS score was halved. Although there are limited case reports and series to date, the efficacy of ECT for TDt has been reported previously.^{3–5} Therefore, when TDt is refractory to pharmacotherapy, ECT may be considered a treatment option. Yasui-Furukori et al. reported that the response rate to ECT for TDt was 30% in 10 cases with depression or schizophrenia; moreover, AIMS scores were reduced by over 25% in all cases.⁵ In our case and others, ECT may be effective for treating TDt; however, it remains unclear whether it should be offered to patients with OCD.

In our case, ECT was also effective for severe OCD. If a patient does not improve with pharmacotherapy and CBT, neuromodulation treatments, such as DBS, rTMS, and ECT, may be options. Although ECT has been shown to be effective for OCD, the response rate is not high, at 60.4%.² Severe OCD symptoms and an absence of depressive symptoms have been reported as factors for a good response,² although complications of TDt have not been studied. Given our case, ECT may be a treatment option for patients who have a complication of TDt with OCD; however, further research is needed.

It is notable that ECT was effective for both TDt and OCD in our case. Although there are no reports on the common ground between OCD and TDt, patients with primary dystonia have been reported to have considerably more obsessive–compulsive symptoms than do normal subjects, with 6.7% of primary dystonia patients having obsessive–compulsive disorder.⁶ Furthermore, it has been suggested that OCD and primary dystonia share a common neurobiological basis associated with frontal cortical-basal dysfunction.⁶ We speculate that OCD may have a vulnerability to dystonia. Taken together, ECT may work by adjusting these common dysfunctions.



As per our experience, when ECT is performed in cases of OCD with TDT, improvement of both may be achieved at once. However, further knowledge is crucial to determine whether ECT should be administered for TDT in patients with OCD.

In conclusion, we report a case of successful ECT for refractory TDT and severe OCD. Our case highlights the following two points: first, when TDT is associated with treatment-resistant OCD, and the effect of pharmacotherapy is inadequate, ECT may be considered as a treatment option; second, given the reported common mechanism of frontal cortex-basal dysfunction in dystonia and OCD, ECT may work by influencing this pathway.

AUTHOR CONTRIBUTIONS

Yasuha Mihara and Koji Otsuki described the case. Yasuha Mihara and Koji Otsuki wrote the draft and carried out the literature search. Mai Hayashi, Satoko Yamashita, Michiharu Nagahama, Maiko Hayashida, Rei Wake, Sadayuki Hashioka, Satoshi Abe, and Masatoshi Inagaki contributed to the conception or design of the work. All authors approved the final version of the manuscript.

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CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

All data generated or analyzed for this case are included in this manuscript.

ETHICS APPROVAL STATEMENT

Not applicable.

PATIENT CONSENT STATEMENT

Written consent to submit this case report for review and publication was obtained from the patient.

CLINICAL TRIAL REGISTRATION

Not applicable.

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