

Can ECT Improve the Motor Symptoms of a Neurological Disease? A Case of Dopa-Responsive Dystonia

Clement Guillet*

La Chartreuse Psychiatric Hospital, Depression Unit, Dijon, France

*Correspondence should be addressed to Clement Guillet; clementguillet@hotmail.com

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Abstract

A 28-year-old woman suffering from a rare genetic disorder, autosomal recessive dopa-responsive dystonia, presented with drug-resistant depression. The motor symptoms of her condition were significantly improved for several months after treatment with electroconvulsive therapy (ECT). This raises the question of whether ECT could be used to treat the motor symptoms of neurological diseases.

Introduction

Electroconvulsive therapy (ECT) is a well-known treatment for mood disorders. There have also been reports of improvement in neurological disorders following ECT, particularly in Parkinson's disease [1], but also in some cases of cervical dystonia [2], orofacial dystonia [3], or blepharospasm [4].

After reporting the case of a patient with a neurological disorder whose motor symptoms were alleviated by a course of treatment with ECT, we now ask ourselves whether the motor symptoms of certain neurological disorders could be improved with ECT sessions.

Case Report

Dopa-responsive dystonia is an extremely rare genetic disease with a European prevalence that varies between 1/1,000,000 and 1/200,000. The autosomal recessive form of dopa-responsive dystonia (DYT5b) is found in only 50 cases in the literature.

The patient has been diagnosed with DYT5b. Her first motor symptoms appeared at the age of 6 years. The symptoms worsened when she was around 15 years old with fixed dystonia of the foot and fatigability of all four limbs. She was wheelchair-bound for 3 years.

The patient's brother, who died by suicide, was also

diagnosed with DYT5b. He presented symptoms of the disorder from birth, while Ms. M was not diagnosed until adulthood because of the atypical presentation of the disease. She was examined by several neurologists over the years, but they were unable to provide the correct diagnosis. She was even hospitalized several times in a psychiatric ward with a diagnosis of conversion disorder.

Then in 2004, when the patient was 23 years old, she was finally diagnosed with dopa-sensitive dystonia with tyrosine hydroxylase deficiency and chronic depression. Treatment with levodopa/benserazide (Modopar®) proved effective, and the patient quickly regained the ability to walk.

In 2007, with the objective of a future pregnancy, she performed a genetic test that identified two mutations in the composite heterozygous state: c.965G>C (Arg31Pro, exon9) and c.1249g>a (Gly414Arg, exon12). This confirmed the diagnosis of dopa-responsive dystonia linked to the tyrosine hydroxylase gene, one of the precursors of dopamine. All members of the paternal lineage are healthy carriers.

In 2016, after her first pregnancy, the patient presented with severe postpartum depression leading to a suicide attempt. Her depression was pharmaco-resistant and none of the antidepressants or mood stabilizers that she was prescribed improved her symptoms. She was therefore referred for ECT sessions.

Neurologically, levodopa/benserazide provided significant but only partial improvement. She presented Parkinson’s syndrome, and though there was no tremor or stiffness, she experienced balance problems and falls, abnormal gait, and brief episodes of dystonia.

The ECT sessions were performed in bilateral temporal stimulation up to the power of 576mC for 25 to 45 seconds (Table 1). From the 13th session onwards, the patient’s psychiatric condition and motor skills improved. It was therefore decided to continue consolidation sessions at a rate of one every 6 weeks, and then every two months. Four months later, the patient was fit to return to work. She presented memory disorders typical of ECT and reversible: the MMS score evaluated before, during and after the treatment remained at 30/30.

Number of stimulations	8	15
Pulse widths (ms)	0,5	0,5
Duration (seconds)	6	8
Current (mA)	800	800
Frequency (Hz)	60	80
Charge (mC)	288mC	576

Table 1: Regulation of the Mecta spECTrum 5000 during the cure.

In dyskinesia and dystonia the patient improved significantly (Table 2). This made it possible for her to delay taking Levodopa/carbidopa at midday, which was not possible before ECT, and to lower the total dose. The patient presented no increase in dyskinesia and dystonia between two ECT sessions: her psychiatric state and motor condition have been stable for one year.

	GDS (Global Dystonia Severity Rating Scale)	UDRS (Unified Dystonia Rating Scale)
Before	5	3,5
After	2	1,5
Comments	Less arms movments, no more jaws movments	

Table 2: Evolution of the dystonia.

The patient’s mood disorder improved progressively over the course of the ECT sessions as expected. But the improvement of the patient’s motor symptoms was unexpected. What neurophysiological explanations could account for both the psychiatric and neurological improvement? And, could ECT treatment be used for other

neurological conditions?

Discussion

Our experience with Ms. M raises the question of the use of ECT in neurological diseases. Neurological and psychiatric disorders often coexist. In Parkinson’s disease, approximately 40% of patients develop depression and 65% suffer from anxiety. Depression is diagnosed in 40% of patients who have had a stroke. In multiple sclerosis, 60% of patients develop depression during their lifetime, with a suicide rate 7.5 times higher than the norm for their age. Patients can develop psychiatric disorders in the aftermath of neurological diseases, just as some neurological diseases are preceded by psychiatric symptoms. This is the case with Parkinson’s disease, multiple sclerosis and Huntington’s disease, which are often preceded by symptoms of depression. Similarly for dystonia, depression is a frequent comorbidity.

ECT is an excellent antidepressant treatment with an efficacy rate of 80% in disorders where conventional antidepressants have an efficacy rate of 60-70% [5].

A meta-analysis published in the *Lancet* and including 1,114 patients [6] showed that ECT is more effective than pharmaceutical treatments, particularly when there are psychotic features like melancholy. The conclusion is simple: ECT is the best available antidepressant treatment.

Patients with comorbid depression of proven biological origin may respond to ECT for both mood and cognitive symptoms. ECT can improve cognitive symptoms in the medium term more than antidepressant drugs, which may be poorly tolerated. ECT is used in particular in depressions secondary to Parkinson’s disease, Creutzfeldt Jakob disease, HIV, neoplastic diseases, and vascular dementia.

The benefit of ECT has been demonstrated in particular in Parkinson’s disease. Whether for associated depressive disorders or for Parkinsonian psychosis, ECT has been shown to be effective.

A 2016 literature review covering 43 articles [7], essentially case reports or case series, analyzed 116 patients with depression and Parkinson’s disease. Depression was improved in 93.1% of cases, and when motor symptoms were severe, 83% of patients were improved. The authors recommended using ECT for patients with Parkinson’s disease and depression, “sooner rather than later”. Directly in case of severe depression (suicidal ideation, psychotic symptoms, catatonia, anorexia ...) and as the 4th line of treatment in case of moderate depression, after treatment with SSRIs, tricyclic antidepressants, then a combination of an antidepressant with lithium or quetiapine. While the improvement of mood disorders by ECT may result

in improvement of motor disorders in cases of co-morbid neurological/affective disorders, isolated motor disorders may also be improved by ECT. Several studies have also shown the positive effect of ECTs in patients with a neurological disease and no psychiatric comorbidity.

One controlled double-blind study [1] conducted in 11 patients with Parkinson's disease, resistant to conventional treatments, without psychiatric comorbidity and with motor fluctuations. Six patients were given sham ECT while the others received actual ECT sessions. There was a statistically significant prolongation of the "on" periods of improvement with levodopa and improvement in motor symptoms compared to the control group.

The question of the use of ECTs in Parkinson's disease has been a subject of debate for many years. A 1989 editorial already recommended a therapeutic trial for each patient with poorly managed or drug-resistant Parkinson's, and particularly those with motor fluctuations. [8]

According to Faber and Trimble [9], in a review of clinical cases found in the literature, 58/75 (77%) patients with Parkinson's disease but no associated psychiatric comorbidity showed signs of motor improvement following ECT.

This raises the question of the neurobiological mechanism and how exactly ECT is able to improve the motor symptoms of certain diseases. The neurobiological theory [10] that explains the major efficacy of ECT on mood disorders hypothesizes that ECT stimulates the transmission of neurotransmitters in the brain, including serotonin and norepinephrine. It also increases the number of receptors for the neurotransmitters that are lacking in depression and that antidepressant drugs attempt to stimulate. But among these neuromediators, dopamine may also be present, which would explain the positive impact of ECT on symptoms in Parkinson's disease, for example. Though this hypothesis is difficult to verify in humans, an animal model found no increase in dopamine metabolites in serum or CSF. The authors' hypothesis was that ECT acts on the sensitivity of post-synaptic dopamine receptors. ECT may therefore increase the sensitivity and the number of dopamine receptors in the substantia nigra.

Because of its anticonvulsant properties, ECT may also be of interest in epilepsy with a reduction in the number of transient seizures in patients with drug-resistant epilepsy. It is particularly indicated for status epilepticus. ECT could increase the efficacy of the endogenous anticonvulsant mechanism through an improvement in the activity of the GABA-ergic system and a reduction in neuronal metabolic activity.

For primary dystonia, the cases found in the literature also report an improvement in neurological symptoms

following ECT sessions. This was the case, for example, of two patients, one with cervical dystonia [2] and another suffering from orofacial dystonia [3], who improved over a period of several days. Patients with blepharospasms also saw improvements in their symptoms, sometimes for several months.

In tardive dyskinesia, the benefit of ECT is controversial. Some authors have found no improvement or even report a worsening of symptoms, while other studies have reported a marked improvement. A 2014 retrospective study [11] found that out of 18 patients suffering from neuroleptic-induced dyskinesia or tardive dystonia, 39% had a good response to ECT with more than 50% improvement on the Abnormal Involuntary Movements Scale (AIMS). In some patients, improvement was noticeable after only 2 sessions. The potential explanation for the improvement in symptoms is the same as for Parkinson's disease: ECT increases dopaminergic transmission. Another explanation involves an increase in blood flow across the blood-brain barrier.

It is therefore clear that ECT should be a treatment of choice in mood disorders occurring in conjunction with neurological diseases, but it can also be considered to treat the motor symptoms of neurological conditions that occur without a psychiatric comorbidity: Parkinson's disease, but also primary dystonias as in the case of our patient, or secondary dystonias. While this treatment option could be included in the therapeutic arsenal, it remains largely unused by neurologists today.

References

1. Andersen K, Balldin J, Gottfries CG, Granérus AK, Modigh K, Svennerholm L, et al. A double-blind evaluation of electroconvulsive therapy in Parkinson's disease with "on-off" phenomena. *Acta Neurologica Scandinavica.* 1987 Sep;76(3):191-9.
2. Garcia RF, Dias AG, De Freitas AR, Fontenelle LF. Short-lived response of cervical dystonia to electroconvulsive therapy. *The Journal of ECT.* 2009 Jun 1;25(2):135-6.
3. Boshes RA, Afonso JA, Tanev K. Treatment of Meige's syndrome with ECT. *The Journal of ECT.* 1999 Jun;15(2):154-7.
4. Sharma A, Hammer S, Egbert M, Sorrell JH. Electroconvulsive therapy and ocular dystonia. *The Journal of ECT.* 2007 Sep 1;23(3):181-2.
5. American Psychiatric Association. Practice guideline for the treatment of patients with major depressive disorder (3rd). <http://psychiatryonline.org/guidelines.aspx>. 2009.

6. The UK. Efficacy and safety of electroconvulsive therapy in depressive disorders: a systematic review and meta-analysis. *The Lancet.* 2003 Mar 8;361(9360):799-808.

7. Borisovskaya A, Bryson WC, Buchholz J, Samii A, Borson S. Electroconvulsive therapy for depression in Parkinson's disease: systematic review of evidence and recommendations. *Neurodegenerative disease management.* 2016 Apr;6(2):161-76.

8. Abrahams R. ECT for Parkinson's disease. *Am J psychiatry* 1989 ; 146 (11) :1391-1393

9. Faber R, Trimble MR. Electroconvulsive therapy in Parkinson's disease and other movement disorders. *Movement disorders: official journal of the Movement Disorder Society.* 1991;6(4):293-303.

10. <https://www.fondationpierredeniker.org/uploads/factSheets/5871f6280fe48cebda94bcbo5394ba4b59f44f2.pdf>

11. Yasui-Furukori N, Kikuchi A, Katagai H, Kaneko S. The effects of electroconvulsive therapy on tardive dystonia or dyskinesia induced by psychotropic medication: a retrospective study. *Neuropsychiatric Disease and Treatment.* 2014; 10: 1209.