CASE REPORT

Psychiatry and Clinical Psychopharmacology 2022;32(1):93-95
DOI:10.5152/pcp.2022.21267

Electroconvulsive Therapy in the Treatment of Catatonia Due To Systemic Lupus Erythematosus

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ABSTRACT

The use of electroconvulsive therapy in case of catatonia due to systemic lupus erythematosus refractory in treatment with immunosuppressant is rare. Therefore, we report a case of catatonia due to systemic lupus erythematosus treated with electroconvulsive therapy as an adjuvant to cyclophosphamide treatment. A 34-year-old female patient diagnosed with systemic lupus erythematosus attended the rheumatology outpatient clinic with a history of catatonia for 8 weeks and laboratory tests revealed high titers of anti-ds DNA positive anti-RNP and polyclonal gammopathy. cyclophosphamide pulse therapy was scheduled in association with electroconvulsive therapy, in 12 sessions, twice a week. The patient evolved with significant clinical improvement after 6 sessions of electroconvulsive therapy and 2 pulses of cyclophosphamide, with negative anti-ds DNA and normalization of gamma globulin levels. Thus, electroconvulsive therapy should be considered as adjuvant therapy in severe cases of neuropsychiatric systemic lupus erythematosus, including catatonia, especially in those with relapses and poor response to immunosuppressant and psychotropic drugs.

ARTICLE HISTORY

Received: October 23, 2021
Accepted: November 26, 2021

KEYWORDS: Behavioral sciences, catatonia, electroconvulsive therapy, psychiatry, systemic lupus erythematosus

INTRODUCTION

Catatonia is a behavioral psychiatric syndrome characterized by the inability to move freely.[1] The presence of 3 or more of the following findings: mutism, dystonia, catalepsy, mannerisms, grimaces, negativism, waxy flexibility, stereotypy, stupor, flexibility, echolalia, or echopraxia were necessary in the diagnosis of catatonia.[2] According to literature data, neuropsychiatric systemic lupus erythematosus (NPSLE) with catatonia is a rare entity whose initial treatment is high-dose benzodiazepine, immunosuppression, and in refractory cases, electroconvulsive therapy (ECT).[3-10]

Therefore, the objective of this study was to report a case of a patient with catatonia due to NPSLE whose treatment with ECT was essential as an adjuvant to immunosuppression.

CASE PRESENTATION

A 34-year-old Afro-Brazilian female patient attended the rheumatology outpatient clinic. She was brought in by her mother with catatonia. No other changes on physical examination. There had been erratic use of medications and loss of appetite for 2 months according to her mother. She was referred to the emergency room and hospitalized for investigation of behavior changes after clinical stabilization.

The patient had been diagnosed with mixed connective tissue disease for 16 years, characterized by Raynaud's phenomenon, puffy hands, arthritis, elevation of muscle enzymes, scleroderma pattern (SD) capillaroscopy, psychosis, and anti-RNP 1:3200. Six years ago, she presented disease activity (catatonia with waxy flexibility), nuclear speckled pattern antinuclear antibody (ANA) (>1/320), and positive anti-dsDNA (enzyme-linked immunosorbent assay: 41.48 U/mL [positive value: ≥20 U/mL]; confirmed by positive anti-ds DNA in *Crithidia* immunofluorescence test), being diagnosed as systemic lupus erythematosus (SLE). The patient had already presented 3 episodes of catatonia associated with poor therapeutic adherence. She was treated with prednisone and 8 cycles of intravenous cyclophosphamide (CYC IV) 1.0 g/m^2 and maintained with azathioprine and prednisone, 15 years ago. In the last 6 years, after a new relapse of the psychiatric condition, she was medicated with 5 cycles of CYC 1.0 g/m^2, and 5 years ago, she showed refractoriness to methylprednisolone pulse therapy and responding partially

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Cite this article as: Gonçalves JJ, Bicalho MET, Guedes LKN, Gallucci-Neto J, Pereira RMR. Electroconvulsive therapy in the treatment of catatonia due to systemic lupus erythematosus. *Psychiatr Clin Psychopharmacol*. 2022;32(1):93-95.

to CYC IV (08 pulses). In this last psychiatric flare, the patient did not respond to benzodiazepine use and was taking azathioprine, hydroxychloroquine, sertraline, and aripiprazole.

During this hospitalization, infections or structural alterations of the central nervous system were ruled out with lumbar puncture (normal cerebrospinal fluid) and cranial tomography, and there were no metabolic alterations and blood count. All tests for common virus, as well as syphilis and blood cultures, were negative. Protein electrophoresis showed polyclonal gammopathy, positive anti-ds DNA (crithidia 1:640), and anti-RNP (1:2300). No complement consumption and anti-Ro, anti-La, antiphospholipid, and anti-P were negative.

Then, it was decided to perform CYC IV (750 mg/m^2), with ECT being scheduled for 12 sessions, twice a week. For the bilateral ECT procedure, the spECTrum 5000Q (Mecta Corporation, Tualatin, Ore, USA) device parameters were set using a pulse width of 1.0 ms, 120 Hz, train duration 6s, and current at 800 mA with a resulting charge of 1152 mC (corresponding 2.5 times the seizure threshold). No parameter adjustment was needed during the ECT treatment, and seizures ranged between 9 seconds and 30 seconds in duration. No post-ECT complications were observed. Previous antipsychotics were retained. After the sixth session of ECT and four cycles of CYC, the patient started to verbalize, sit on the bed, and eat alone. There was normalization of gamma globulin (1.1 ng/dL) and negative anti-dsDNA antibodies. Scores on the Bush-Francis Catatonia Rank Scale decreased from 33 to 0 at the end of ECT treatment.

Finally, it should be noted that the Free and Informed Consent Term was signed by the patient, as well as all the ethical principles of the Declaration of Helsinki, the Brazilian National Health Council (CNS, acronym in Portuguese) and international standards were observed.

DISCUSSION

Neuropsychiatric systemic lupus erythematosus has been demonstrated with highly variable incidences from 20% to 97%.[11] There is a consensus that in patients diagnosed with SLE, neuropsychiatric manifestations are more prevalent (20%-40%) when compared to the general population[12] and rare cases described in other rheumatic diseases.[7-9] As in the literature, our patient evolved with catatonia as the only manifestation of lupus activity[1-4] but also presented anti-dsDNA antibodies confirming the diagnosis of activity of NPSLE.

Interestingly, prospective research has shown that the prevalence of catatonia caused by organic disease in psychiatric units ranged from 20% to 25%;[11] however, when evaluating catatonia as an SLE activity, the literature reports mainly in the pediatric age group[1,7-9] (Figure 1). In addition, the American College of Rheumatology describes 19 syndromes of the central and peripheral nervous system that can occur in SLE[13] does not describe catatonia as one of them, denoting the rarity of this manifestation.

Regarding the treatment of our patient, ECT was introduced due to the seriousness of the psychiatric manifestation, poor response to psychotropic drugs, and several relapses even with the use of immunosuppressants. In fact, currently, ECT is indicated in cases of (i) need for rapid response to the clinical condition, significant functional impairment, starvation (severe depression with a high risk of suicide, with catatonic or psychotic characteristics); and (ii) lack of response to psychotropic drugs.[8,9] Thus, in the context of catatonia, ECT is highlighted as an adjuvant treatment and in refractory cases (Figure 1).

In an attempt to explain the effects of ECT on NPSLE, it is necessary to understand that regarding the pathophysiology of this disease, there is a conjunction of factors: (i) the neurotoxic action of autoantibodies such as anti-ds DNA and anti-NMDA, (ii) microvascular lesions due to thrombotic or embolic events, (iii) damage by the complement system, and (iv) overlap with other diseases such as antiphospholipid antibody syndrome. Thus, the literature delineates that ECT seems to cause immunosuppression of innate and adaptive immunity. Integrative review of preclinical studies published in 2020 states that with chronic ECT sessions, there is stimulation of astrocyte activation and cell proliferation of neurons and glial cells, while pro-inflammatory molecules such as COX-2 and prostaglandin D synthase are reduced.[3-11]

Although the mechanism is not well understood, it is important to emphasize that the use of ECT in severe catatonia (organic etiology or not) is widely supported by literature reviews,[7,10] retrospective studies,[14] and clinical trails.[15] In the present case, ECT treatment was quite effective, as per the previous case reports.[3-9] However, in the case of NPSLE, neither there seems to be uniformity in the number of sessions needed nor the impact of ECT on the need for adjuvant immunosuppression.

MAIN POINTS

- Catatonia is a rare manifestation of neuropsychiatric systemic lupus erythematosus (NPSLE) with 9 reports since 1990;
- Electroconvulsive therapy (ECT) should be considered as adjuvant therapy in severe cases of NPSLE;
- There seems to be no uniformity in the number of ECT sessions for NPSLE cases.

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - J.G.J., R.M.R.P.; Design - L.K.N.G., J.G.J., M.E.T.B.; Supervision - R.M.R.P., J.G.N.; Resource - J.G.J. M.E.T.B.; Materials - J.G.J., M.E.T.B., L.K.N.G., R.M.R.P.; Data Collection and/or Processing - M.E.T.B., J.G.J., J.G.N.; Analysis and/or Interpretation - J.G.J.,

J.G.N., L.K.N.G., M.E.T.B., R.M.R.P.; Literature Search - J.G.J., J.G.N., L.K.N.G., M.E.T.B., R.M.R.P.; Writing - J.G.J., J.G.N., L.K.N.G., M.E.T.B., R.M.R.P.; Critical Reviews - J.G.J., J.G.N., L.K.N.G., M.E.T.B., R.M.R.P.

Acknowledgment: Conselho Nacional de Ciência e Tecnologia (CNPq) and Rheumatology Division, Hospital das Clínicas - FMUSP.

Declaration of Interests: The authors declare no conflict of interest.

Funding: The authors declared that this study has received financial support from Rheumatology Division, Hospital das Clinicas HCFMUSP, Faculdade de Medicina, Universidade de Sao Paulo.

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