

Case Report

Systemic Lupus Erythematosus Presenting as Catatonia and its Response to Electroconvulsive Therapy

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ABSTRACT

Neuropsychiatric systemic lupus erythematosus (SLE) encompasses various psychiatric and neurological manifestations that develop in SLE patients, secondary to involvement of central nervous system. Neuropsychiatric SLE, presenting as catatonia is very uncommon, and treatment of this condition is not well defined. Previously the role of benzodiazepines, immunosuppression, plasma exchange, and electroconvulsive therapy (ECT) has been described in its management. Here we describe a case of neuropsychiatric lupus presenting as catatonia that did not respond to benzodiazepines or immunosuppression. The symptoms of catatonia showed improvement with ECT. Furthermore, we have discussed the pathology of the disorder and the role of ECT in the treatment of cases of catatonia associated with SLE, who do not respond to benzodiazepines.

Key words: Benzodiazepines, catatonia, electroconvulsive therapy, systemic lupus erythematosus

INTRODUCTION

Catatonia is a clinical syndrome characterized by two of the following psycho motor abnormality — motor excitation, negativism, motoric immobility, echolalia, echopraxia and abnormalities of voluntary movements.^[1] Catatonic states occur in the context of a wide variety of both psychiatric and medical conditions.^[2] This syndrome is a significant clinical problem and in certain situations it represents a psychiatric and even vital emergency because it is associated with dehydration, inter current infection, and

pulmonary embolism).^[3] Systemic lupus erythematosus (SLE) is a chronic autoimmune disease characterized by multisystem involvement and can affect the central nervous system resulting in a broad range of psychiatric syndromes such as psychosis, mood disorders, acute confusion and cognitive dysfunction.^[3,4] The prevalence of neuropsychiatric symptoms in patients with SLE is reported to be 81-91%.^[5] There are rare cases of catatonia described in SLE.^[6] These aforementioned cases illustrated the effectiveness of high-dose steroid, followed by plasma exchange 6 times, steroid alone, or five sessions of electroconvulsive therapy (ECT) in SLE patients with catatonia. Case reports of neuropsychiatric SLE treated with ECT alone are very rare. Herein, we describe a case of catatonia due to neuropsychiatric SLE who was successfully treated with ECT.

CASE REPORT

A 30-year-old married Indian female, diagnosed as SLE 10 years ago presented to the Medical Emergency Department with a 1-month history of mutism,

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nonresponse to commands, continuous staring, refusal to eat, rigidity of limbs, decreased self-care, urinating in bed and insomnia. She was on oral prednisolone 30 mg/day and hydrochloroquine 200 mg BD for last 10 years in view of her SLE. Some years ago, she also had received one cycle of 200 mg of cyclophosphamide intravenously for 1-month. There was a past history 2 months ago of psychiatric illness in which she had an episode of agitated behavior, tendency to run away from home, muttering to self, which was precipitated by a single episode of seizure. She was worked up for organic precipitating cause, and haloperidol 5 mg intravenously was given immediately at the time of admission, in view of considering delirium as a provisional diagnosis.

A psychiatric consultation was sought in view of her continuing symptoms. She was conscious, but withdrawn and vacant, with decreased touch from reality. She was mute and did not obey commands showing features of ambitendency. She made no spontaneous movements and could not stand or walk without support. There was rigidity of all limbs more so in upper limbs. The remainder of neurological examination was normal. She was afebrile, and no abnormalities were found in cardiovascular, respiratory and abdominal systems.

All baseline investigations including complete blood count, liver function test, kidney function test, blood sugar, serum electrolytes and electrocardiogram were normal. Imaging studies including ultrasonography, magnetic resonance imaging brain and electroencephalogram showed no abnormality. Anti-nuclear antibodies and anti-ds DNA antibody was negative. Examination of the cerebrospinal fluid, including virological studies, was normal. Renal biopsy showed features of membranous nephropathy consistent with lupus nephritis Class Vb with mild tubulointerstitial nephritis.

Haloperidol was continued for 5 days (5 mg twice daily). On day 6, in view of her catatonic symptoms, she was given lorazepam orally 2 mg/day in three divided doses (6 mg/day). On day 10, the dose of lorazepam was increased to 10 mg/day for the next 10 days. The patient however, showed no improved and the drug was tapered off. She was then given amisulpride 600 mg/day for the next 3 days, but with no response. She was advised ECT by a consultant psychiatrist after the patient showed no response to drugs [Figure 1]. On day 24, ECT was administered bilaterally with 30% energy stimulation with thiopentone (15 mg) and suxamethonium (25 mg) respectively. Her Bush Francis Catatonia Rating scale (BFCRS) score decreased from 27 before ECT treatment to 5 after the fourth ECT. She was given a total of six sessions of modified ECT twice weekly on outpatient department (OPD) basis for a total duration

of 3 weeks by the end of sixth ECT session she was completely symptom free (BFCRS of 0) [Figure 2]. On day 45, she was discharged to outpatient follow-up. In the outpatient follow-up, her mental status was stable. Her husband also reported marked improvement in her behavior at home in the form of taking care of herself and her children, doing household work with interest, remaining cheerful and taking part in family discussions. Other drugs were slowly tapered, and she is presently taking only 50 mg cyclophosphamide. She has presently no neuropsychiatric manifestations at 1-year of follow-up at psychiatric OPD [Figure 3].

DISCUSSION

Systemic lupus erythematosus is a multi-system disorder with protean clinical manifestation. Neuropsychiatric manifestation of SLE can result in damaging presentations.^[4] Catatonia is one of the neuropsychiatric symptom that can occur in SLE.^[6] In a



Figure 1: Patient before electroconvulsive therapy. She is catatonic, withdrawn, and mute, with cushingoid features



Figure 2: Patient immediately after six sessions of electroconvulsive therapy. Improvement in affect and catatonic features, but the cushingoid features persist



Figure 3: One-year follow-up. No neuropsychiatric manifestations and no cushingoid features

review of the literature done by Grover *et al.* on catatonia in SLE, has shown ECT to be an effective mode of treatment.^[7] In the current study, our patient showed marked improvement in neuropsychiatric symptoms with ECT in conjunction with cyclophosphamide which was reflected in her family life and behavior with the physician. This finding of ours is in accordance with Mak *et al.*, who also reported improvement of catatonic symptoms in SLE with cyclophosphamide and ECT.^[5] The patient reported to OPD without recurrence.

There have been several theories explaining the mechanism of action of ECT, which still remains an enigma. One possible mechanism of action of ECT has been described due to enhanced gamma-amino butyric acid (GABA) transmission.^[8] ECT has been shown to decrease glutamate in an animal study.^[9] In another animal study, it was found that ECT produces increased responses to 5-hydroxytryptamine and dopamine receptor stimulation.^[10]

Studies have described the possible neurobiological mechanisms underlying catatonia. Alteration in top-down modulation of basal ganglia due to deficiency of GABA has been described as a possible cause of catatonia.^[11] The role of hyperactivity of glutamate has also been suggested as a possibility.^[12] Furthermore, hypothesis that catatonia could be caused due to a sudden and massive blockade of dopamine neurotransmitters has also been put discussed in studies.^[13] In addition the pathogenesis of neuropsychiatric SLE is multifactorial and involves various inflammatory cytokines, autoantibodies, and immune complexes resulting in vasculopathic, cytotoxic and autoantibody-mediated neuronal injury.^[14]

In view of the possible theories, ECT is arguably a very effective mode of treatment of catatonia. Benzodiazepines also enhance GABA function which explains their role in catatonia. However, there are cases

of catatonia not responding to benzodiazepines that do show response to ECT. This implies the likelihood of multiple pathological mechanisms playing a role in medical catatonia, apart from GABA dysfunction. The effectiveness of ECT in neuropsychiatric SLE can be explained by the varied likely mechanisms of actions of ECT and additionally by its role in neurogenesis.^[15]

There has been recent literature supporting the role of cytokines in depression and mood disorders.^[16-19] ECT is a very effective mode of treatment of major depression. Cytokines have also been implicated in the pathogenesis of neuropsychiatric SLE.^[19] There however, is a scarcity of literature about the effect of ECT on cytokines, but it does seem to alter the expression of various cytokines which could implicate its role in neuropsychiatric SLE.

CONCLUSION

Electroconvulsive therapy may play an important role in the treatment of neuropsychiatric manifestations of SLE including catatonia and should always be considered as a treatment option if resistant to medications. Furthermore, work could be done in elucidating the role ECT could play in modulation of cytokines.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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