Case Report

BIPOLAR AFFECTIVE DISORDER IN A PATIENT WITH NEUROSARCOIDOSIS – A CASE REPORT

Arun Joy[1], Nithin Murali TN[1], Anithakumari Ayirolimeethal[*1], Harish M Tharayil[1]

[1]Department of Psychiatry, Government Medical College, Thrissur, Kerala
[*]Corresponding address: Professor (CAP), Department of Psychiatry, Government Medical College, Kannur, Kerala, PIN – 670503. Email address: akumeethal@rediffmail.com

Abstract

Background: Sarcoidosis is a rare clinical condition due to an autoimmune process that can involve the central nervous system. 20% of cases of neurosarcoidosis (NS) can have psychiatric manifestations. Here, we highlight a case of bipolar affective disorder (BPAD) for 25 years with NS. **Case report:** A 50-year-old woman presented with progressive sensorineural hearing loss from 15 years of age and BPAD for 25 years. She developed generalized tonic clonic seizure and catatonia while on sodium valproate, leading to a detailed medical evaluation. Neuroimaging showed leptomeningeal enhancements, atherosclerosis, and stenosis of the internal carotid artery. Her serum angiotensin-converting enzyme level was elevated, and chest X-ray showed parenchymal lesions. She was diagnosed with neurosarcoidosis. Her condition improved with steroids and mycophenolate mofetil. BPAD was treated with sodium valproate. **Discussion:** Rare medical conditions like NS can present as psychiatric disorders. Poor response and vulnerability to adverse effects of psychotropics warrant a detailed clinical evaluation for organic etiology.

Keywords: Neurosarcoidosis, bipolar disorder, leptomeningeal enhancement

Introduction

Sarcoidosis is a chronic condition that typically leads to non-caseating granuloma in different organs. It is of unknown etiology, has a variable course, and is thought to be due to an auto-immune process involving CD4+ and CD8+ T cells, TNF alpha, and interleukin-2, which disturbs normal tissue functioning.[1] It occurs in 152-215/100,000 population, more commonly in young adults between 20-40 years of age.[2] Neurological complications are found in 5-10% of sarcoidosis.[3] It can involve the central and peripheral nervous systems, producing myriad signs and symptoms. It is diagnosed after excluding many other conditions with similar clinical features.[4] About 10-20% of Neuro-sarcoidosis (NS) may not have other identifiable systemic involvement.[5,6,7] The symptoms are multifocal and involve cranial nerves (seventh being the most common, followed by second and eighth), cause leptomeningitis, parenchymal disease, stroke, or peripheral neuropathy.[1,4]

Depression is the most common psychiatric manifestation of NS, occurring in 60-66% of affected individuals, and around one-fifth of the patients may have psychosis.[1] Previous literature also reported bipolar affective

Access this article online:
https://kjponline.com/index.php/kjp/article/view/420
DOI: 10.30834/KJP.37.1.2024.420
Received on: 29/08/2023. Accepted on: 02/04/2024.
Web publication: 10/04/2024

QR code:

Please cite this article as: Joy A, Murali TNN, Ayirolimeethal A, Tharayil HM. Bipolar affective disorder in a patient with neurosarcoidosis – A case report. Kerala Journal of Psychiatry 2024;37(1):51-54.

disorder in patients with NS.[8] The parenchymal involvement of the disease, stress of illness, treatment complications (e.g., steroids), and other comorbidities may be related to psychiatric manifestation.[1]

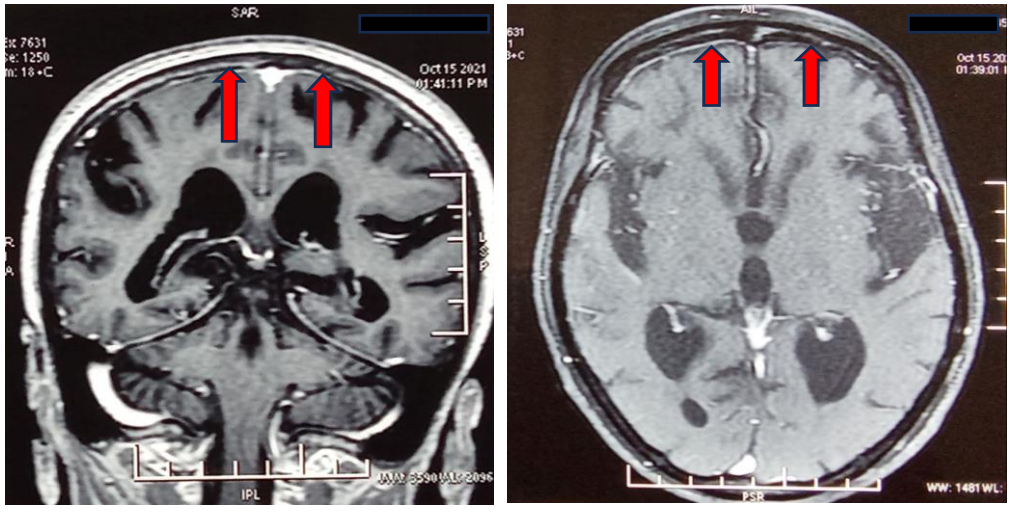
Here, we present a patient with BPAD who was diagnosed with NS. We consider this case unique because it presented with mania and developed seizure episodes while on sodium valproate, which led to the diagnosis of NS.

Case report

A 50-year-old female presented with current symptoms of elated mood, increased energy, increased talk, over-religiosity, suspiciousness, and reduced sleep suggestive of mania. There was a previous history of several similar episodes for the past 25 years. There was no history suggestive of any depressive episodes. She was on irregular medication till five years back, details of which were unavailable. The last episode was in March 2022. She was on Tab. Quetiapine 150mg/day at the time of onset of current symptoms, which were of three months duration. She was diagnosed to have progressive sensorineural hearing loss (SNHL) with onset at the age of 15 years and diabetes mellites for the past seven years. There was no family history of mental illness. Her father and brother had died of chronic pulmonary disease. On examination, her psychomotor activity and talk were increased, her mood was elated, and delusion of persecution and second-person auditory hallucination were present. Her judgment was impaired, and she had no insight. Other cognitive functions were normal.

Two years ago, while on Tab. Lithium 600mg/day and Tab. Haloperidol 1.5mg/day, she developed severe tremors, hypothyroidism, recurrent hyponatremia, and delirium. Tab. Lithium was changed to Tab. Sodium valproate. While on Tab. Sodium valproate 1000 mg/day, she had two episodes of generalized tonic clonic seizures and catatonic stupor. MRI brain showed leptomeningeal enhancement in bilateral frontoparietal regions and periventricular chronic small vessel ischemic changes (Figure 1). MR angiogram showed atherosclerosis in bilateral internal carotid arteries (ICA) and stenosis in the right ICA. Her Serum angiotensin converting enzyme (ACE) level was elevated (293 U/L; normal – 8-52 U/L). The chest X-ray showed bilateral lower zone heterogeneous opacities, suggesting parenchymal infiltrates. Other investigations, including routine blood tests, USG abdomen, CSF study, EEG, and autoimmune assay, were within normal limits. Following this, a diagnosis

Figure 1: MRI Brain images showing leptomeningeal enhancement periventricular chronic small vessel ischemic changes



of neurosarcoidosis was made, and she was started on oral steroids initially and later Tab. Mycophenolate Mofetil 500mg twice a day from the General Medicine department. She was also given Tab. Levetiracetam 500mg twice a day as an antiepileptic. No further seizure episodes occurred.

A diagnosis of organic bipolar affective disorder (F 06.31) was made, levetiracetam was stopped, and sodium valproate was restarted. Her symptoms improved with Tab. Sodium valproate 1000mg/day, and Tab. Amisulpride 200mg/day. Quetiapine was reduced and stopped due to excessive sedation. Tab. Mycophenolate Mofetil was continued at 500mg twice daily. Her biological functions became normal, and personal care was adequate. She was well-maintained on medications.

Informed consent was obtained from the patient to report the case.

Discussion

Neurosarcoidosis is a rare clinical condition in which up to 20% of patients develop psychiatric complications, including depression, psychosis, and bipolar affective disorder.[1,8] Diagnosis is mostly based on clinical features and corroborative evidence. Diagnosis of NS is often difficult due to its varying clinical presentation.[9]

In our case, the diagnosis was made only after the development of seizures. But, she was diagnosed with bipolar affective disorder several years prior to the diagnosis of NS. The patient developed hearing loss at 15 years of age, which was later diagnosed as SNHL. This could have been due to the involvement of the eighth cranial nerve, which is common in NS.[4] She developed mood disorder at 25 years of age, which indicates that there is a temporal correlation with the onset of NS. Here, the diagnosis was made based on the clinical presentation, brain imaging, ACE level, and chest X-ray findings. Having a family history of death due to chronic pulmonary disease in two first-degree relatives could also probably be due to sarcoidosis, which may indicate a genetic basis that needs further exploration.[1]

Though she responded to Lithium, it led to delirium and hypothyroidism and had to be stopped. Marked proneness to adverse effects at higher doses of Lithium has been reported in NS earlier.[8] Leptomeningeal involvement is common in NS, and aseptic meningitis was the reason for the admission and detailed workup in this case. Her MR angiogram revealed atherosclerosis and stenosis in the internal carotid artery, which is also reported in NS.[10] Though treatment with steroids can worsen psychiatric disorders, this patient showed improvement while on steroids, which may again indicate the organic nature of her mood disorder. A similar observation was made in a recent case report on NS with mania.[11]

Conclusion

Neurosarcoidosis, though a rare condition, can present as BPAD. Patients who do not respond or are overly prone to adverse effects of psychotropic medications need to be investigated for the presence of rare multisystem illnesses like NS. The use of steroids in the treatment of NS may pose a challenge in treating associated psychiatric disorders like mood and psychotic disorders. Identifying and treating NS at an early stage can help reduce patient morbidity.

References

1. Bradshaw MJ, Pawate S, Koth LL, Cho TA, Gelfand JM. Neurosarcoidosis: pathophysiology, diagnosis, and treatment. Neurol Neuroimmunol Neuroinflam. 2021;8(6):e1084. Available from: DOI: 10.1212/NXI.0000000000001084.
2. Arkema EV, Grunewald J, Kullberg S, Eklund A, Askling J. Sarcoidosis incidence and prevalence: a nationwide register-based assessment in Sweden. Eur Respir J 2016;48:1690-9.
3. Burns TM. Neurosarcoidosis. Arch Neurol 2003;60:1166-8.
4. Lacomis D. Neurosarcoidosis. Curr

Neuropharmacol 2011;9:429-36.
5. Scott TF, Yandora K, Valeri A, Chieffe C, Schramke C. Aggressive therapy for neurosarcoidosis: long-term follow-up of 48 treated patients. Arch Neurol 2007;64:691-6.
6. Carlson ML, White Jr JR, Espahbodi M, Haynes DS, Driscoll CLW, Aksamit AJ, et al. Cranial base manifestations of neurosarcoidosis: a review of 305 patients. Otol Neurotol 2015;36:156-66.
7. Gelfand JM, Bradshaw MJ, Stern BJ, Clifford DB, Wang Y, Cho TA, et al. Infliximab for the treatment of CNS sarcoidosis: a multi-institutional series. Neurology 2017;89:2092-100.
8. Stern BJ, Royal 3rd W, Gelfand JM, Clifford DB, Tavee J, Pawate S, et al. Definition and consensus diagnostic criteria for neurosarcoidosis: from the Neurosarcoidosis Consortium Consensus Group. JAMA Neurol 2018;75:1546-53.
9. McLoughlin D, McKeon P. Bipolar disorder and cerebral sarcoidosis. Br J Psychiatry 1991;158:410-3.
10. Hoss S, Grinberg T, Eisen A. The interrelationship between sarcoidosis and atherosclerosis—complex yet rational. J Clin Med 2022;11:433. Available from: DOI: 10.3390/jcm11020433.
11. Shoib S, Das S, Saleem SM, Nagendrappa S, Khan KS, Ullah I. Neurosarcoidosis masquerading as mania with psychotic features—A case report. Ind Psychiatry J 2023;32:187-9.