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Manic Episode in a Woman with Sjögren's Syndrome: About a Case Report

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Abstract **Case Report**

Sjögren's syndrome (SS) is an autoimmune disorder that often presents with neuropsychiatric manifestations, either at the onset or during the course of the disease. Psychiatric symptoms such as depression, anxiety, and cognitive dysfunction are common in SS. However, bipolar disorder (BD), characterized by alternating periods of mania and depression, is a rare and under-documented complication of SS. Recent studies suggest a possible connection between autoimmune diseases, including SS, and an increased risk of developing BD, although this relationship remains poorly understood. This case report presents a 62-year-old woman diagnosed with SS who developed a manic episode after starting corticosteroid and immunosuppressant therapy.

Keywords: Manic episode, Sjögren's syndrome, bipolar disorder, corticosteroid therapy, immunosuppressants.

INTRODUCTION

Sjögren's syndrome (SS) is an autoimmune disease characterized by mononuclear cell infiltration and subsequent damage to the salivary and lacrimal glands. This syndrome can present as either secondary Sjögren's syndrome, occurring alongside another autoimmune disease, or primary Sjögren's syndrome when it manifests independently [1]. The prevalence of primary Sjögren's syndrome varies across populations and regions, but it is estimated to affect approximately 0.1% to 3% of the general population [2, 3]. The female-to-male ratio is approximately 9:1 [4].

At the onset of the disease and throughout its progression, it most often presents with neuropsychiatric symptoms [5, 6]. Depression, anxiety, psychiatric features, and cognitive dysfunction are generally the most common [7, 8]. However, the association with bipolar disorder appears to be rare and is not frequently documented [9, 10].

Bipolar disorder is a chronic and debilitating mental illness characterized by recurrent manic, hypomanic, and depressive episodes. Its prevalence has been estimated at between 1% and 2%. The etiology of bipolar disorder is not fully understood, and various factors have been implicated over time, including genetic, biological, and psychosocial factors.

Recently, multiple pieces of evidence have suggested that the immune system, the central nervous system, and the endocrine system are involved in the pathophysiology of bipolar disorder. Indeed, the association between systemic autoimmune diseases and bipolar disorder has been reported in several epidemiological studies. Diseases such as Crohn's disease, autoimmune hepatitis, rheumatoid arthritis, systemic lupus erythematosus (SLE), psoriasis, and autoimmune thyroiditis have been found to increase the relative risk of bipolar disorder [11]. However, the number of studies published on Sjögren's syndrome remains limited.

We present here the case of a patient who is being treated for Sjögren's syndrome and developed a manic episode during the course of her illness.

PATIENT AND OBSERVATION

A 62-year-old woman, married with three daughters, unemployed, with a medical history of a major depressive episode in 2011, treated with paroxetine 20mg/day. Nine months ago, she developed daily fatigue, a 5 kg weight loss, anorexia, and abdominal pain. She consulted several doctors and was prescribed various symptomatic treatments without improvement. She then developed a chronic dry cough and dizziness. A chest X-ray revealed a diffuse bilateral alveolar-interstitial syndrome at the lung bases. A CT scan

showed ground-glass opacities, consolidations at the bases, and bronchiectasis in the trunks. A periungual capillaroscopy was performed. Due to the dryness of the mucous membranes observed during the examination, a facial CT scan was carried out, which revealed chronic sialadenitis classified as Chisholm and Mason grade. The diagnosis of Sjögren's syndrome with pulmonary involvement was made, and the patient was started on prednisone 20mg/day and azathioprine 50mg/day. The dose of prednisone was increased to 40mg/day, and azathioprine to 150mg/day, with slight improvement in her pulmonary symptoms. A brain MRI was performed without abnormalities. Her condition gradually changed, unnoticed by her family, who described her as having a persistent irritability and a high need to talk. However, this irritability worsened, and she became increasingly unstable psychomotorically, highly agitated, unable to stay in one place, excited, and logorrheic, with relentless energy and total insomnia for more than two weeks. She also exhibited excessive generosity, giving her personal belongings—such as television, clothes, and furniture—to strangers. Given this marked change, her older sister took her to a psychiatric consultation. During the psychiatric interview, the patient was unstable, logorrheic with flight of ideas, and exhibited a delusion of grandeur. The diagnosis of bipolar disorder induced by medication was made. The patient was started on carbamazepine 400mg/day, and after one week, a significant clinical improvement was noted.

DISCUSSION

In our case, the patient was diagnosed with medication-induced bipolar disorder according to the DSM-5-TR criteria, as she developed a manic episode shortly after starting corticosteroid and immunosuppressant treatment. The relationship between the psychiatric manifestations and the increase in dosage, as well as the exclusion of organic pathologies of the central nervous system, were essential in making this diagnosis.

In the literature, two cases [5-12] have reported bipolar disorder during the progression of Sjögren's syndrome, but these two patients had a different presentation compared to our case. They showed lesions in the central nervous system (periventricular white matter lesions on weighted MRI images) and clear neurological symptoms, unlike our patient, who mainly had pulmonary involvement. Both patients were treated with high doses of prednisone and azathioprine and developed bipolar disorder after an increase in corticosteroid doses, with significant improvement following carbamazepine treatment.

Psychiatric disorders associated with Sjögren's syndrome (SS) are a clinical reality. Indeed, these disorders can represent a complication of SS due to central nervous system involvement. They may also be an early manifestation of SS, in which case they would precede somatic symptoms. According to Ampelas *et al*., [13], mental disorders in SS could be explained by secondary psychological stress. They suggested that the slow progression and fluctuating course of SS create constant discomfort, leading to a depressive or anxious reaction to a chronic illness. However, in line with other reports, the psychiatric presentation of SS also suggests that mental disorders do not solely arise in response to psychological stress or as a reaction to chronic illness but may constitute an early manifestation of the same autoimmune process, implying a direct immunological activity of SS on the central nervous system (via T lymphocytes, autoantibodies, cytokines, or apoptosis) [14]. It has been suggested that autoantibodies reactive to adrenocorticotropic hormone (ACTH) and alpha-melanocyte-stimulating hormone (α-MSH) may be involved in the pathogenesis of psychiatric symptoms [15].

The etiology of bipolar disorder (BD) in Sjögren's syndrome (SS) is also unclear. Genetic associations and immunological effects, due to autoantibodies against neurons and the ganglionic acetylcholine receptor, as well as the classification of BD itself as an autoimmune disease, are the main suspected factors [16-18].

Some researchers, such as Stevenson *et al*., [19], have confirmed that depression is the most common comorbidity in patients with SS. However, based on the role of autoimmunity in the etiology of bipolar disorder, epidemiological studies have also reported an association between systemic autoimmune diseases and bipolar disorder [19]. Wang *et al*., [20], in a recent study, provided further evidence that autoimmune diseases, including Sjögren's syndrome, are associated with a higher incidence of bipolar disorder.

La relation entre le trouble bipolaire et les maladies immunitaires est bidirectionnelle, des recherches antérieures indiquant une augmentation significative de l'incidence des maladies immunitaires chez les individus atteints de TB [21].

However, the underlying pathophysiological mechanisms remain unclear. Patients with both bipolar disorder (BD) and autoimmune diseases tend to have a shorter average life expectancy, an increased risk of self-inflicted injuries, higher readmission rates, and face more difficult treatment and management. This includes a higher risk of death in the hospital [5-22]. Therefore, rapid diagnosis and management of BD in patients with Sjögren's syndrome are crucial.

In our case, we diagnosed medication-induced bipolar disorder due to corticosteroid therapy in a patient being treated for Sjögren's syndrome, given the absence of current neurological symptoms. However, based on the literature and the significant association between immunological phenomena and bipolar disorder, as well

as the presence of irritability and logorrhea symptoms before corticosteroid therapy, this case warrants particular attention in this patient.

Our study appears to support the hypothesis that an autoimmune process is associated with an increased expression of psychiatric symptoms. Further studies on patients with Sjögren's syndrome may be needed to better understand this association, as treatment and prognosis can vary.

CONCLUSION

Neuropsychiatric manifestations in Sjögren's syndrome are common and can occur not only during the course of the disease but also at its onset. Clinicians (rheumatologists, neurologists, ophthalmologists, and psychiatrists) should be aware that psychiatric disorders are possible in patients with Sjögren's syndrome (both at the onset and during the course of the autoimmune syndrome), and these patients may require psychiatric assistance and appropriate psychotropic treatment. Psychiatric evaluations should be systematically performed in these patients to avoid delays in diagnosis and make better therapeutic decisions.

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