

Sjögren syndrome associated with obsessive-compulsive disorder

J. FREIRE DE CARVALHO¹, F. MACHADO RIBEIRO²

¹Institute for Health Sciences from Federal University of Bahia, Salvador, Bahia, Brazil

²Hospital Universitário Pedro Ernesto from State University of Rio de Janeiro, Rio de Janeiro, Brazil

Abstract. – OBJECTIVE: To describe a patient with Sjögren syndrome (SS) associated with obsessive-compulsive disorder (OCD).

PATIENTS AND METHODS: Case report and systematic review of the literature.

CASE REPORT: A 40-year-old female patient with a history of xerostomia and xerophthalmia initiated in 2015. She also had a history of changing her behavior, and she practices rituals, recurrent obsessions, and compulsions. She was diagnosed with OCD. She was treated with fluoxetine associated with risperidone and then was changed for aripiprazole 10 mg/day. Cognitive-behavioral therapy was also applied. She had good control of the obsessions with these drugs and psychotherapy. Laboratory tests showed positive antinuclear antibodies, anti-Ro/SS-A, and-La/SS-B antibodies. Schirmer test, break up time, and positive green lisamin were all positive. Scintigraphy and ultrasound of salivary glands were positive. A diagnosis of Sjögren syndrome was determined. She was treated with HCQ, vitamin D3 50,000 IU/week, omega-3 2 g, and artificial tears with a good response. Currently, 5 years later, the patient is asymptomatic and has OCD under adequate control even without drugs.

CONCLUSIONS: This case illustrates a rare case of a patient with SS who evolved with OCD.

Key Words:

Sjögren syndrome, Obsessive-compulsive disorder, Psychiatric disease, Neuropsychiatric disease, Autoimmunity.

Introduction

Sjögren syndrome (SS) is a systemic autoimmune disease characterized by an inflammatory process's exocrine glands invasion with consequent reduction of these glands. Autoantibodies' presence plays a role in this disorder; besides the gland involvement, several organs and systems may be impaired, such as the hematological sys-

tem, renal, gastrointestinal, and neurological system¹.

In fact, the nervous system was previously described in this syndrome². The most common manifestations are peripheral neuropathy, aseptic meningitis, and demyelinating lesions. Neuropsychiatric disorders are also reported and vary from 18 to 45%². Very few patients were described to have obsessive-compulsive disorders (OCD). Only one previous case report describes a primary SS patient with this condition³. The other descriptions are case series or observational ones in which obsessive symptoms were reported, but the patients did not fulfill the diagnostic criteria for OCD.

The purpose of this article is to report a patient with SS who developed OCD. A systematic review of the literature on this rare association of SS and OCD was also performed.

Case Report

A 40-year-old female patient with a history of xerostomia and xerophthalmia was initiated in 2015. She also had a history of changing in her behavior, and she practices rituals. The patient suffered from recurrent obsessions regarding persecution and ideas of was suffering diseases, contamination, and compulsions, including hand-washing, showering, cleaning, and checking. Her symptoms had caused significant distress and interference with social functioning. There was no drug use or diagnosed medical illnesses to account for her symptoms. Therefore, her symptomatology satisfied DSM-IV criteria for the diagnosis of OCD. She was treated with fluoxetine 60 mg/day associated with risperidone 0.25 mg/day, but she had galactorrhea, and then this drug was changed for aripiprazole 10 mg/day. Cognitive-behavioral therapy was also applied. She had good control of the obsessions. Laboratory tests showed positive antinuclear antibodies with a titer of 1:640, an-

ti-Ro/SS-A, and-La/SS-B antibodies. Anti-RNP, anti-dsDNA, rheumatoid factor, anti-CCP, ANCA, anti-tissue transglutaminase, anti-gliadin, anti-endomysium, IgG and IgM anticardiolipin, and lupus anticoagulant were all negative. Normal C3 84 mg/dL and C4 29mg/dL. Blood cell count and blood chemistry were normal. Erythrocyte sedimentation rate was 13 mm/1st hour and C-reactive protein < 5 mg/dL. Normal levels of vitamin B12 was 579 (nr: > 300 pg/mL), TSH 1.48 mU/mL, and free T4 0.92 ng/dL. Serology for infectious disease, including HIV 1 and 2, HTLV I and II, hepatitis B and C, syphilis, and IgM for Epstein-Barr, human cytomegalovirus, rubella, and toxoplasmosis were negative. Schirmer test was 6 mm in the right eye and 5 mm in the left eye, with a break-up time of 4 sec in both eyes and positive green lisamin grade 3. Scintigraphy of salivary glands showed marked dysfunction of salivary glands, and salivary gland ultrasound showed alterations compatible with *sicca* syndrome. She denied biopsy of the minor salivary glands. A diagnosis of Sjögren's syndrome was determined. She was treated with HCQ 400 mg/day, vitamin D3 50,000 IU/week, omega-3 2g, and artificial tears with a good response. Currently, 5 years later, the patient is asymptomatic and has OCD under adequate control even without drugs.

Discussion

This article reports a rare association of Sjögren syndrome who had OCD.

Shen *et al*⁴ evaluated psychiatric symptoms in an extensive nationwide population-based retrospective cohort study including 2,686 patients with primary SS and 10,744 matched controls. The authors found a higher risk of depression, anxiety disorder, and sleep disorder in SS compared to controls. Furthermore, importantly, some patients with anxiety also had OCD symptoms⁴.

Two other studies, from the same author, probably evaluating the same population, found neuropsychiatric symptoms, some compatible with OCD in some SS patients, but they do not describe that these cases fulfilled the diagnosis criteria for OCD^{5,6}. Moreover, Karaiskos *et al*⁷, evaluating 103 primary SS found obsessiveness as personality traits in 27.2% of these patients⁷. Again, the author does not describe patients with OCD diagnosis.

More specifically, regarding OCD diagnosis, only two previous studies assessed the relationship between SS and OCD. One of them was a

case report of a 17-year-old female hospitalized due to OCD and major depression. During the hospital stay, she was diagnosed as SS. Interestingly, she received immunosuppressive drugs and evaluated with good control of the psychiatric disorder³. This exciting good response of OCD to immunosuppressive drugs was also observed in our patient since she currently is out of psychiatric drugs and with good control of OCD. In other study from Wang *et al*⁸ evaluating a large nationwide population-based cohort study, the authors evaluated if a history of systemic autoimmune diseases was associated with an increased risk of subsequent onset OCD. A total of 63,165 patients with autoimmune diseases were compared to 315,825 patients in a follow-up of 10 years. The authors found an increased frequency of OCD in patients with systemic autoimmune diseases (HR: 1.85; 95% CI 1.41–2.43), mainly in systemic lupus erythematosus, dermatomyositis, and SS (HR: 2.38; 95% CI: 1.53–3.72)⁸.

Regarding pathophysiological hypothesis on SS and OCD, some researches believe in the role of cytokines present in SS that may cross the hematoencephalic through a leaky brain barrier, active transport, endothelial cell activation, and cytokine receptor binding⁹. Furthermore, SS might exacerbate the OCD syndrome in the presence of subclinical OCD vulnerability *via* a direct mechanism that includes acting on the biological substrate of OCD, or an indirect one using the vulnerability to depression or other anxiety disorder might then exacerbate OCD¹⁰. Moreover, a third hypothesis could be related to a common genetic etiology or epigenetic mechanism observed in other autoimmune diseases linked to OCD, such as rheumatic fever¹¹.

Future studies to report more patients with SS and OCD diagnoses are desired, and studies that evaluate the immunosuppressive drug's role on the OCD symptoms.

Conclusions

The present study adds a rare case of SS associated with OCD to the literature. Immunosuppressive drugs seem to be an option to deal with this psychiatric condition in the SS context.

Conflict of Interest

The Authors declare that they have no conflict of interests.

Ethical statement

The authors declare that he followed the World Medical Association Declaration of Helsinki in this study. An informed consent was obtained from the patient for publication of her case. No image of her is used.

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