Brief communication

Secondary Sjögren's Syndrome in Sero-Positive Rheumatoid Arthritis

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Nauman Ismat Butt[1], Barak Waris[2], Muhammad Sohail Ajmal Ghoauri[3], Ayesha Afzal[4], Muhammad Waseem Riaz[5]
[1,2,4,5]Azra Naheed Medical College, Superior University, Lahore. [3]Quaid e Azam Medical College, Bahawalpur

ABSTRACT

We report a case of secondary Sjogren syndrome in a 46-year-old woman with a 4-month history of bilateral symmetrical joint pains, morning stiffness, dry mouth, dry eyes, fatigue, and lethargy. Examination revealed pallor, dry eyes, dry tongue and synovitis in multiple joints. Investigations showed iron deficiency anemia, positive rheumatoid factor, ANA, anti-CCP antibodies, anti-SSA/Ro and anti-SSB/La antibodies. The patient was diagnosed with Sero-positive rheumatoid arthritis complicated by secondary Sjögren's syndrome. She was started on Methotrexate, Hydroxychloroquine, oral Steroids and dietary supplements. At 8-week follow-up, her joint pains improved, steroids had been discontinued, and she was tolerating medications without adverse effects.

Keywords: Sjögren's Syndrome, Rheumatoid Arthritis, Anti-CCP antibodies, anti-SSA/Ro and anti-SSB/La antibodies, Methotrexate, Hydroxychloroquine.

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Correspondence: Nauman Ismat Butt
Azra Naheed Medical College, Superior University, Lahore
Email: nauman_ib@yahoo.com

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INTRODUCTION

Sjögren's syndrome is classified into two types: primary, which occurs independently without association with other diseases, and secondary, which arises in individuals with an existing autoimmune condition such as rheumatoid arthritis or systemic lupus erythematosus.[1] Primary Sjögren's syndrome mainly results in dry eyes and mouth, caused by inflammation of the salivary and tear glands. In half of the cases, it can also affect other parts of the body such as skin, lungs, digestive tract, nervous system and kidneys, leading to a broader range of symptoms beyond glandular issues.[2] For primary Sjögren's syndrome, diagnosis typically relies on the presence of dry eyes and mouth, along with laboratory findings that show autoantibodies, including anti-SSA/Ro) and anti-SSB/La), and occasionally a lip biopsy that reveals inflammation of the salivary glands.[3] Secondary Sjögren's syndrome is diagnosed when a patient with an autoimmune disease begins to experience severe dryness of the mouth and eyes. Unlike primary Sjögren's syndrome, a lip biopsy is rarely required for diagnosing secondary Sjögren's, as the clinical symptoms and the presence of another autoimmune condition generally provide enough evidence.[4]

This case report presents a 46-year-old female diagnosed with Sero-positive RA complicated by secondary Sjögren's syndrome and iron deficiency anemia, who showed significant improvement with Methotrexate, Hydroxychloroquine, and oral Steroids, along with supportive measures for dry symptoms. This case is significant as it highlights the complex interplay between RA and secondary Sjögren's syndrome, two autoimmune conditions that often coexist but can present with overlapping and subtle clinical features. The patient's Sero-positive RA status with secondary Sjögren's syndrome highlights the importance of considering additional autoimmune etiologies in patients presenting with symptoms like dry eyes and mouth, which are often overlooked or attributed to other causes. The combination of these

Antinuclear antibodies (ANA) by IFA

Test	Result	Titer	Normal Titer
ANA	Speckled	1:640	<1:80

CONCLUSION :

ANA shows Speckled pattern with titres of 1:640. ENA quantrix is recommended for further workup.

INTERPRETATIVE GUIDELINE:-

ANTIBODY	PATTERN	DISEASE ASSOCIATION
ANA	Speckled	Scleroderma
		Sjogren"s" Syndrome
		Mixed Connective Tissue Disease
		SLE.

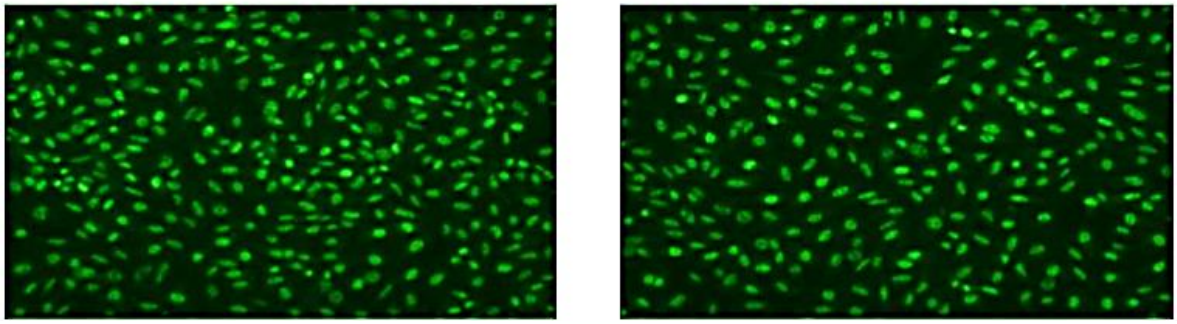

Figure 1: Report of Antinuclear antibodies (ANA) by IFA.

conditions can complicate diagnosis and treatment, making early recognition and appropriate management crucial.

CASE PRESENTATION

A 46-year-old female reported bilateral symmetrical joint pain affecting hands, wrists, elbows and ankles, accompanied by joint swelling and morning stiffness lasting over an hour for the last 4 months. She also reported symptoms of dry mouth, dry eyes, generalized lethargy and fatigue. There were no associated symptoms such as oral ulcers, skin rashes, photosensitivity or changes in bowel or urinary function. The patient was married with four children and had normal menstrual cycles. She had no history of smoking, alcohol consumption, or illicit drug use. Her family history was positive for rheumatoid arthritis, affecting her mother and two maternal aunts. On examination, the patient had pallor, but her vital signs were normal. Mild conjunctival redness and dryness of the eyes were noted. The patient had a dry tongue and difficulty forming a saliva pool in the mouth. There was synovitis observed in both elbows, wrists, and multiple proximal interphalangeal joints, with sparing of the distal interphalangeal joints. Both ankles were tender, although there was no visible swelling. Her cardiac, respiratory, and neurological examinations were all unremarkable.

According to investigations, the patient had microcytic anemia, with a hemoglobin level of 9.1 g/dL and a mean corpuscular volume (MCV) of 65 fl. Her total leukocyte count (TLC) and platelet count were normal. Liver and renal function tests, along with urinalysis were normal. The autoimmune profile revealed positive rheumatoid factor (RA factor) at 332 U/ml, anti-cyclic citrullinated peptide (Anti-CCP) antibodies at 117 u/ml, antinuclear antibodies (ANA)

by IFA at 1:640 in speckled pattern as shown in Figure 1, and anti-SSA/Ro60kd, anti-SSA/Ro52kd and anti-SSB/La antibodies at 2.8, 2.9 and 1.9 respectively through membrane-based enzyme immunoassay. Serologic testing for hepatitis B, hepatitis C and HIV showed negative results. Her serum iron was low at 30 mcg/dL and total iron-binding capacity (TIBC) was elevated at 760 mcg/dL. Additional investigations including ECG**,** chest X-ray, and abdominal ultrasound were unremarkable, ruling out any other major systemic complications.

Considering the clinical presentation and laboratory results, a diagnosis of Sero-positive rheumatoid arthritis complicated by secondary Sjögren's syndrome and iron deficiency anemia was established. The patient was started on Methotrexate, Hydroxychloroquine and a brief course of oral Prednisolone. Iron, calcium and vitamin D supplements were prescribed to manage anemia and support bone health. Artificial tears and oral lubricant gel were also prescribed to manage the symptoms of dryness associated with Sjögren's syndrome. At her 8-week follow-up, the patient showed significant improvement in her joint pains, with steroids having been tapered and eventually discontinued. She was tolerating the Methotrexate and Hydroxychloroquine without any adverse effects.

DISCUSSION

Sjögren's syndrome affects up to 1.0% of the general population.[5] In the United States around four million people are affected by Sjögren's syndrome, with over 90% being women. The disorder is observed globally in both adults and, less frequently children, with no significant racial or geographic preference. While the disease can develop at any age, the average age of onset is in the late 40s. However, it exhibits a notable gender imbalance, with a female-to-male ratio of approximately 9:1.[5] The patient in this case was a 46-year-old woman and falls within the typical age range for the onset of Sjögren's syndrome, which commonly occurs in individuals in their late 40s to early 50s. Furthermore, her gender aligns with the pre-existing data indicating that the disorder primarily affects women, emphasizing the relevance of her demographic characteristics in the context of this autoimmune condition.

The ACR/EULAR 2010 diagnostic criteria are commonly used for diagnosing RA.[6] Our patient met these criteria, with positive RA factor and anti-CCP antibodies, confirming the diagnosis of Sero-positive RA. The diagnosis of secondary Sjögren's syndrome should be considered when a patient develops severe dryness of the eyes and mouth on the history of an existing autoimmune disease. Unlike primary Sjögren's, a lip biopsy is usually not required for diagnosis, as the clinical symptoms, along with the presence of another autoimmune condition, are typically enough to confirm the diagnosis. The present case emphasizes the importance of a thorough autoimmune workup in patients with inflammatory joint symptoms, particularly in those with a family history of RA. The detection of positive anti-SSA/Ro and anti-SSB/La antibodies provides additional evidence supporting the diagnosis of secondary Sjögren's syndrome in this case.

The treatment of Sjögren's syndrome focuses on managing symptoms and reducing dryness, as there is no cure. Key approaches include maintaining good oral hygiene, using artificial tears and thicker eye ointments, and employing punctal plugs or cautery for severe cases of dry eyes.[7,8] Medications such as Pilocarpine and Cevimeline help stimulate the secretion of saliva, while immunosuppressants like Methotrexate may be prescribed for more severe cases. A regular exercise is beneficial in managing fatigue and joint pain. In our case, the patient was treated with Methotrexate, Hydroxy chloroquine and Steroids in addition to artificial tears and oral lubricants. This case is important as it illustrates the successful multimodal treatment approach as well as the positive outcomes in symptom control and medication tolerance, offering clinical evidence for clinicians in managing similar cases. It is also vital to look for extra-articular complications and associated conditions in patients with RA and other autoimmune disorders especially hematological conditions which may be attributable to chronic inflammation, nutrient deficiencies, blood loss or hemolysis.[9,10] Our patient had co-existing iron deficiency anemia. The management of iron deficiency anemia associated with chronic inflammation and autoimmune disease is also an important aspect of comprehensive care.

CONCLUSION

The present case highlights a patient with Sero-positive rheumatoid arthritis and secondary Sjögren's syndrome, presenting with joint pain and dryness of

the eyes and mouth. Treatment with Methotrexate, Hydroxychloroquine and a short course of oral Steroids led to significant improvement in symptoms within 8 weeks, highlighting the significance of prompt diagnosis and intervention in autoimmune disorders, particularly in relation to ocular complications. In conditions like rheumatoid arthritis and Sjögren's syndrome, regular eye examinations are crucial for detecting and addressing issues such as dry eye syndrome and corneal damage.

Patient's Consent: Researchers followed the guide lines set forth in the Declaration of Helsinki.

Conflict of Interest: Authors declared no conflict of interest.

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Authors Designation and Contribution

Nauman Ismat Butt; Assistant Professor: *Concepts, Design, Literature Search, Data Acquisition, Manuscript Preparation, Manuscript Editing, Manuscript Review.*

Barak Waris; House Officer: *Concepts, Design, Literature Search, Data Acquisition, Manuscript Preparation.*

Muhammad Sohail Ajmal Ghoauri; Postgraduate Resident: *Concepts, Design, Literature Search, Data Acquisition, Manuscript Preparation.*

Ayesha Afzal; House Officer: *Literature Search, Manuscript Editing, Manuscript Review.*

Muhammad Waseem Riaz; House Officer: *Data Acquisition, Manuscript Editing, Manuscript Review.*