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Case Report

Sjögren's syndrome after silicone breast implantation

Lütfi Akyol, Soner Önem, Metin Özgen, Mehmet Sayarlıoğlu

Abstract

Sjögren's syndrome, an autoimmune disease characterized by lymphocytic infiltration of the lacrimal and salivary glands, leads to dryness of the mouth and eyes. Herein, we present a case of Sjögren's syndrome that developed after silicone breast implantation. A cause-effect relationship between breast implantation and Sjögren's syndrome has not been established. However, the possibility of such an association should be considered when a patient with silicone implants is admitted to the hospital for treatment of Sjögren's syndrome.

Keywords: Silicone, Sjögren's syndrome, breast implantation

Introduction

Sjögren's syndrome is an autoimmune illness characterized by lymphocytic infiltration of the lacrimal and salivary glands that causes dryness of the eyes and mouth. A cause-effect relationship between Sjögren's syndrome and silicone implantation has not been established. Recently, a new syndrome, namely the Autoimmune/inflammatory syndrome induced by adjuvants (ASIA) (1), has been defined. The conditions included in ASIA syndrome share different common signs and symptoms and seem to be induced by the presence of adjuvants. These so-called adjuvants are substances able to boost the immune response and act as trigger for autoimmune diseases development (2). Herein, we present a case of Sjögren's syndrome that developed after silicone breast implantation.

Case Presentation

A 34-year-old female patient was admitted to our hospital for a 2-year history of dry mouth, dry eyes, and cyanosis of her fingers. The cyanosis manifested more distally on her fingers when exposed to cold weather. The patient reported no other abnormalities. She had undergone silicone breast implantation 5 years ago, and her symptoms began after the surgery. There was no history of illness or drug use. Physical examination findings were normal.

The patient's laboratory results were as follows: leukocyte count, 2800/μL; lymphocyte count, 950/μL; erythrocyte sedimentation rate, 24 mm/h; and C-reactive protein level, <3 mg/L (Table 1).

The patient was negative for anti-human immunodeficiency virus, anti-hepatitis B surface antigen, and anti-hepatitis C virus anticore antibodies. She was positive for antinuclear antibody (granular pattern, 1:3200) and anti-SS-A and anti-SS-B were positive in the extractable nuclear antigen profile; she was negative for all other serologic markers. Moreover, she was negative for rheumatoid factor and anti-cyclic citrullinated peptide antibody. No atypical cells were found on a peripheral smear. Chest radiograph was normal. Dry eye was diagnosed with a Schirmer test. A salivary gland biopsy specimen revealed >50 lymphocytic periductal lymphoid aggregates within four areas. Capillaroscopy was normal. The patient was diagnosed with Sjögren's syndrome, and treatment with hydroxychloroquine was initiated. The patient's leukocyte count normalized during the follow-up period, and the hydroxychloroquine treatment is being continued.

Discussion

Silicone breast implantation is associated with several illnesses, the most controversial of which are connective tissue disorders (3). Silicone in various tissues of the bodies of women with breast implants has led to the incrimination of these implants as causative factors of connective tissue disorders (4). Karlson et al. (5) demonstrated a considerably increased risk of any specific connective tissue disorders, all specific connective tissue disorders combined, and other autoimmune or rheumatic diseases in association with silicone. The expected summary relative risk for scleroderma or systemic sclerosis, rheumatoid arthritis, and systemic lupus erythematosus is generally ≤1. According to Okano et al. (6), breast augmentation with silicone may have facilitated the development of scleroderma and Sjögren's syndrome.

Department of Internal Medicine, Division of Rheumatology, Ondokuz Mayıs University Faculty of Medicine, Samsun, Turkey

Address for Correspondence: Lütfi Akyol, Ondokuz Mayıs Üniversitesi Tıp Fakültesi, İç Hastalıkları Anabilim Dalı, Romatoloji Bölümü, Samsun, Türkiye

E-mail: drakyol19@gmail.com

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Table 1. Laboratory parameters

Parameter	Results	Normal values
Complete blood count		
Leukocyte	2800/μL	4000-10000
Lymphocyte	950/μL	1.300-3.600
Hemoglobin	13 g/dL	13.5-17.5
Platelet	378,000/μL	150-350
Blood biochemistry		
Glucose	81 mg/dL	70-110
Creatinine	0.8 mg/dL	0.7-1.3
Aspartate transaminase (AST)	19 U/L	5-34
Alanine transaminase (ALT)	15 U/L	0-55
Erythrocyte sedimentation rate	24 mm/h	0-20
C-reactive protein	<3 mg/L	0-3
Urinalysis	Normal	
Rheumatoid factor	<10 IU/L	0-10

A large meta-analysis by Janowsky revealed no distinct relationship between breast implantation and any of the specific connective tissue diseases, all connective tissue diseases combined, or other autoimmune or rheumatic diseases with the possible exception of Sjögren's syndrome (7). A large cohort study by Sánchez-Guerrero found no association between silicone breast implants and connective tissue diseases or the signs/symptoms of these diseases according to a variety of standardized criteria (8).

Anemia, leukopenia, and lymphopenia can develop in patients with Sjögren's syndrome and connective tissue disorders, such as systemic lupus erythematosus (9). Our patient had no malar rash, discoid rash, arthralgia, renal disease, neurologic disease, oral or nasal ulcers, alopecia, pleuritis, pericarditis, hemolytic anemia, or thrombocytopenia. Therefore, we did not consider systemic lupus erythematosis.

ASIA syndrome was defined to summarize for the first time the spectrum of immune-mediated diseases triggered by an adjuvant stimulus, such as chronic exposure to silicone and other adjuvants that also may have an adjuvant effect. All these environmental factors have been found to induce autoimmunity by themselves. Several mechanisms have been hypothesized to be involved in the onset of adjuvant-induced autoimmunity; a genetic favorable background plays a key role. However, our patient was not a known genetic predisposition. Although there were defined minor and major criteria for ASIA, the most prominent feature being emphasized was the development of the autoimmunity/autoimmune disease after exposure to the adjuvant (10). Therefore, this presented case of Sjögren's syndrome that developed after silicone breast implantation was evaluated as an example of ASIA Syndrome.

In conclusion, a causal relationship between Sjögren's syndrome and silicone breast implantation was found in the present case. From a community health viewpoint, breast implants seem to have a minimal effect on the number of women in whom connective tissue disorders develop. However, they cannot be ignored. The present case serves to remind clinicians of silicone breast implants as an important cause of Sjögren's syndrome.

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