

Serologic Activity against HTLV-I in Patients with Primary Sjögren's Syndrome in Mashhad, an Endemic Area

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Abstract

Background: There are accumulating evidences that Human T cell Leukemia Virus type I (HTLV-I) plays a part in the development of Sjögren's Syndrome (SS). This virus is endemic in some areas of the world. The objective of this study was to estimate the seroprevalence rates of anti-HTLV-I in SS patients in Mashhad and to determine whether any association between SS patients and HTLV-I could be observed.

Methods: We recruited 21 patients with primary SS who consecutively attended our Rheumatology Clinic in Ghaem Hospital affiliated to Mashhad University of Medical Sciences in Mashhad, Northeastern Iran during May 1, 2005 to September 30, 2008. Serum samples were screened for HTLV-I, using an enzyme-linked immunosorbent assay (ELISA). To compare the seroprevalence rates of HTLV-I found in SS patients with those in the community, we used the viral infection rates calculated for the entire population of blood donors in Mashhad by Blood Transfusion Organization.

Results: All patients with SS were negative by ELISA versus HTLV-I.

Conclusion: This study demonstrates no prevalence of positivity for HTLV-1 in SS patients in Mashhad. These data are against the role of HTLV-I in the development of SS in our region.

Keywords: Sjögren's syndrome; Human T cell Leukemia Virus type I (HTLV-I); Northeastern Iran

Introduction

Human T Cell Leukemia Virus type I (HTLV-I) is a human retrovirus endemic in Southern Japan, intertropical Africa, Melanesia, Latin America, and the Caribbean basin.¹ HTLV-I is etiologically associated with adult T cell leukemia,² and HTLV-I-associated myelopathy/tropical spastic paraparesis.^{3,4} HTLV-I may also be the cause of some other inflammatory disorders, such as uveitis,⁵ chronic arthropathy,⁶ pulmonary alveolitis,⁷ and Hashimoto's thyroiditis.⁸ In addition, there is accumulating evidences that HTLV-I plays a part in the development of Sjögren's Syndrome (SS), a systemic

disease in which the exocrine glands are destroyed by autoimmune mechanisms.^{9,10} The virus is also endemic in Mashhad in Northeastern Iran.¹¹ To the best of our knowledge, there has been no prevalence study of HTLV-I seropositivity in SS in our region. The objective of this study was to estimate the seroprevalence rates of anti-HTLV-I in SS patients in Mashhad and to determine whether any association between SS and HTLV-I could be observed.

Materials and Methods

We recruited 21 patients with primary SS (18 females and 3 males) who consecutively attended our Rheumatology Clinic in Ghaem Hospital affiliated to Mashhad University of Medical Sciences in Mashhad, Northeastern Iran during May 1, 2005 to September 30, 2008. The diagnosis of SS was based on the criteria of

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Received: February 25, 2009

Accepted: July 5, 2009

American-European consensus group classification of Sjögren's syndrome,¹² and rheumatoid arthritis. Systemic lupus erythematosus and systemic sclerosis was ruled out in each patient. After the aim of the study was explained to the patients, those who gave informed consent were asked to enter our study. Serum samples were screened for HTLV-I, using an enzyme-linked immunosorbent assay (ELISA; Dia. Pro diagnostic Bioprobes, Italy). To compare the seroprevalence rates of HTLV-I found in SS patients with those in the community, we used the viral infection rates calculated for the entire population of blood donors in Mashhad (n=60,892) while referring to Blood Transfusion Organization from March 2001 to March 2002.¹¹

Results

A total of 21 patients were tested for HTLV-I. They consisted of 18 women and 3 men with a mean age of 46±4.6 years. All the patients were Iranians living in Mashhad (HTLV-I endemic areas), in the northeastern part of Iran. SS was diagnosed according to the criteria of American-European consensus group classification of Sjögren's syndrome.¹² There was no prevalence of positivity for HTLV-1 in our SS patients. (0/21=0% in our patients compared with 404/60,892=0.663% in the control group).

Discussion

HTLV-I is a human retrovirus endemic in some areas of the world.¹ Although HAM/TSP is the major syndrome caused by HTLV-I, the virus may cause systemic immune-mediated inflammatory disorders of various organs including SS.

Transgenic mice expressing Tax protein has an exocrinopathy similar to SS in man.¹³ To clarify the pathogenesis of HTLV-I in "HTLV-I with SS", the TCR V β gene usage was examined by the infiltrating lymphocytes in the target organ.¹⁴ The V β families predominantly used in the labial salivary gland (LSG) from the HTLV-I-seropositive (HTLV- I⁺)

SS patients were more restricted than those from the HTLV-I-seronegative (idiopathic) SS patients. They were commonly V β 5.2, V β 6, and V β 7. These T cells were commonly present in patients with idiopathic SS, being strongly suggested to be most likely involved in the pathogenesis of both HTLV-I-associated and idiopathic SS.

An association between SS and HTLV-I has been suggested indirectly by case reports of HTLV-I infected patients with tropical spastic paraparesis who developed features of SS. Vernant and colleagues were the first researchers who reported this association.⁹ They studied the prevalence of antibodies to HTLV-1 in 34 women with primary SS and found it in 13 of them.⁹ Another study also investigated the seroprevalence rate of HTLV-1 among 76 patients with SS in Nagasaki, an endemic area, and found it in 23% of them. The rate was significantly more than that in the blood donors (3%).¹⁰ Leon-Monzon *et al.* evaluated the presence of antibodies against HTLV and HIV virus in 14 patients with SS in Spain, but the results were different and all the patients were negative versus known retroviruses.¹⁵

Mashhad, a city in Northeastern Iran is also an endemic area for HTLV-I.¹¹ The present study evaluated the prevalence rates of HTLV-I in SS patients for the first time in Iran. All of our patients with SS were negative by ELISA versus HTLV-I. These results are similar to the findings of a similar study in Spain,¹⁵ and in contrast with the results of studies performed in Nagasaki.^{9,10}

This study demonstrated no prevalence of positivity for HTLV-1 in SS patients in Mashhad. There was no association between HTLV-I infection and SS in our region. These data are against the role of HTLV-I in the development of SS in our region.

Acknowledgement

The authors wish to thank Mashad University of Medical Sciences for financial support.

Conflict of interest: None declared.

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