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(CASE REPORT)

Sjögren's Syndrome Revealing HIV Infection in a 36-Year-Old Woman: A Case Report and Literature Review

Jean Caude Bucumi [1,*], Sanaa Jebbar [1], Hassan Ahmed Daoud [2], Sylvestre Nimubona [1], Mwana-Yile Hassan [1], Bongungu Raissa [1], Inas Ouggane [1], Hanani Badi [1], Fatima Ihbibane [1], Ahd Oulad Lahsen [1], Mustapha Sodqi [1] and Latifa Marih [1]

[1] *Department of Infectious Diseases, Ibn Rochd University Hospital of Casablanca, Morocco.*
[2] *Department of Radiology, Ibn Rochd University Hospital of Casablanca, Morocco.*

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Abstract

Human immunodeficiency virus (HIV) infection may present with a wide spectrum of systemic manifestations, occasionally mimicking autoimmune diseases. Sjögren's syndrome is classically considered an autoimmune exocrinopathy but may also occur in association with HIV infection. We report the case of a 36-year-old woman with no previous medical history who presented with progressive bilateral salivary gland enlargement evolving over three months without fever. Imaging revealed bilateral parotid and submandibular sialadenitis with cervical and inguinal lymphadenopathy. Cervical magnetic resonance imaging showed bilateral multicytic parotidomegaly consistent with diffuse lymphoepithelial cysts. Histopathological examination demonstrated grade IV chronic sialadenitis according to Masson's classification. Etiological investigations led to the diagnosis of HIV infection with high viral load and moderate immunosuppression. This case highlights the importance of HIV screening in patients presenting with unexplained chronic bilateral sialadenitis.

Keywords: HIV; Sjögren's syndrome; Salivary gland disease; Lymphoepithelial cysts; Sialadenitis

1. Introduction

Human immunodeficiency virus (HIV) infection remains a major global health concern despite significant advances in antiretroviral therapy [1,2]. Beyond opportunistic infections, HIV is associated with a wide range of inflammatory, autoimmune-like, and lymphoproliferative manifestations that may reveal the disease [3,4].

Salivary gland involvement represents a well-recognized but often underdiagnosed manifestation of HIV infection. It may present as chronic sialadenitis, parotid enlargement, or sicca symptoms, mimicking primary Sjögren's syndrome [5,6]. Distinguishing between primary Sjögren's syndrome and HIV-associated salivary gland disease is essential due to differences in pathophysiology, prognosis, and therapeutic approach [7,8].

We report a case of HIV infection revealed by a Sjögren-like presentation in a young woman with no particular medical history.

* Corresponding author: Jean Caude Bucumi

2. Case Report

A 36-year-old woman with no notable past medical history was referred for evaluation of bilateral salivary gland swelling evolving progressively over three months. The clinical course occurred in an afebrile context, without weight loss, night sweats, or general deterioration [9].

Physical examination revealed bilateral, symmetrical, painless enlargement of the parotid and submandibular glands with firm consistency. Multiple non-tender, mobile cervical lymph nodes were noted, as well as bilateral inguinal lymphadenopathy. Oral examination revealed mild oral dryness without candidiasis or mucosal lesions [10].

Laboratory investigations showed hypochromic microcytic anemia with hemoglobin at 11.8 g/dL. Leukocyte count was 6,490/µL, including neutrophils at 2,170/µL and lymphocytes at 3,700/µL. Platelet count was normal. Renal and hepatic function tests were within normal limits. Serological tests for hepatitis B, hepatitis C, and toxoplasmosis were negative [11,12].

Cervical ultrasound demonstrated bilateral parotid and submandibular sialadenitis associated with cervical lymphadenopathy [13]. Magnetic resonance imaging revealed bilateral parotidomegaly with a multicytic appearance suggestive of diffuse multifocal lymphoepithelial cysts [14,15].

Histopathological examination of minor salivary gland biopsy showed dense lymphocytic infiltration with severe architectural destruction, consistent with grade IV chronic sialadenitis according to Masson's classification [16,17].

Given the clinical and radiological findings, HIV serology was performed and returned positive. Plasma HIV viral load was 105,000 copies/mL, and CD4 T-cell count was 333 cells/mm^3 [18].

2.1. Parotid MRI Images

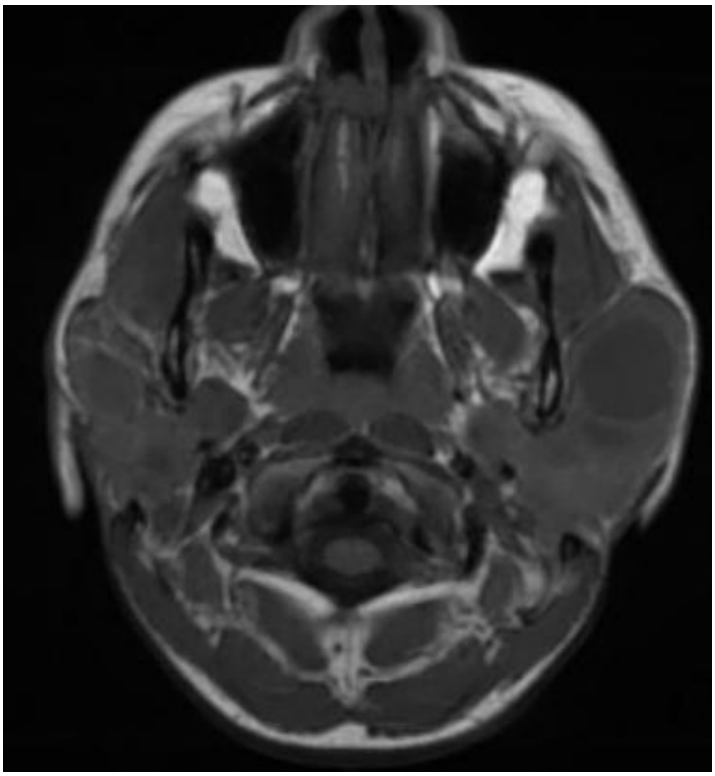

Figure 1a

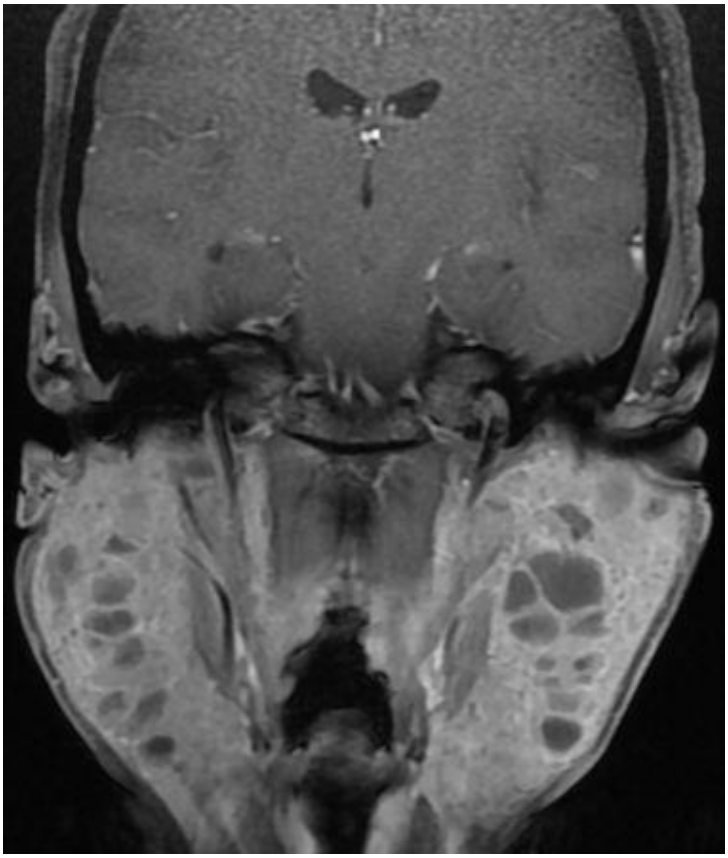

Figure 2b

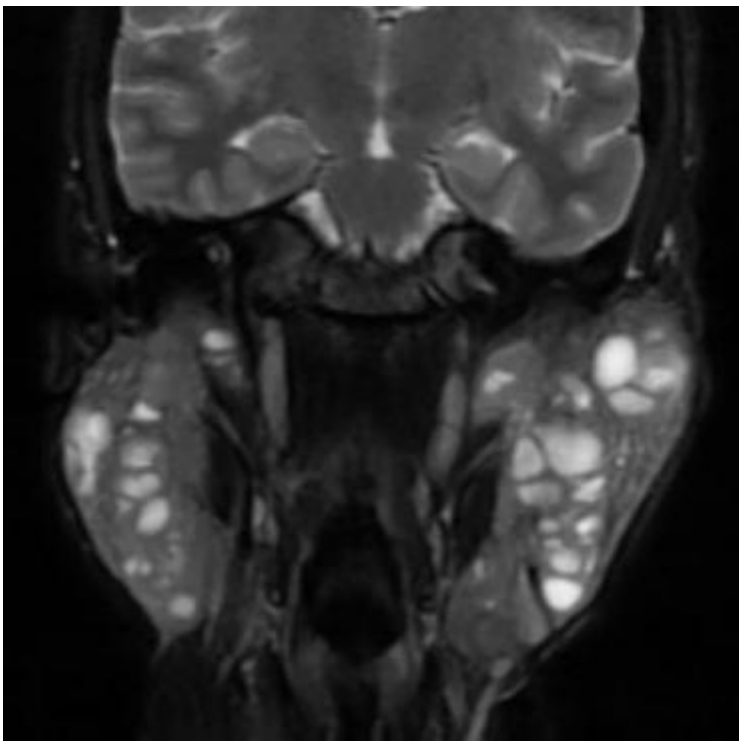

Figure 3 c

Figure 1 MRI of parotid gland with axial T1(a), coronal T2(b)and T1 after godolinium injection (c) images showing multiple intraparotid cystic formations with bilateral parotidomegaly

3. Discussion

HIV-associated salivary gland disease constitutes a distinct clinical entity characterized by chronic inflammation of the salivary glands, parotid enlargement, sicca symptoms, and lymphoepithelial cyst formation [19,20].

Unlike primary Sjögren's syndrome, which is mediated predominantly by CD4+ T lymphocytes and B-cell hyperactivity, HIV-associated sicca syndrome is characterized by CD8+ lymphocytic infiltration and chronic immune activation [21,22]. Autoantibodies such as anti-SSA and anti-SSB are frequently absent, and lymphadenopathy is more prominent [23,24].

The 2016 ACR/EULAR classification criteria for Sjögren's syndrome emphasize the need to exclude conditions such as HIV infection that may mimic the disease [25]. In our patient, although histopathology was compatible with Sjögren's syndrome, the presence of diffuse lymphadenopathy, multicytic parotidomegaly, and positive HIV serology supported a diagnosis of HIV-associated salivary gland disease [26,27].

Antiretroviral therapy remains the cornerstone of management and has been shown to induce partial or complete regression of salivary gland enlargement and improvement of sicca symptoms [28,29]. Immunosuppressive therapy, commonly used in primary Sjögren's syndrome, is not routinely recommended in HIV-associated cases due to limited efficacy and increased infectious risk [30].

This case underscores the importance of systematic HIV testing in patients presenting with chronic bilateral salivary gland enlargement, even in the absence of classic risk factors [31–33].

4. Conclusion

This case illustrates an unusual presentation of HIV infection revealed by a Sjögren-like syndrome. Chronic bilateral salivary gland enlargement associated with lymphadenopathy should prompt systematic HIV screening, particularly in young patients without autoimmune background. Early diagnosis allows appropriate antiretroviral therapy and prevents unnecessary immunosuppressive treatment [34,35].

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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