

Spontaneous Bladder Rupture in a Patient With Human T-cell Lymphotropic Virus Type 1: A Case Report

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Received: December 14, 2013; Accepted: December 18, 2013

Introduction: Spontaneous bladder rupture is a rare manifestation of hyporeflexic bladder due to *HAM/TSP* (human T-cell lymphotropic virus type 1 (HTLV1) associated myelopathy/tropical spastic paraparesis).

Case Presentation: We report a case of spontaneous bladder rupture in a 62-year-old woman with a HTLV1 infection.

Discussion: Bladder over-distention and spontaneous bladder rupture, although very rare in *HAM/TSP*, should be considered in a differential diagnosis of bladder rupture, especially in patients with a history of HTLV1.

Keywords: Human T-lymphotropic virus; Urinary Bladder; Rupture

1. Introduction

Spontaneous bladder rupture is a rare condition, with an incidence rate of 1/126000. In total, 70% of cases have been reported in men, and the overall mortality was 47% (1). It is usually the result of an underlying disease such as: blunt trauma to the lower abdomen, carcinoma of the pelvic organs, radiation therapy of pelvic tumors, large ureterovesical stones, catheterization of the bladder, atonic bladder, bladder irrigation, and neurogenic bladder dysfunction (2). One of the rare underlying diseases that can exhibit neurogenic bladder dysfunction is human T-cell lymphotropic virus type 1 (HTLV) (3). We present a case of spontaneous rupture of the urinary bladder in a patient with a history of *HTLV1*.

2. Case Presentation

A 62-year-old woman from the city of Mashhad (north-east of Iran) was admitted to Mehr Hospital, Tehran; with a chief complain of sudden onset generalized abdominal pain from four hours ago. The pain was continuous in nature, diffusely involved the whole abdomen, and the pain was non-radiating, no other lower urinary tract or gastrointestinal (GI) symptoms were present. In her past medical history, there was a history of *HTLV1* from one year ago (positive *HTLV1* antibodies Elisa and Western blot). She also complained of gradually progressive voiding problems, such as: frequency, dysuria, nocturia, and urge incontinence, for a period of six months. Treatment with conventional interferon α three times a week was started

based upon a diagnosis of overactive bladder syndrome due to *HAM/TSP*. However, one month ago the sensation of incomplete voiding and hesitancy became worse, regrettably this situation was not followed up by the patient. No other risk factors such as: trauma, malignancy, or radiation were found in her past medical history.

On physical examination: she was alert, ill, and pale. Vital signs: blood pressure (BP) = 80/60 mmHg, pulse rate (PR) = 150/minute, respiratory rate (RR) = 24/minute, temperature (T) = 38, an abdominal examination revealed a distended abdomen, bowel sounds were absent, and diffuse tenderness over the whole abdomen with positive rebound tenderness. Laboratory tests were as hemoglobin = 9 mg/dL, white blood cell = 18000/mm³ (net: 78% L: 16%), platelet count = 138000/mm³, creatinine level = 1.9 mg/dL, blood urea = 36 mg/dL, LDH = 498 U/L, AST = 83 U/L, Alt = 33 U/L, total bilirubin = 0/6 (direct = 0/4), negative hepatitis B surface (HBS) Ag, anti HCV Ab, anti-human immunodeficiency virus (HIV) Ab.

An x-ray of the abdomen showed gas under the diaphragm and an abdominal ultrasound revealed free fluid in the abdomen. Based on the clinical and radiological findings the patient underwent a laparotomy, which showed a rupture of the urinary bladder fundus. A bladder biopsy showed necrotizing inflammation and gangrene, but negative for granuloma and malignancy. According to her past medical history of *HTLV1*, detrusor hypo-sensitivity, hypo-activity and bladder rupture due

Implication for health policy/practice/research/medical education:

We report a rare manifestation of human T-cell lymphotropic virus type 1 (*HAM/TSP*).

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to bladder over-distension associated with HAM/TSP, were diagnosed. During a laparotomy, the site of the perforation was repaired and the patient is now undergoing treatment for HTLV1, as a result she has remained asymptomatic after her surgery till the present time.

3. Discussion

HTLV1 is a complex retrovirus belonging to the delta virus family (4). HTLV1 infects 10 to 20 million people worldwide: Southern Japan, the Caribbean, Central and South Africa, South America, and the northeast of Iran are the endemic foci (5). The prevalence of HTLV1 infections in Mashhad City (northeast of Iran) is 1.16% (6, 7), and HTLV1 infects the body's CD4 cells. There are several routes of viral transmission, including: sexual, mother to baby, transfusion, needle stick, and breast feeding (8). The standard practice for a diagnosis of HTLV1 infection is the detection of antibodies by an enzyme immunoassay (EIA) technique, followed by a confirmatory test with Western blot or molecular methods (9). HTLV1 has two major clinical presentations: 1) adult T-cell leukemia lymphoma (ATLL) and 2) a neurologic disease called HAM/TSP (10). Patients with HAM/TSP present with spastic paraparesis in the lower extremities, mild disturbance of sensation and urinary dysfunction (8). The pathogenesis of neurological manifestations in a HTLV1 infection are poorly understood. Up to 90% of HAM/TSP patients have urologic manifestations, such as: urgency, nocturia, urge incontinence, dysuria, and straining (11, 12). In the aforementioned group, the major abnormality is over-active bladder syndrome, and detrusor over-activity, followed by detrusor external sphincter dyssynergia (13). However, 3.84% of patients showed a hyporeflexic bladder and detrusor hypo-sensitivity and hypo-activity. In this group, the urological manifestation is bladder over-distention, secondary to a large volume of urine, which in turn causes hesitancy, sensation of incomplete voiding, and spontaneous bladder rupture (14). A diagnosis of bladder rupture is difficult to make and it requires a high index of suspicion. The most common presentation is diffuse abdominal pain and lower abdominal tenderness, confirmation of the diagnosis is almost always done with a laparotomy (5). In conclusion, bladder over-distention and spontaneous bladder rupture, although very rare

in HAM/TSP patients, should be kept in mind when making a differential diagnosis of spontaneous bladder rupture, especially in a high HTLV1 prevalence country such as ours. In addition, aurodynamic evaluation should be performed in all HTLV1 infected individuals with voiding complaints.

Authors' Contributions

All authors contributed equally in data acquisition, analysis, writing, and final approval of the manuscript.

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