Case Report

Chronic Urinary Schistosomiasis in a 25-Year-Old Man, a Case of Neglected Tropical Disease

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Abstract

Schistosomiasis is a parasitic disease caused by several species of trematodes, a parasitic worm of the genus Schistosoma. Out of the two major forms of schistosomiasis that exist, Schistosoma haematobium affects the genitourinary system. Radiologic imaging manifestations of urinary tract schistosomiasis are observed mainly in the ureters and bladder due to deposition of eggs on them, which elicits chronic granulomatous injury. This eventually causes nodules, polypoid lesions and ulcerations of the lumen of the ureter and bladder that manifest clinically in urinary frequency, dysuria, and terminal hematuria. It may ultimately end in renal failure or carcinoma of the bladder. This endemic but poorly reported neglected tropical disease is presented to increase the index of suspicion and reduce its complications and prevalence.

Keywords: 25-year-old man, chronic urinary schistosomiasis, neglected tropical disease

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Introduction

Schistosomiasis (also known as bilharzia, bilharziasis, or snail fever) is a parasitic disease caused by several species of trematodes (platyhelminth infection or "flukes"), a parasitic worm of the genus *Schistosoma*. In 1851, German physician Theodor Bilharz described this parasitic infection, which consequently became known as bilharziasis but was later renamed schistosomiasis.[1]

Two major forms of schistosomiasis exist: intestinal and genitourinary. Intestinal schistosomiasis may be caused by any of five main species of blood flukes. However, only *Schistosoma haematobium* affects the genitourinary system.[2] *S. haematobium* is endemic throughout Africa, Madagascar, Mauritius, the southern shore of the Mediterranean, and the Middle East, including Turkey.[3,4] Schistosomiasis is endemic in Egypt, exacerbated by the country's dam and irrigation projects along the Nile. The ancient Egyptians recognized schistosomiasis as a cause of bloody urine and knew that the condition was due to worms.[3]

Radiologic imaging manifestations of urinary tract schistosomiasis are observed mainly in the ureters and bladder. The kidneys appear normal until a late stage of disease. Trapped eggs mature normally, secreting antigens that elicit a vigorous immune response. The eggs themselves do not damage the body. Rather, it is the cellular infiltration resulting from the immune response that causes the pathology classically associated with schistosomiasis. Hematuria, the first clinical sign of established genitourinary schistosomiasis, appears 10–12 weeks after infection. Dysuria and hematuria are common in both early and late stages of the disease.[4,5]

Males are generally more infected and with higher intensity than the females, particularly children who may acquire the disease by swimming or playing in infected water.[6]

In urinary schistosomiasis, the risks of hematuria, dysuria, nutritional deficiencies, lesion of the bladder, kidney failure, an elevated risk of bladder cancer, and growth retardation in children are well established.[4,5,7] Ureteral involvement

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in schistosomiasis has been reported in as many as 65% of cases.[8,9] In regions where schistosomiasis is endemic, it constitutes a major risk factor for bladder malignancies, predominantly for squamous cell carcinomas, which account for more than 50% of bladder cancers.[3]

This endemic but poorly reported disease is presented to increase the index of suspicion and reduce its complications and prevalence in resource-constrained tropical areas of the world like Nigeria.

Case Report

BM is a 25-year-old male sugarcane farmer who presented at the General Outpatient Clinic of the Ahmadu Bello University Teaching Hospital, Zaria, with a 6-month history of abdominal pain associated with cough and fever. He also developed pruritic rash and hematuria before presentation. The abdominal pain was colicky and more localized at the suprapubic and lumbar regions bilaterally. The pain was neither aggravated nor relieved by any known factor and did radiate to the waist. The symptoms were also associated with a significant fatigue, despite good appetite and nutrition.

The medical history revealed previous treatment of hematuria with native medication. No history of trauma or symptoms referable to gastrointestinal and central nervous systems was noted. There was no history or sign of past abdominal surgery. The patient is the fifth of eight children in a polygamous setting. His cousin also had a history of hematuria.

On general physical examination, he was febrile, mildly pale, anicteric, not dehydrated, not irritable, or in respiratory distress. He had no pedal edema but had inguinal lymphadenopathy. Skin discoloration of the lower extremities was noted.

Abdominal examination revealed mild tenderness in the renal angles and suprapubic area, but no palpable mass was demonstrated, and rectal examination was unremarkable.

The initial clinical impression was acute pyelonephritis, and radiological investigations were then requested. Plain abdominopelvic radiograph showed oval-shaped calcifications in the pelvic cavity [Figure 1].

The bowel gas distribution and the bony architecture are preserved. The abdominal ultrasound scan revealed bilateral hydronephrosis and hydroureter as well as thickened urinary bladder walls. These findings were also confirmed on intravenous urographic (IVU) study [Figure 2]. In addition, IVU showed bilateral marked tortuous ureteral dilatation that involved their entire length, and dilatation was more on the right side. The urinary bladder showed irregular outline, reduced capacity, and multiple filling defects [Figures 2 and 3]. The preliminary film demonstrated urinary bladder wall calcifications [Figure 1].

The computed tomography (CT) urography and magnetic resonance imaging were however not done due to financial constraints. In view of the aforementioned findings,

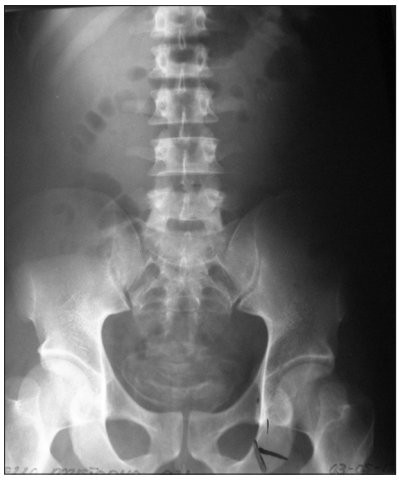

Figure 1: Anteroposterior plain radiograph of the abdomen and pelvis showing peripheral circumferential calcification (arrow) of the urinary bladder wall with coarse and thicker calcifications in the center pathognomonic of chronic urinary tract schistosomiasis. Spina bifida of the sacral vertebrae are also noted

radiological diagnosis of chronic urinary schistosomiasis was made which was further confirmed by the findings of *Schistosoma* ova at urine microscopy and in histological report.

The patient was commenced on praziquantel and broad-spectrum antibiotics. He was managed on outpatient basis after initial investigation and stabilization on admission to correct electrolyte imbalance and infections. The patient is still being followed up at the urology clinic.

Discussion

In 1996, the World Health Organization[4] estimated that more than 200 million people worldwide were affected by schistosomiasis, mainly those living in rural agricultural and periurban areas. Of that number, it was estimated that 20 million were severely affected by the disease and that another 120 million were symptomatic. Schistosomiasis is the second most socioeconomically devastating parasitic disease after malaria.[2]

Nigeria is also one of the tropical countries known to be highly endemic for urinary schistosomiasis.[6] This patient probably may have got infected during repeated daily contact with contaminated freshwater sources from his sugarcane plantation, which constitutes a major occupational health risk in these rural areas. Unsanitary disposal of human and animal wastes was the other source of infection.

Male gender in the present patient is a predisposing factor, similar to the observations made in Southeastern Nigeria.[6] These presumably due to more frequent water contact activities by male pupils, particularly in the swamp-rice farming and fishing, where fathers engage every male in their household in the profession.

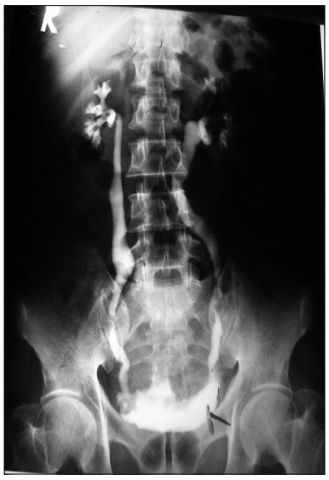

Figure 2: Excretory urogram demonstrates bilateral tortuous pelviureteric dilatation. Multiple filling defects along the course of the distal ureters are noted representing ureteritis cystica due to schistosomiasis

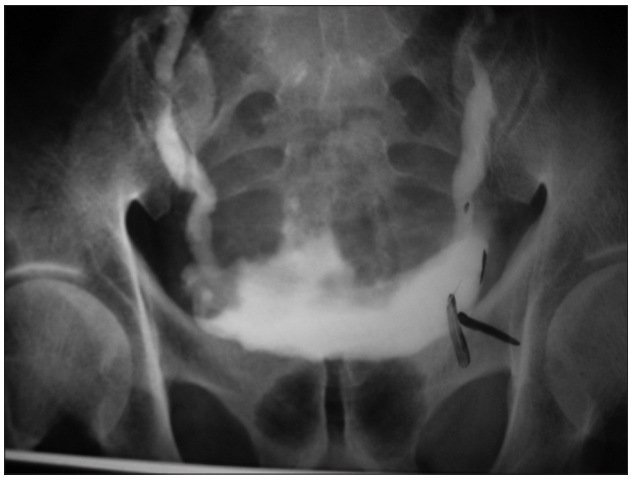

Figure 3: Bladder view of intravenous urographic showing persistent filling of both distal ureters. Multiple filling defects and irregularities of the urinary bladder wall with associated reduced capacity are seen indicative of cystitis cystica due to chronic schistosomiasis

Clinical manifestations reflect the parasite's developmental stage and the host's response to toxic or antigenic effects of the parasite and its eggs. During the early stage of infection, the patient may present with dermatitis caused by cercarial penetration of the skin. This may account for the skin discoloration noted on the lower limbs of this patient. The stage of egg deposition is manifested by genitourinary symptoms of cystitis, dysuria with terminal hematuria, dull suprapubic pain, and hemospermia, which were consistent with the presentation in the current patient with the exception of hemospermia. Hematuria, the first clinical sign of established genitourinary schistosomiasis, appears 10–12 weeks after infection.[4,5]

This symptom is so common in some communities where it was named male menstruation, a term occasionally unofficially used for a type of bleeding in the urine, reported in some tropical countries. Mostly noted in peoples who work in tropical wet places such as rice fields where majority of the boys pick up *Schistosoma*, and the bleeding manifestation start at about puberty stage. The uneducated ones think that it is normal and refer to it as the male equivalent of female menstruation. However, this is to be distinguished from genuine menstruation in an anatomically intersex human who has a functioning menstruating womb but male external sexual organs.

Pathologic changes in the urinary tract due to schistosomiasis are far more common in chronic infections than in acute ones. Such changes result from the deposition of eggs (not adult flukes) in and around vessels, which leads to chronic inflammatory lesions and induces an immune response with granuloma formation and associated fibrotic changes.[10] Radiologic imaging manifestations of urinary tract schistosomiasis are thus observed mainly in the ureters and bladder where large amount of eggs are deposited; the kidneys appear normal until a late stage of disease [Figures 1 and 2].

The disease usually starts at the urinary bladder trigone and base, with the formation of submucosal granulomas, leading to inflammatory patches and hematuria as already explained. In the early stages of schistosomal infection, the bladder outline becomes hazy and ill-defined at urography because of submucosal edema and pseudotubercles. Schistosomiasis is the most common cause of bladder wall calcification in regions where *S. haematobium* is endemic; it accounts for as many as 56% of cases of such calcification.[8,11] Calcification becomes detectable when CT attenuation exceeds 160 HU.[8] This however was not done due to high cost.

At radiography, bladder calcifications are visible first at the bladder base, forming a linear pattern that parallels the upper border of the pubic bone; eventually, calcium deposits encircle the entire bladder.[11] This is consistent with the findings in the present patient [Figure 1]. The classic presentation of a calcified bladder, which resembles a fetal head in the pelvis, is pathognomonic of chronic urinary tract schistosomiasis. Various calcification patterns have however been reported, depending on the state of bladder filling. In empty bladder, calcifications appear coarser and thicker because the collapsed bladder wall has thick folds. Other patterns that may be seen include fine granular, fine linear, and thick irregular calcification. Calcification may encircle the bladder as seen in this patient [Figure 1] or may affect only parts of it; calcification may be more marked on the one side than the other, or more marked at the base than at the dome. A shell-like rim of calcification, an appearance produced by the submucosal deposition of the eggs and not caused by fibrosis, has little or no effect on bladder capacity or emptying. Later, in the course of infection, the bladder wall becomes fibrotic and the bladder contracts, leading to a reduction in capacity as seen evolving in the present patient with irregularities of its outline[12] [Figure 3].

In addition, in late-stage cystitis glandularis, a papillary or polypoid mass may arise that mimics carcinoma which IS demonstrated as filling defects in contrast filled urinary

bladder as seen in the radiograph of this patient. In regions where schistosomiasis is endemic, it constitutes a major risk factor for bladder malignancies, predominantly for squamous cell carcinomas, which account for more than 50% of bladder cancers.[7]

Ureteral involvement in schistosomiasis has been reported in as many as 65% of cases.[8,9,13] More than 80% of the earliest ureteral strictures occur in the intravesical segment of the ureter, and as fibrosis progresses, the entire length of the ureter may be involved, with multiple strictures.[14] The peristaltic movement of the ureter is affected, and vesicoureteral reflux also occurs as a late complication of schistosomiasis and present in about 30% of patients.[15] The kidneys are not the main target of *S. haematobium*, but they may be affected in cases of ureteral obstruction and vesicoureteral reflux. The dilated ureters found in the current patient are believed to result from atony consequent upon the urethritis as no clear ureteral stenosis, stricture, or calcification was seen.

Schistosomiasis is readily treated using a single oral dose of the drug praziquantel. As with other major parasitic diseases, there is ongoing extensive research into developing a schistosomiasis vaccine that will prevent the parasite from completing its life cycle in humans.

Conclusion

A 25-year-old man with an endemic but neglected tropical disease like chronic urinary schistosomiasis has been presented to increase the index of suspicion and reduce its complications and prevalence in tropical areas.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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