EJR European Journal of Rheumatology

DOI: 10.5152/eurjrheum.2017.16084

Case Report

Bilharzial arthropathy: Rare cause of chronic arthritis in tropical areas

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Abstract

Bilharziasis is a parasitic disease that affects the urinary tract and intestines. Finding bilharzia in joints is exceptional. We report two cases of Malagasy patients living in a highly endemic bilharziasis area and having chronic arthritis due to bilharziasis. The first case was a 32-year-old man presenting with a clinical picture of spondylitis with chronic oligoarthritis and paroxysmal asymmetric in his lower limbs, accompanied with sacroiliac pain and episodic dysentery. We retained the diagnosis of bilharziasis arthritis because of positive schistosomiasis serology in the joint fluid and the blood, poor response to therapy with nonsteroidal anti-inflammatory drugs (NSAIDs), and high efficacy of treatment with specific antiparasitic drugs. The second case was a 42-year-old woman with pain in her knees, hips, and low back for 1 year. She was presented inflammatory pain which were persisting despite the use of non steroidal anti inflammatory drugs. She had a history of splenectomy resulting to a splenomegaly by liver-spleen schistosomiasis. Results of paraclinical examinations were similar with the first case as well as the high efficacy of anti-parasitic treatment. *Schistosoma haematobium* and *S. mansoni* are the endemic strains found in Madagascar. The lifestyle of the Malagasy population exposes them to recurrent infestations, even massive. If the affinity for joint of schistosoma is known, the diagnosis of bilharzial arthropathy is exceptional. The diagnosis is based on a set of clinical and biological arguments as well as evolution. Serological testing for bilharzia in joint fluid could be an efficient way of diagnosis. So, in a country where bilharzia is endemic, clinical presentation of spondylitis or nondestructive chronic polyarthritis should lead to bilharzia arthropathy.

Keywords: Bilharzia, arthritis, serological testing, praziquantel

Introduction

Frequently observed in tropical areas, bilharziasis is a parasitic disease that affects the urinary tract and intestines. Joint location of bilharzia is exceptional. We distinguish parasitic rheumatism by immunological mechanism and bilharziasis arthritis characterized by the presence of the parasite inside the joint. Diagnosing these articular manifestations remains difficult and is based on a set of clinical and biological arguments.

We report on two cases of Malagasy patients living in a highly endemic bilharziasis area and having chronic arthritis due to bilharziasis.

Case Presentation

Written informed consent was obtained from patients before performing different examinations.

The first case was a 32-year-old man presenting with chronic oligoarthritis and paroxysmal asymmetric in his lower limbs for the past 7 years, accompanied with sacroiliac pain and episodic dysentery.

Accordingly, we diagnosed him with non-radiological spondylitis. Treating it with nonsteroidal anti-inflammatory drugs (NSAIDs) and sulfasalazine (Salazopyrin; Pfizer, Paris, France) resulted in transitional efficiency. The presence of hypereosinophilia in a blood test at 1008/mL justified a search for parasites, such as bilharzia; we consequently obtained a positive blood serology of 1/1280. An examination of joint fluid revealed an inflammatory-type fluid, but no microorganisms were identified. The schistosomiasis serology in the joint fluid was also positive at a dilution of 1/1280.

An histological examination of the synovial biopsy was in favor of non-specific synovitis. An X-ray of the painful joints showed they were normal.

We have mentioned the diagnosis of bilharziasis arthritis, and the patient was administered a single dose of praziquantel at 40 mg/kg (Biltricide; Bayer Santé, Elberfeld, Allemagne), renewable after 5 months. A year later, the patient showed no joint pain- bilharziasis arthritis was considered to be another possible reason.

Cite this article as: Rakotomalala HN, Ranaivoarison MV, Andrianjafison F, Ralandison DS.Bilharzial arthropathy: Rare cause of chronic arthritis in tropical areas. Eur J Rheumatol 2017; 4: 229-30.

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Submitted: 19 October 2016
Accepted: 5 March 2017
Available Online Date: 04 July 2017

The second case was a 42-year-old-woman with bilateral knee pain and sciatica, accompanied with low back pain, inflammation, and NSAID resistance, all occurring over 1 year. There were no extra-articular findings. She had a history of splenectomy following a splenomegaly by hepatosplenic schistosomiasis. An X-ray revealed no specific anomaly, particularly the absence of sacroiliitis. A biological test showed biological inflammatory syndrome with hypereosinophilia at 1600/mL. The joint fluid was of an inflammatory-type with 40,000 white cells/mL, lymphocyte predominance (92%), and germ-free. The clinical settings justified bilharziasis serology in the blood and joint fluid, both revealing a positive result at a 1/640 dilution. We then raised a diagnosed of bilharzial arthritis. After 3 days of treatment with 40 mg/kg praziquantel (Biltricide; Bayer Santé, Elberfeld, Allemagne), the joint pain disappeared in 3 weeks. One year later, there was no recurrence.

Discussion

Bilharziasis, or schistosomiasis, is part of a group of parasitic diseases caused by the helminth worm species that are non-hermaphroditic blood-sucking trematodes. Six species are harmful to humans and are highly prevalent in warm climates across three continents. Schistosomiasis is believed to affect approximately 200 million people worldwide and causes >300,000 deaths annually (1).

Schistosoma haematobium and *S. Mansoni* are the endemic strains in Madagascar. They are primarily responsible for urogenital schistosomiasis and intestinal and/or hepatosplenic revealed by hematuria or dysentery (1). The lifestyle of the Malagasy population exposes them to recurrent infestations, even massive. Madagascar's national prevalence is 31%; in regions most infested by bilharziasis, the prevalence is 70% (2, 3).

If the athrophile character of schistosoma is known, the diagnosis of bilharzial arthropathy is exceptional. On the one hand, we distinguish schistosomiasis arthritis, which causes mono- or oligoarthritis mainly in the large joints of the lower limbs. The symptomatology might suggest septic arthritis or the beginning of spondylitis; the presence of parasites in the joint fluid from an infected joint facilitates the diagnosis (4, 5). One the other hand, there is parasitic rheumatism, which presents a polymorphous clinical picture; most of the time an inflammatory polyarthritis with synovitis affecting both small and large joints with acute, subacute, or chronical evolution in which we do not find parasites in the joint but in another part of the organism. Antirheumatic drugs are inefficient in this case (5-7). Based on the Doury criteria, the diagnosis of these manifestations is based on a set of clinical arguments (oligo-, mono-, or polyarthropathy or living in a endemic area) and biological arguments: blood hypereosinophilia, increased erythrocyte sedimentation rate, inflammatory joint fluid but no visible parasites, parasite identification (in the stool, in the duodenal fluid, in the urine, into the dermis, or parasite immunology tests), lack radiographic evidence, failure of NSAIDs, and dramatic effectiveness of specific antiparasitic treatment (7, 8).

Many blood tests of parasitic immunology are available, but the research of antibodies in the joint fluid has not been the aim of any study (7-10). Positivity of the schistosomiasis serology in joint fluid from our patients could open up an area of research in the diagnosis of this disease.

Few cases of schistosomiasis arthritis and parasitic rheumatism have been reported in the literature. The most important one was reported by Bassouini et al. (13); in their study, 124 patients were included. Arthralgia concerned both large peripheral joints than the spine, they were not disabling and not accompanied by morning stiffness, no significant inflammatory signs or deformity. Synovial biopsy showed the presence of parasite eggs in only three cases. Two other cases of coxarthritis schistosomiasis were described with the discovery of schistosoma eggs in the synovium, reflecting its scarcity and non-essential character to make diagnosis of joint bilharzia (13-15).

In 1966, Girge described 36 cases of arthritis among patients with a history of schistosomiasis. The disappearance of arthritis following antiparasitic treatment was one of the strong arguments, enabling the diagnosis of parasitic rheumatism (14). The same evolutionary profile was found in the cases diagnosed by May et al. (15). However, in none of these cases, the parasite in question was found in either the liquid or in the synovial membrane.

Serological testing for antischistosomal antibody in joint fluid was not realized in these reported cases, which makes our two cases unique in that they met all the diagnosis criteria of Doury (7). The positive serology reaction caused by schistosomiasis in joint fluid could be an additional argument for the diagnosis of bilharzial arthropathy, a technique that is technically and financially accessible in several tropical areas.

So, in a country where bilharzia is endemic, a clinical presentation of spondylitis or nondestructive chronic polyarthritis with hypereosinophilia should lead to bilharzia arthropathy. Serological testing for bilharzia in joint fluid could be an efficient way of diagnosis.

Ethics Committee Approval: N/A.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - R.D.S., R.H.N.; Design - R.D.S., R.H.N.; Supervision - A.F., R.D.S.; Resources - R.D.S., R.H.N.; Materials - A.F., R.D.S.; Data Collection and/or Processing - R.V., R.H.N.; Analysis and/or Interpretation - R.V., R.H.N.; Literature Search - R.H.N.; Writing Manuscript - R.H.N.; Critical Review - R.D.S.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

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