





Case Report

# **Expecting the Unexpected: Schistosomial** Appendicitis in Nonendemic Area

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## **Abstract**

Background Schistosomiasis is a granulomatous disease prevalent in tropical and subtropical areas. Appendicitis secondary to schistosomiasis is a rarely encountered presentation, even in endemic areas. Most acute intestinal schistosomiasis presents with abdominal pain and bloody diarrhea. Other atypical clinical presentations include intestinal obstruction, peritonitis, and acute appendicitis.

## **Keywords**

► acute appendicitis

schistosomiasis

► endemic disease

► acute abdomen

**Case Description** We report a case of a 37-year-old male from the Philippines who presented with the clinical picture of acute appendicitis and was treated with laparoscopic appendectomy. Pathological study revealed calcified Schistosomia japonicum's egg in a background of acute inflammation.

**Conclusion** Worldwide increase in immigration and tourism resulted in an increased number of schistosomiasis in nonendemic areas. Considering this, we are faced with

the uncommon etiology of acute appendicitis.

## Introduction

Schistosomiasis is a granulomatous disease that is prevalent in tropical and subtropical areas. 1 It is transmitted after contact with infested water. Schistosomiasis can infect intestinal or urogenital tracts, and symptoms can vary from abdominal pain to hematuria as per the affected system.1

Most acute intestinal schistosomiasis presents with abdominal pain and bloody diarrhea. There are few cases of atypical presentation, such as intestinal obstruction, peritonitis, and acute appendicitis. 1-3

Despite its prevalence worldwide, schistosomiasis is a rarely encountered presentation, even in the endemic areas.<sup>4</sup> However, the United Arab Emirates is not considered an endemic area for the disease, although it is a multicultural country with immigrants from endemic regions. The clinical manifestation of acute appendicitis represents an extremely uncommon situation, even in endemic areas.

## **Case Description**

A 37-year-old male patient from the Philippines with a medical background of hypertension and gout presented to the emergency department complaining of generalized abdominal pain localizing to the right lower quadrant of 1-day duration. On examination, he was found to be afebrile and hemodynamically stable with positive rebound tenderness in the right lower

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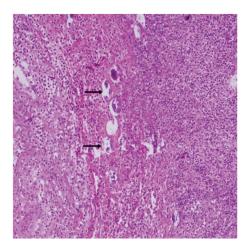
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**Fig. 1** Hematoxylin and eosin stain (x20 magnification) showing nonviable calcified *Schistosoma japonicum* eggs present among acute suppurative inflammation.

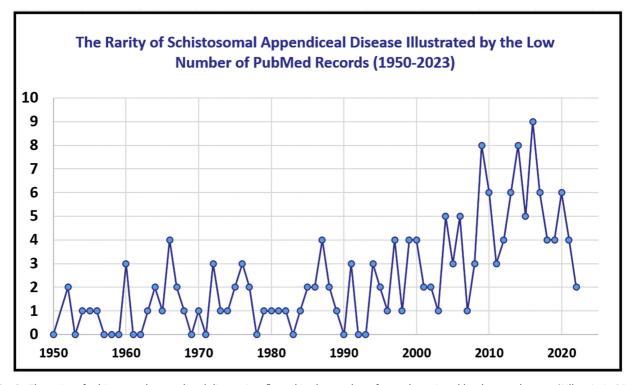
quadrant. Laboratory findings were positive for an increase in the inflammatory markers. His white blood cell count was  $24.13 \times 10^9 / L$  and C-reactive protein of  $103.8 \, mg/L$ . Based on clinical manifestations, the patient was suspected to have acute appendicitis and was taken for surgery. Surgical findings demonstrated an acutely inflamed appendix with early mass formation and adhesions to the abdominal wall. An appendectomy was done, and the patient was discharged after 2 days in stable general condition. Unexpectedly, histopathology demonstrated appendicitis secondary to schistosomial disease (**Fig. 1**). The patient was treated for his schistosomiasis on his return to the clinic.

## **Discussion**

Schistosomiasis is a parasitic infection caused by five different species. *Schistosoma mansoni, japonicum, mekongi,* and *intercalat*e predominantly affect the intestinal tract, whereas *Schistosoma crematorium* mainly affects the urogenital tract. Our cases were from the Philippines, where the most common species is *Schistosoma japonicum*. The disease is transmitted through contact with infested water. The infective cercariae penetrate the skin, entering the venous system to reach the heart. They reside in the liver before entering the portal system to spread to the abdominal viscera. Species, as per their biological preference, will end up infesting the gastrointestinal or urinary tracts and exiting through feces or urine.

Schistosomiasis of the appendix is a well-recognized disease, but it rarely results in acute appendicitis (**Fig. 2**). Turner reported the first case in 1909. The exact pathology resulting in acute appendicitis is not clear, but few theories have been postulated over the years. The most common theory is the activation of the immune response around the ova in the lumen of the appendix, resulting in inflammation and fibrosis, leading to lumen obstruction and appendicitis. Another theory is that an embolic event caused by the ovum leads to ischemia of the serosa and the formation of a perpendicular granulomatous reaction, ending with fibrosis. The exact pathology resulting in acute appendicitis and the formation of a perpendicular granulomatous reaction, ending with fibrosis.

A systematic review of the international literature demonstrated a total prevalence of 1.3%, while stratification by continents revealed a prevalence of 2.8% in Africa compared with 0.5% in the Middle East. By contrast, studies in nonendemic countries reveal a prevalence rate of 0.1 to 0.2%.



**Fig. 2** The rarity of schistosomal appendiceal disease is reflected in the number of records retrieved by the search term: (Bilharziasis OR Schistosomiasis) AND (Appendix OR Appendicitis OR "Appendiceal mass") between 1950 and 1923.

The incidence of acute appendicitis as a result of schistosomiasis is very low in nonendemic regions such as the Middle East. The largest reported case series were from Kuwait, where a 5-year retrospective study was reported. They included 3,012 appendectomies and only 8 cases of schistosomial appendicitis. All of which were expats from endemic areas like our reported case.<sup>8,9</sup> However, the incidence of schistosomial appendicitis in endemic areas is reported to be between 0.02 and 6.3%<sup>10</sup> This represents over quarter (28.6%) of chronic appendicitis.<sup>11</sup>

Histological examination is the gold standard diagnosis of schistosomial appendicitis. Colonic mural calcification on preoperative computed tomography scan can raise suspicion for schistosomal appendicitis, but this is not specific and can be found in other colonic diseases such as sclerosing encapsulating peritonitis, phlebosclerotic chronic ischemic colitis, mucinous adenocarcinoma, and leiomyosarcoma.<sup>12</sup> Once the diagnosis of schistosomial appendicitis has been confirmed, the disease needs to be eradicated by praziquantel in the form of two doses of 20 mg/kg body weight.<sup>13</sup>

## **Conclusions**

Surgeons practicing in nonendemic areas, such as the Middle East, must be aware of the possibility of seeing atypical presentations of parasitic diseases, especially considering the worldwide increase in tourism and immigration.

#### **Patient Consent**

The authors confirm that they had the patient's consent for publication. The full report is based on an anonymized presentation, and the patient's identity is protected.

#### **Authors' Contributions**

All named authors contributed to the clinical care of the patient, the preparation of the case report, and its revision and finalization.

#### **Compliance with Ethical Principles**

No prior ethical approval is required for single cases.

Funding and Sponsorship None.

#### Conflict of Interest

None declared.

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