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Case Report

Acute Schistosomal Colitis Preceded by Katayama Syndrome

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ABSTRACT

Schistosomiasis is prevalent in tropical and subtropical areas. It manifests as an acute or chronic illness caused by the body's reaction to the worms' eggs. In view of its clinical similarity to various other diseases, the disorder may cause diagnostic errors. We present a case of a Sudanese man, who presented with fever, headache, fatigue, myalgia, excessive sweating, abdominal cramps, and a high eosinophil count on blood testing. He was diagnosed with a connective tissue disorder and was started on prednisolone, but 3 weeks later, he presented with rectal bleeding. Colonoscopy showed features of moderate distal colitis. Colonic biopsies revealed several viable schistosome ova associated with aggregates of eosinophils, compatible with active colonic schistosomiasis.

Keywords: *Katayama syndrome, schistosomal colitis, schistosomiasis*

Introduction

Schistosomiasis is a parasitic disease caused by blood flukes (trematodes) of the genus Schistosoma, it is the third most devastating tropical disease in the world after malaria and intestinal helminthiasis.[1]

It can present as acute or chronic illness.

We report a case of schistosomiasis that shows how the acute infection (Katayama syndrome) in an endemic country can be misdiagnosed at the time of presentation, with the diagnosis made only after development of chronic complications of schistosomiasis.

Case Report

A 29-year-old male, engineer, living in Khartoum state, presented with a 1-week history of fever, headache, fatigue, myalgia, excessive sweating, and abdominal cramps. He underwent thorough investigations that showed a high eosinophil count of 4.96 × 10^3 cell/μL, C-reactive protein (CRP) of 287.9 mg/L, erythrocyte sedimentation rate (ESR) of 90 mm/h, serum IgE of 841.5 IU/ml, and a strongly positive A/Scl 70 [Table 1]. He was seen by a rheumatologist who considered him as a case of systemic sclerosis; the patient was admitted to hospital and started on prednisolone 40 mg daily. After 9 days, he was discharged with much improvement clinically and biochemically. In spite of that, he continued to have fatigability, myalgia, and mild abdominal cramps. He consulted a second rheumatologist who suggested the diagnosis of Churg–Strauss syndrome in view of eosinophilia and a history of wheezy chest in childhood. This time he was put on azathioprine 50 mg twice daily orally, in addition to the previously prescribed dose of prednisolone.

One week later, he presented to the gastroenterology department with bright red bleeding per rectum which lasted for 3 days without associated change in bowel habits, fever, or weight loss. He was a nonsmoker and had no family history of inflammatory bowel disease or bowel malignancy. The patient denied any contact with contaminated water. Physical examination was unremarkable. Laboratory investigations revealed a high eosinophil count of 1.5 × 10^3 cell/μL, ESR of 60 mm/h, and CRP of 45 mg/L, and trace protein in urinalysis. He had a normal stool examination with no red blood cells, ova, or parasites, and normal urea and creatinine level [Table 2].

Colonoscopy revealed moderate colitis noted distally with edematous, inflamed mucosa up to mid-transverse colon, inflammation was most severe in the rectum.

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Histopathological examination of the colonic mucosa sections showed normal crypt architecture. Several viable schistosome ova associated with aggregates of eosinophils were noted; features of active colonic schistosomiasis [Figure 1].

He was treated with praziquantel 40 mg/kg, the dose was repeated after 6 weeks, he showed immediate marked clinical improvement and most of his symptoms disappeared within the 1st week. He showed complete resolution of symptoms on follow-up visits. His repeat complete blood count showed normal eosinophil count.

Discussion

Acute schistosomiasis can present as swimmer's itch or as Katayama fever, the latter is a systemic hypersensitivity reaction against the migrating parasites, which occurs between 2 and 8 weeks after exposure.[2,3]

Table 1: Laboratory investigations of the patient on initial presentation with acute febrile illness

Test	Patient values	Normal values
Hemoglobin concentration	15.4	11.0-16.0 mg/dl
White blood cell count	11.45×10^3	$4.0\text{-}11.0\times10^3$ µL
Neutrophils	3.55	$1.8\text{-}6.8\times10^3$ µL
Lymphocytes	2.24	$1.2\text{-}4.9\times10^3$ µL
Platelet count	209×10^3	$150\text{-}450\times10^3$ µL
Eosinophil	4.96×10^3	$0.1\text{-}0\times10^3$ µL
Erythrocyte sedimentation rate	90	5-20 mm/1 h
IgE	841.5	0-100 IU/ml
C-reactive protein	287.9	<5 mg/L
Blood urea	23.9	10-50 mg/dl
Serum creatinine	0.8	0.7-1.2 mg/dl
Liver function test	Normal	-
ANA profile	Strongly positive A/Scl 70	-

ANA=Antinuclear antibody

Table 2: Laboratory investigations of the patient after development of schistosomal colitis

Test	Patient values	Normal values
Hemoglobin concentration	15.2	11.0-16.0 mg/dl
White blood cell count	9.8×10^3	$4.0\text{-}11.0\times10^3$ µL
Neutrophil	5.2	$1.8\text{-}6.8\times10^3$ µL
Lymphocytes	2.8	$1.2\text{-}4.9\times10^3$ µL
Platelet count	270×10^3	$150\text{-}450\times10^3$ µL
Eosinophil	1.5×10^3	$0.1\text{-}0.4\times10^3$ µL
Erythrocyte sedimentation rate	60	5-20 mm/1 h
C-reactive protein	45	<5 mg/L
Blood urea	18	10-50 mg/dl
Serum creatinine	1.2	0.7-1.2 mg/dl
Urine analysis	Trace protein	-
Stool analysis	No ova or parasites	-

Symptoms are more likely to occur in travelers and other nonimmune hosts,[4] these include sudden onset of fever, chills, myalgia, arthralgia, dry cough, diarrhea, and headache, often resembling serum sickness. Lymphadenopathy and hepatosplenomegaly may be prominent findings on physical examination. The symptoms usually resolve spontaneously over a period of a few weeks, but neck stiffness and coma can occur and occasional deaths have been reported related to intense infection.[5] Patients may develop eosinophilia and patchy infiltrates on chest X-ray.[3]

The diagnosis of Katayama fever relies upon appropriate epidemiology for potential exposure and consistent clinical findings. The diagnosis can be complicated by the fact that eggs are rarely excreted in detectable amounts during this stage and antibody tests are usually negative. However, a peripheral eosinophilia will usually be present and often serves as an important clue.[6]

Due to the lack of clinical trials in nonimmune populations, the optimal treatment regimen for Katayama syndrome is not known.[7] Praziquantel is not particularly effective in early infection, so therapy for Katayama fever is mostly supportive. Glucocorticoids (e.g., prednisolone 40 mg daily for 5 days) can be considered to ameliorate significant symptoms related to hypersensitivity.[7,8]

The optimal timing of praziquantel therapy is unclear; if it is not instituted immediately, it should be administered within 6–10 weeks when the infection is established and adult worms have developed. An alternative approach is to commence praziquantel at the time of diagnosis as this may decrease the worm load and reduce the risk of ectopic localization.[4]

The pathology of chronic schistosomiasis, which is far more common than the acute form of the infection, results from egg-induced immune response, granuloma formation, and associated fibrotic changes.

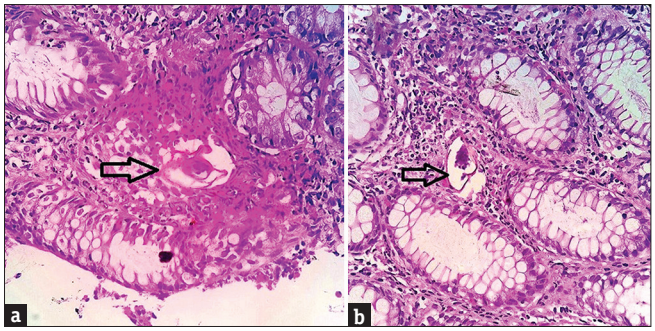

Figure 1: (a and b) Two histology slides of the colonic mucosa of a 29-year-old male with distal colitis due to *Schistosoma mansoni* infection. In the center of the field (arrow) is a viable Schistosoma mansoni egg (intact miracidial nuclei) in the lamina propria associated with aggregates of eosinophils

The symptoms of colonic schistosomiasis are nonspecific and may mimic other gastrointestinal problems.[9] Severe complications have been reported with schistosomiasis such as hemorrhagic diarrhea, obstruction secondary to an inflammatory mass, acute appendicitis, intestinal intussusception arising from a mucocele of the appendix, and ischemic colitis.[10-14]

For a patient coming from an endemic country with a suspected light infection, the reasonable approach for diagnosis would be serum antibody titer or polymerase chain reaction assay.[6] Other investigations include serologic tests for the detection of the parasite antigens.[15] Another means involve the demonstration of parasite eggs in the stool by microscopic examination (Kato-Katz smear).[16]

The treatment of choice for all schistosome species is praziquantel.[17,18]

As maturing schistosomes are less susceptible to therapy than adult worms, a second course of treatment is necessary. This is given several weeks after the first course of therapy.[19]

This patient presented first with an acute illness, Katayama syndrome, which was misdiagnosed as connective tissue disease owing to the nonspecific clinical presentation. Patients with acute *Schistosoma mansoni* infection have been diagnosed at times as having typhoid fever, hepatitis, pancreatitis, and appendicitis, only to be cured when infection with schistosomiasis has been discovered after a long search.[14] In this case, the patient's illness was complicated by acute colonic schistosomiasis, confirmed by the finding of viable ova on histopathological examination of colonic mucosa specimens. The absence of ova in stools does not rule out schistosomiasis since it has been estimated that thousands of eggs per day must be excreted to be readily visualized on routine microscopic examination of stools.[20]

Conclusion

Compared to chronic schistosomiasis, Katayama syndrome is rather an uncommon acute presentation of the disease, especially in endemic areas. Clinical symptoms and laboratory tests are nonspecific, so the diagnosis could be missed till the time of appearance of features of specific organ involvement. Colonic mucosa is one of the common sites of ova deposition, giving rise to colonic schistosomiasis, again with nonspecific clinical symptoms and laboratory tests, as well as endoscopic features, but only in conjunction with the histopathological findings, the correct diagnosis can be revealed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Schistosomiasis, Fact Sheet No 115; February, 2010. World Health Organization. Available from: http://www.who.int/mediacentre/factsheets/fs115/en/. [Last accessed on 2016 Apr 20].
2. Gryseels B, Polman K, Clerinx J, Kestens L. Human schistosomiasis. Lancet 2006;368:1106-18.
3. Ross AG, Vickers D, Olds GR, Shah SM, McManus DP. Katayama syndrome. Lancet Infect Dis 2007;7:218-24.
4. Bottieau E, Clerinx J, de Vega MR, Van den Enden E, Colebunders R, Van Esbroeck M, *et al.* Imported Katayama fever: Clinical and biological features at presentation and during treatment. J Infect 2006;52:339-45.
5. Hoeffler DF. Cercarial dermatitis. Arch Environ Health 1974;29:225-9.
6. Gundersen SG, Ravn J, Haagensen I. Early detection of circulating anodic antigen (CAA) in a case of acute *Schistosomiasis mansoni* with Katayama fever. Scand J Infect Dis 1992;24:549-52.
7. Harries AD, Cook GC. Acute schistosomiasis (Katayama fever): Clinical deterioration after chemotherapy. J Infect 1987;14:159-61.
8. Chapman PJ, Wilkinson PR, Davidson RN. Acute schistosomiasis (Katayama fever) among British air crew. BMJ 1988;297:1101.
9. Mohamed AR, al Karawi M, Yasawy MI. Schistosomal colonic disease. Gut 1990;31:439-42.
10. van Leerdam ME, Dingemans-Dumas AM, Boldewijn JK, Dees A. A man from Surinam with haemorrhagic diarrhoea after long-standing schistosomiasis. Ned Tijdschr Geneeskd 2004;148:1928-30.
11. Lamyman MJ, Noble DJ, Narang S, Dehalvi N. Small bowel obstruction secondary to intestinal schistosomiasis. Trans R Soc Trop Med Hyg 2006;100:885-7.
12. Gabbi C, Bertolotti M, Iori R, Rivasi F, Stanzani C, Maurantonio M, *et al.* Acute abdomen associated with schistosomiasis of the appendix. Dig Dis Sci 2006;51:215-7.
13. Wong MT, Goh L, Chia KH. Intestinal schistosomiasis manifesting as colonic intussusception arising from a mucocele of the appendix: Report of a case. Surg Today 2008;38:664-7.
14. Neves J, Raso P, Pinto Dde M, da Silva SP, Alvarenga RJ. Ischaemic colitis (necrotizing colitis, pseudomembranous colitis) in acute *Schistosomiasis mansoni*: Report of two cases. Trans R Soc Trop Med Hyg 1993;87:449-52.
15. Sandoval N, Siles-Lucas M, Pérez-Arellano JL, Carranza C, Puente S, López-Abán J, *et al.* A new PCR-based approach for the specific amplification of DNA from different *Schistosoma* species applicable to human urine samples. Parasitology 2006;133:581-7.
16. King C, Mahmouud AA. Schistosomiasis. In: Guerrant R, Walker DH, Weller PF, editros. Tropical Infectious Diseases: Principles, Pathogens and Practice. Vol. 2. Philadelphia: Churchill Livingstone; 1999. p.1031.
17. Ross AG, Bartley PB, Sleigh AC, Olds GR, Li Y, Williams GM, *et al.* Schistosomiasis. N Engl J Med 2002;346:1212-20.
18. Danso-Appiah A, Olliaro PL, Donegan S, Sinclair D, Utzinger J. Drugs for treating *Schistosoma mansoni* infection. Cochrane Database Syst Rev 2013;(2):CD000528.
19. Fenwick A, Rollinson D, Southgate V. Implementation of human schistosomiasis control: Challenges and prospects. Adv Parasitol 2006;61:567-622.
20. Elliott DE. Schistosomiasis. Pathophysiology, diagnosis, and treatment. Gastroenterol Clin North Am 1996;25:599-625.