<u>study</u>

4 ABSTRACT

5 Aim: Schistosomiasis is a water-borne trematode infestation and is one of the most widespread

6 parasitic diseases in the world. Schistosomiasis can affect any organ. This case report highlights

7 the importance of awareness of unusual cause of common surgical presentations of

8 schistosomiasis in endemic areas and the need for appendectomy specimens to be sent for

9 histological review so that patients can benefit from full investigations and specific antiparasitic

- 10 treatment.
- 11 Case Presentation: In this paper, the authors present a case of appendicular schistosomiasis
- 12 that was incidentally discovered in a 33-year-old female patient from Lagos, Nigeria who

13 underwent appendectomy for acute appendicitis. Appendectomy specimens removed from the

14 patient appeared macroscopically normal but histopathological analysis revealed schistosomal

15 eggs confirming the diagnosis of appendicular schistosomiasis.

16 Discussion: Confirmation of appendicular schistosomiasis is a purely histological diagnosis,

because there are no pathognomonic clinical or operative findings. Physicians practising in the

18 endemic areas must be aware of the possibility of seeing atypical presentations of parasitic

19 diseases.

20 Conclusion: The authors therefore strongly recommend that all appendectomy specimens

- should be sent for histological review so that patients can benefit from full investigations and
- 22 specific anti-parasitic treatment.

23 Key Words: Appendicular schistosomiasis, Nigeria, Schistosomiasis, Appendicitis.

24 INTRODUCTION

25 Schistosomiasis is endemic in many parts of the world with highest burden in South Asia and Sub-

26 Sahara Africa (1–3). In Nigeria, *Schistosomiasis* is endemic and causes intestinal Schistosomiasis which

27 affects various internal organs. Schistosomal ova are frequently found in the appendix of patients

suffering from schistosomiasis. The prevalence of appendicular Schistosomiasis is high in endemic areas

29 compared to developed countries (2). In a prospective study carried out in Nigeria over a 5 year period,

- **30** 518 consecutive appendices were removed at Surgery for symptomatic appendicitis examined
- histologically, of these, 32 (6.2%) appendices showed evidence of chronic Schistosoma Haematobium
- appendicitis (4). There are two types and pathogenic pathways of appendicular schistosomiasis. First,
 'granulomatous acute appendicitis' is caused by an immunological granulomatous reaction to newly
- 35 granulomatous acute appendicitis is caused by an immunological granulomatous reaction to newly 24 deposited over with tissue percess and tissue accinential in the infection. I a with infection is a with infection of a with the infection of a with infection of a with the infection.
- 34 deposited ova, with tissue necrosis and tissue eosinophilia; it may occur early in the infection, i.e., within
- 35 weeks. Second, 'obstructive acute appendicitis' is caused by long-standing inflammation and fibrosis
- around dead eggs, leading to obstruction of the appendiceal lumen and increasing the risk of infection
- 37 from faecal contaminants; this may occur in the late phase after several months or years (5,6).

38 Confirmation of appendicular schistosomiasis is a histological diagnosis, because there are no 39 pathognomonic clinical or operative findings. Schistosomiasis in endemic areas is a major public health 40 problem. Specific drug treatment involves the use of antihelmintics. Praziguantel is the most commonly 41 used for treating both urinary and gastrointestinal forms of the disease and is administered as a single 42 dose (7). Most patients with schistosomiasis experience gradual improvement following treatment 43 depending on the severity of their presenting symptom. If symptoms persist after 2 rounds of Praziguantel treatment, more urine or stool should be taken and tested for viable eggs and re- treatment may be given 44 if persistent infection is detected (7). Physicians practising in endemic areas must be aware of the 45 46 possibility of seeing atypical presentations of parasitic diseases. The authors present a case of acute 47 appendicitis caused by schistosomiasis, a rare effect of the parasitic infection in a 33 years old Nigerian 48 woman.

49

50 CASE REPORT

A 33 year-old female Lagos resident originally from a rural village in Osun State, Nigeria, presented at Subol Hospital Limited, Lagos, Nigeria with complains of intense abdominal pain that had started five days earlier. This pain had started suddenly, initially periumbilical, with radiation to the right iliac fossa. There was a distinct history of anorexia and nausea but no associated vomiting and fever. The patient had no obvious urinary symptoms or any obvious signs of constipation, diarrhea, melena, hematochezia, and hematuria. Her Last menstrual period was 3 weeks prior her presentation. She was not a known patient with Hypertension or Diabetes

On examination she was noted to be afebrile, not pale and hemodynamically stable with a pulse rate of 58 59 84b/m and blood pressure of 110/60mmHg. Her abdomen was not distended, with localized tenderness in the right iliac fossa. There was no obvious organomegaly. Other systems were essentially normal. Blood 60 laboratory results revealed a packed cell volume of 35%, a white cell count of 11000/mm³ (70% 61 neutrophils, 25% lymphocytes, 3% eosinophils), and a negative urinalysis. Pelvic ultrasound revealed a 62 non-gravid uterus, normal ovary, both adnexae are free. A diagnosis of acute appendicitis was made and 63 64 the patient was prepared for appendectomy. During appendectomy, she was noted to have an acutely 65 inflamed appendix with no perforation, clear peritoneal fluid and essentially normal bowels. The patient 66 made an uneventful post-operative recovery and was discharged after three days.

67 She was seen in the outpatient clinic 2 weeks post operation with the Histology report. She had recovered 68 without any sequels. Macroscopic examination of the appendix specimen showed an appendix which 69 measured 8.5cm in length and 0.5cm in maximum diameter. The serosa surface was smooth with prominent vascular markings and devoid of exudates. The cut section revealed patent lumen. 70 71 Histological sections showed an appendix with mucosa exhibiting hyperplastic lymphoid follicles with 72 tingible body macrophages (FIGURE 1). The submucosal revealed numerous ova of schistosoma 73 haematobium most of which were calcified (FIGURE 2). They were surrounded by dense fibrosis. The 74 muscularis propria was devoid of neutrophils transmigration (FIGURE 3). Histological diagnosis of 75 appendicular schistosomiasis was made. After histological report, the patient received a single dose of 76 praziguantel (40 mg/kg), which was well tolerated.

77 DISCUSSION

In endemic areas, schistosomiasis is a major public health problem. Schistosomiasis is the second most prevalent parasitic disease worldwide. More than 200 million people are infected, 120 million are symptomatic, and 20 million suffer from severe disease. An estimated 85% of all cases, and virtually all of the most severe, are concentrated in African countries (1,5,8).

Appendicitis is a common cause of acute abdomen in developing countries (9–11). Schistosomiasis as accountable for acute appendicitis are reported between 0.02%–6.3%, representing 28.6% of chronic appendicitis in endemic areas (3,4,6,12,13). Schistosomiasis of the appendix was first described by Turner in 1909 (1), and has been reported in endemic areas. The most usual organisms are Schistosoma
 haematobium and *S. mansoni* (5,7,14,15).

The actual role of infestation plays a significant factor to the development of appendicitis is still confusing and has been much controversy. The characteristic pathological tissue response is believed to be a granulomatous inflammatory reaction to the schistosomal ova, with the lesion predominantly in the submucosa and serosa. There is a formation of epitheloid cell granulomas that ultimately undergo fibrosis. Intramuscular oviposition (submucosa) causes an obstructive type of appendicitis with a greater risk of perforation. Serosal involvement causes inflammation and the formation of adhesions (3,5,6,16,17).

- 94 Confirmation of appendicular schistosomiasis is a purely histological diagnosis, because there are no 95 pathognomonic clinical or operative findings (5,6,15). Physicians practising in the endemic areas must be 96 aware of the possibility of seeing atypical presentations of parasitic diseases.
- 97 Praziquantel is the drug of choice to treat all forms of schistosomiasis. This is active against mature 98 worms. Although resistance to the drug is suspected, drug can still be used reliably at 40 to 60 mg/kg as

99 a single dose in most of the cases. Repeated dosages of praziguantel might be necessary in early stages

100 of the disease and to treat long-standing infections. Abdominal discomfort is the most frequently reported

101 side effect of this well-tolerated drug (7,14,18,19).

102 CONCLUSION

This case report highlights the importance of awareness of unusual causes of common surgical presentations of schistosomiasis in endemic areas. It also shows that intestinal schistosomiasis can cause appendicitis, therefore appendectomy specimens should be sent for histological review so that patients can benefit from full investigations and specific antiparasitic treatment. Thus, a clear communication between surgeons and pathologists for management of patients with suspected appendicitis is required for proper management.

109 COMPETING INTEREST

- 110 There is no competing interest
- 111 ETHICAL APPROVAL
- 112 It is not applicable
- 113 CONSENT
- 114 Patient signed a written informed consent
- 115 **REFERENCES:**

- Mazigo HD, Giiti GC, Zinga M, Heukelbach J, Rambau P. Schistosomal peritonitis secondary to perforated appendicitis. Brazilian J Infect Dis [Internet]. Elsevier; 14(6):628– 30. Available from: http://dx.doi.org/10.1016/S1413-8670(10)70122-7
- Salih MA. A case of acute appendicitis due to intestinal schistosomiasis. Ann Med Surg
 [Internet]. Elsevier; 2019;37(October 2018):1–3. Available from: https://doi.org/10.1016/j.amsu.2018.11.015
- Badmos KB, Komolafe AO, Akintola L, Rotimi O. Schistosomiasis presenting as acute appendicitis. East Afr Med J. 2006;83:528–32.
- 4. Duvie S.O, Diffang C, Guirguis M.N. The effects of Schistosoma haematobium infestation on the vermiform appendix: the Nigerian experience. J Trop Med Hyg. 1987;90(1):13–8.
- Satti MB, Tamimi DM, Sohaibani MOAL, Quorain AAL. Appendicular schistosomiasis : a cause of clinical acute appendicitis ? 2014;(May).
- Boudier B, Parola P, Dales JP, Linzberger N, Brouqui P, Delmont J. Schistosomiasis as an unusual cause of appendicitis. Clin Microbiol Infect [Internet]. European Society of Clinical Infectious Diseases; 2004;10(2):89–91. Available from: http://dx.doi.org/10.1111/j.1469-0691.2004.00805.x
- 132 7. Madavo C, Hurriez H. Schistosomiasis of the appendix. 2006;
- Vennervald BJ, Dunne DW. Morbidity in schistosomiasis: an update. Curr Opin Infect Dis.
 2004;17(5):439–47.
- Kong VY, Bulajic B, Allorto NL. Acute appendicitis in a developing country. World j Surg.
 2012;36:2068–73.
- Fashina IB, Adesanya AA, Atoyebi OA, Osinowo OO, Atimomo CJ. Acute appendicitis in Lagos: a review of 250 cases. Niger Postgr Med J. 2009;16(4):268–73.
- 139 11. Mangete ED, Kombo BB. Acute appendicitis in Port Harcourt Nigeria. Orient J Med.
 2004;16:1–3.
- 141 12. Gali BM, Nggada HA, Eni EU. Schistosomiasis of the appendix in Maiduguri. 2006;
 142 36:162-3. Trop Dr. 2006;36:162–3.
- 143 13. Helmy A.H, Shousha T.A, Magdi M, Sabri T. "Appendicitis; appendicectomy and the value of endemic parasitic infestation." Egypt J Surg. 2000;19(2):87–91.
- Olveda DU, Li Y, Olveda RM, Lam AK, Mcmanus DP, Chau TNP, et al. International Journal of Infectious Diseases Bilharzia in the Philippines : past , present , and future. Int J Infect Dis [Internet]. International Society for Infectious Diseases; 2014;18:52–6.
 Available from: http://dx.doi.org/10.1016/j.ijid.2013.09.011

- Cenarruzabeitia D, Landolfi S, Carrasco MA. Case Report Intestinal Schistosomiasis as
 Unusual Aetiology for Acute Appendicitis , Nowadays a Rising Disease in Western
 Countries. 2012;2012.
- 152 16. Nandipati K, Parithivel V, Niazi M. Schistosomiasis: a rare case of acute appendicitis in 153 the African American population in the United States. Am Surg. 2008;74:221–3.
- 17. Terada T. Schistosomal appendicitis: Incidence in Japan and a case report. World J
 Gastroenterol. 2009;15:1648–9.
- 156 18. Cox N.D, Yates P.J. Schistosomiasis : a rare cause of acute appendicitis. JSCR.
 157 2010;4(4):3–5.
- 158
 19. Limaiem F, Bouraoui S, Bouhamed M. Schistosomiasis : A rare cause of acute appendicitis. 2017; (January 2015).

160



161 162

Figure 1: Histopathologic examination of the mucosa with hyperplastic lymphoid follicles and underlying extensive submucosa fibrosis with numerous Schistosoma haemantobium ova (H and E, ×10)

165



166

- 167 Figure 2: Histopathologic examination of the submucosa, muscularis propria and serosal surfaces of the
- appendix showing extensive submucosa fibrosis with Showers of Schistosoma haemantobium ova as well as congested serosa (arrows) (H and E, ×10)



- 175 Figure 3: Histopathologic examination of the submucosa of the appendix showing chronic inflammation,
- 176 fibrosis with embedded Schistosoma ova and vascular channels (H and E, ×20)