

Schistosomal appendicitis in Kuwait A5-year study

Fawaz Abo-Alhassan (B.Sc., B.M.B.CH)^{a,*}, Fatemah Faras (B.Sc., M.D.)^b,
Yousef M. Malek (B.Sc., B.M.B.CH)^c,
Munish Joneja (MD, DNB) (Consultant Histopathology)^d,
Piyaray M. Dhar (MBBS, MS, FRCS(I)) (Consultant General Surgery)^e

^a Department of Surgery, Al-Adan Hospital, Ministry of Health, State of Kuwait, P.O. Box 12244, Kuwait

^b Department of ENT, Zain and Al-Sabah Hospitals, Ministry of Health, State of Kuwait, Kuwait

^c Department of Surgery, Al-Adan Hospital, Ministry of Health, State of Kuwait, Kuwait

^d RNMLC Yiacco Medical Co., Al-Adan Hospital, Ministry of Health, State of Kuwait, Kuwait

^e Department of Surgery, Al-Adan Hospital, Ministry of Health, State of Kuwait, Kuwait

ARTICLE INFO

Article history:

Received 21 September 2016

Accepted 10 October 2016

Available online 17 October 2016

Keywords:

Schistosomal appendicitis

Schistosomiasis

Kuwait

Surgery

Acute appendicitis

ABSTRACT

BACKGROUND: Appendicular schistosomiasis is an unusual etiology of acute appendicitis, which has been reported in countries endemic in schistosomiasis, such as sub-saharan Africa and South America. Nowadays, due to globalization, this disease has been diagnosed in non-endemic countries. Kuwait is a country possessing a larger percentage of foreigners than national citizens. Therefore, several cases of schistosomal appendicitis were found.

METHOD: The clinicopathological records of all patients that underwent appendectomy during January 2007 and December 2011 were recorded from the archives of Al-Adan Hospital in Kuwait. All cases of schistosomal appendicitis were retrieved and the histopathologic slides reconfirmed by the histopathologist.

RESULTS: During the 5-year study period, 3012 appendectomies were performed and 8 schistosomal appendicitis were found. They were all Egyptian males that were admitted for a clinical suspicion of acute appendicitis. The age ranged between 24 and 42 years, with a mean age of 32.75 years. All cases showed histological features of acute or acute suppurative inflammation, with ova seen in the vasculature of all layers of appendicular wall.

CONCLUSION: Although schistosomiasis is a rare causative agent of acute appendicitis, this however can't be confirmed until histological evaluation. Therefore, adequate follow up postoperatively is necessary to insure eradication of the disease and to prevent further serious consequences.

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1. Introduction

Schistosomiasis is a very common tropical disease, endemic in Africa. It is a parasitic infection caused by a trematode, and transmitted by contact with infested waters. According to the WHO, schistosomiasis transmission has been reported from 78 countries. In 2012, 42.1 million have been treated from this disease [1]. It is rarely found in countries that have clean water supply. Schistosomiasis has a very long lifespan, reaching up to 30 years [2]. Because of this reason and globalization, schistosomiasis has reached non-endemic countries.

The two forms of schistosomiasis are urogenital and intestinal. Individuals with the intestinal form can present with a wide range of symptoms as this disease can affect any part of the gastrointestinal (GI) tract. However, the appendix is one of the rarely affected organs. Patients with appendicular schistosomiasis can present with a typical picture of acute appendicitis, and only histopathologic review of the appendix can reach the diagnosis.

The objective of this study is to report appendicular schistosomiasis in a non-endemic country, discuss the clinicopathological findings and to review the literature.

2. Materials and method

This retrospective study was conducted in Al-Adan Hospital in Kuwait. This hospital serves the southern part of Kuwait, which includes the health needs of a big proportion of expatriates. The study was conducted over five years between January 2007 and December 2011. All histopathologic reports of excised appendixes,

* Corresponding author.

E-mail addresses: drfawaz86@gmail.com, fawazow@hotmail.com

(F. Abo-Alhassan), falfaras@gmail.com (F. Faras), dr.yousef89@hotmail.com

(Y.M. Malek), munishjoneja@yahoo.com (M. Joneja), Pmd49@hotmail.com

(P.M. Dhar).

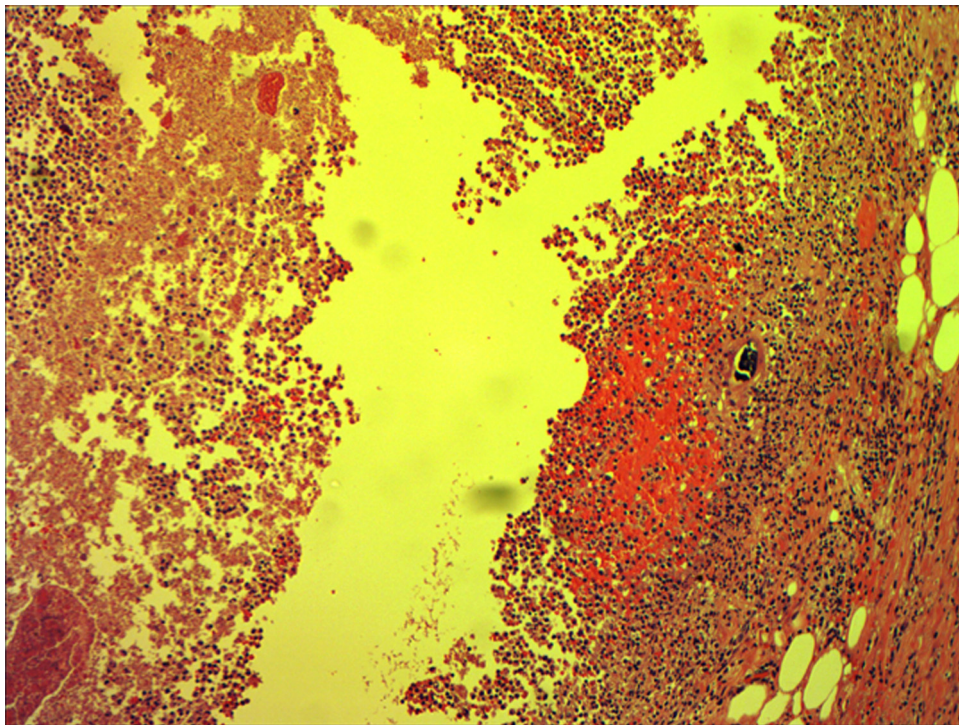


Fig. 1. Microphotograph showing acute suppurative appendicitis with schistosomal egg (H&E 40 $\times$).

and their corresponding files, were retrieved and reviewed. 3012 appendectomies were done during this period, and 8 cases of appendicular schistosomiasis were found. The specimens were retrieved and re-examined by our histopathologist. Detailed clinical data, including history, examination, investigation, and follow up were obtained. These data in addition to histopathologic images are presented in tables and figures respectively.

3. Results

During the 5-year study period, 3012 appendectomies were performed because of clinical suspicion of acute appendicitis. Table 1 summarizes the clinicopathological data. Out of the total number, 8 cases were diagnosed as schistosomal appendicitis. All cases were Egyptian males, and no females were found. The age group ranged between 24 and 42 years (Mean: 32.75 years).

All patients complained of right iliac fossa pain. Other associated symptoms included vomiting in 6 (75%), fever in 2 (25%), and dysuria in 1 (12.5%). The duration of symptoms was 1 day in 7 cases, and 2 days in one case only. However 1 case reported to have similar symptoms 2 months ago. One patient had a previous history of schistosomiasis, and another one gave a history of bladder stones. On examination, all cases demonstrated right iliac fossa tenderness.

Preoperative blood investigation are shown in Table 1. Leukocytosis is defined as a total white blood cell count $>10 \times 10^9/L$. All patient had leukocytosis. As for the differential white cell count, it demonstrated eosinophilia in one case, and neutrophilia in four cases.

All patients were diagnosed preoperatively as acute appendicitis. Half of the cases underwent emergency laparoscopic appendectomy, and the rest underwent an emergency open appendectomy. Intraoperatively, three cases had appendicular masses. Postoperative period was uneventful in all except one that had a short period of low-grade fever, which resolved conservatively.

Grossly three specimens showed fibrino-purulent exudates on the surface, whereas 4 specimens showed pus within the lumen (Fig. 1). All cases had morphological features of acute or acute-

suppurative inflammation (Fig. 1). One showed features of healing with submucosal fibrosis in addition to fibrosis around the ova with only focal mild inflammation (Fig. 2). Three cases showed granulomatous immune response with giant cell formation (Figs. 3–5). Two cases morphologically showed prominent concentric fibrosis around ova (Fig. 2).

The ova were prominently calcified with degenerative changes and were invariably seen in vasculature of all layers of the appendicular wall. Two cases revealed heavy parasitic colonization in the wall (Figs. 6 and 7). Infiltration by eosinophils was not a prominent feature. Only one case, however, showed associated considerable infiltration by eosinophils. Further typing of particular species was not attempted as ova appeared to be mineralized, suggesting that suppurative inflammation was a secondary bacterial infection following luminal obstruction or following ischemia.

4. Discussion

Schistosomiasis is one of the most widely spread parasitic infections. It is mostly endemic in sub-saharan Africa and South America [3]. There are 5 known species of schistosome; *schistosoma mansoni* (SM), *haematobium* (SH), *japonicum*, *mekongi* and *guineensis*. SH causes urinary schistosomiasis while the rest have a predilection to the GI tract.

It is important to note that Kuwait's population is composed of nearly 2 thirds non-Kuwaitis, including immigrants and expatriates. Additionally, Kuwait has no rivers or lakes, and its water system is clean. According to WHO 2012, Kuwait is a non-endemic country of schistosomiasis. Therefore considering the long lifespan of the parasite, it is wise to conclude that globalization has caused this disease to reach this part of the world, months or years after migration. It was not surprising to have all 8 cases being Egyptians, as Egypt is known to be endemic for schistosomiasis, and a big proportion of Kuwait's population is composed of Egyptians.

In this study, schistosomal appendicitis occurred in 0.0027% of patients with acute appendicitis. This is comparable to the incidence found in a similar study done in USA (0.0018%). However,

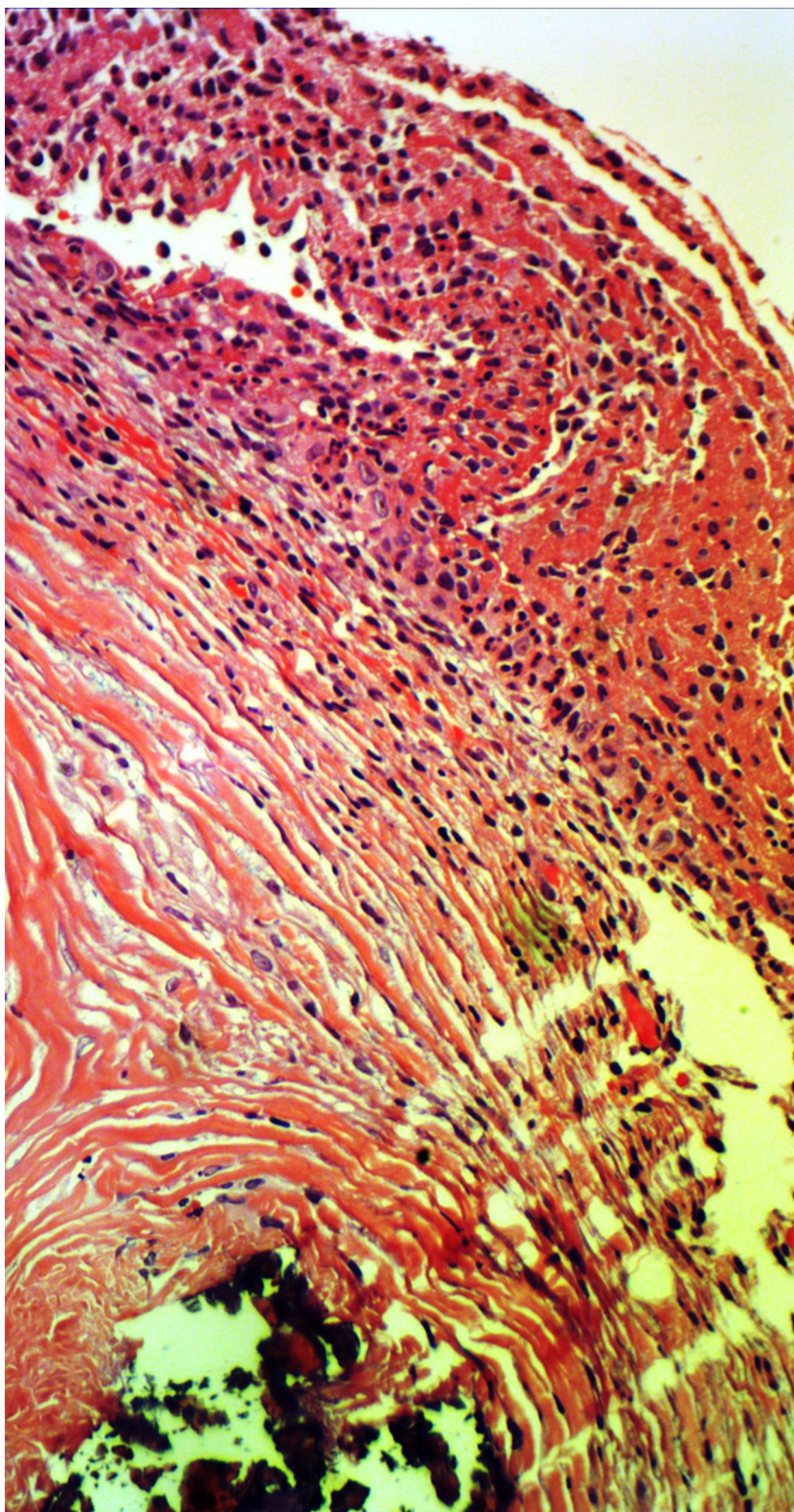


Fig. 2. Microphotograph showing *peri-fibrotic histiocytic granulomatous reaction* (H&E 200 $\times$).

our incidence is lower than Saudi Arabia, which was found to be 1.5% [4] and 1.3% [5] from two different regions. In an endemic country like Nigeria, the incidence was found to be 2.1% [6].

All patients presented with signs and symptoms suggestive of acute appendicitis, similar to other reported cases. Even the duration of symptoms was similar to that of an acute appendicitis.

Therefore, it is difficult to differentiate simple acute appendicitis from schistosomal appendicitis. It is a pure histological diagnosis, with no gold standard preoperative investigative tool. Even serological testing of schistosomal antibodies will not be able to differentiate past from ongoing infections. It might however be of a value in an endemic area were it can be used as a screening tool.

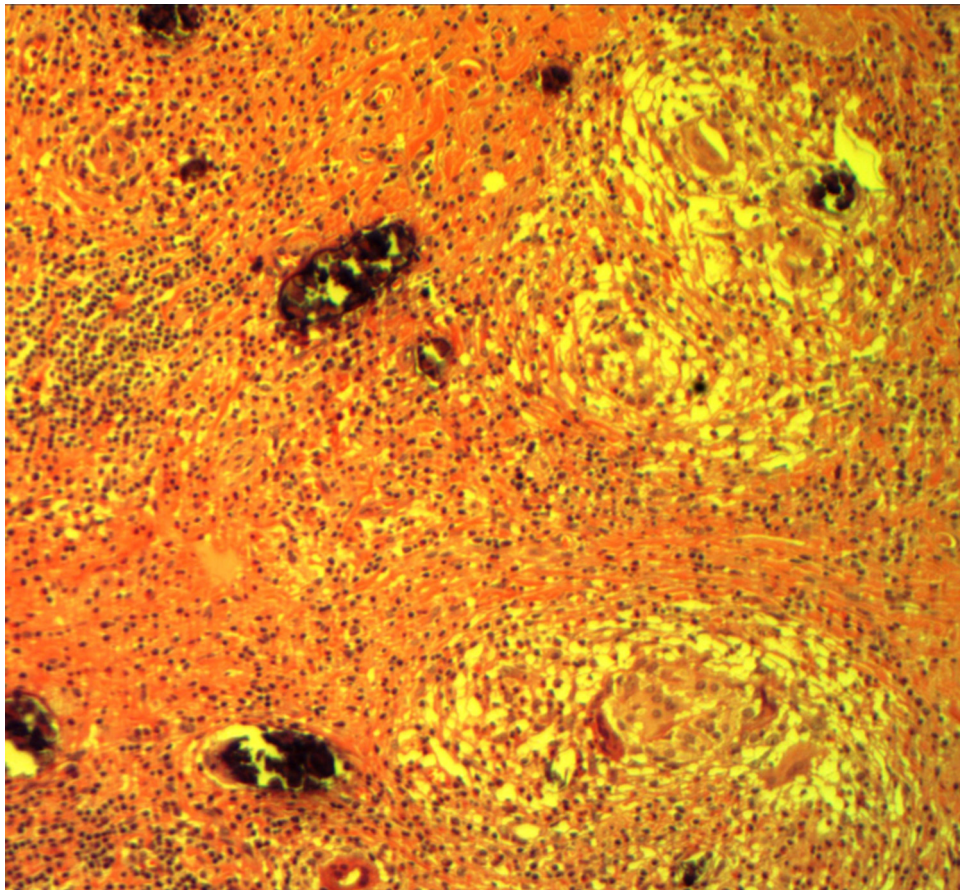


Fig. 3. Microphotograph showing granulomatous reaction around schistosomal eggs (H&E 100×).

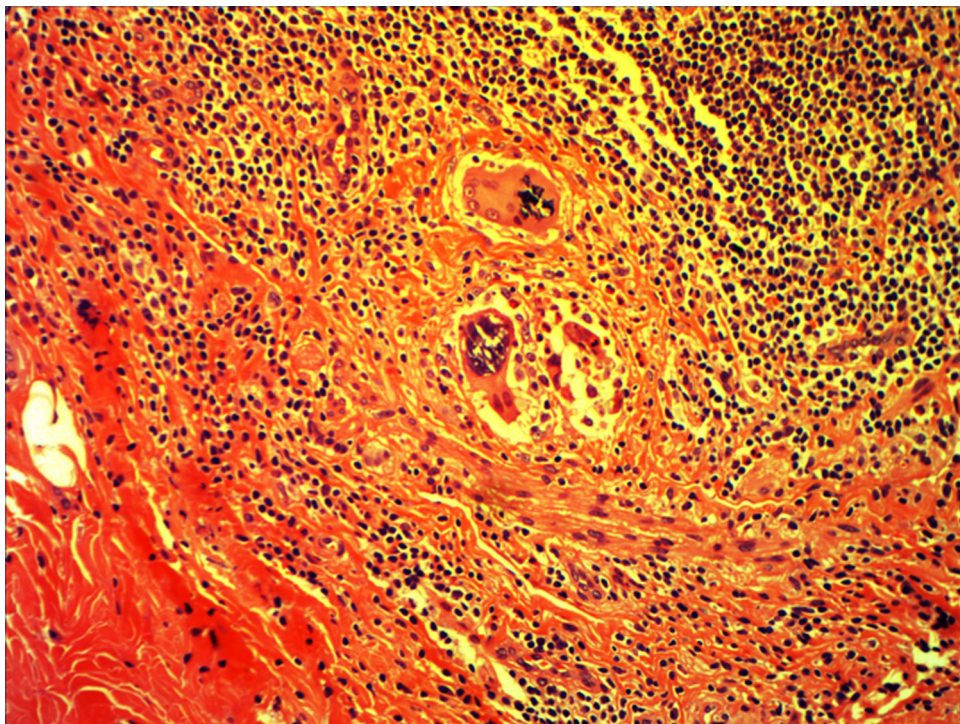


Fig. 4. Microphotograph showing foreign body giant cell engulfing the calcified ova of schistosoma (H&E 200×).

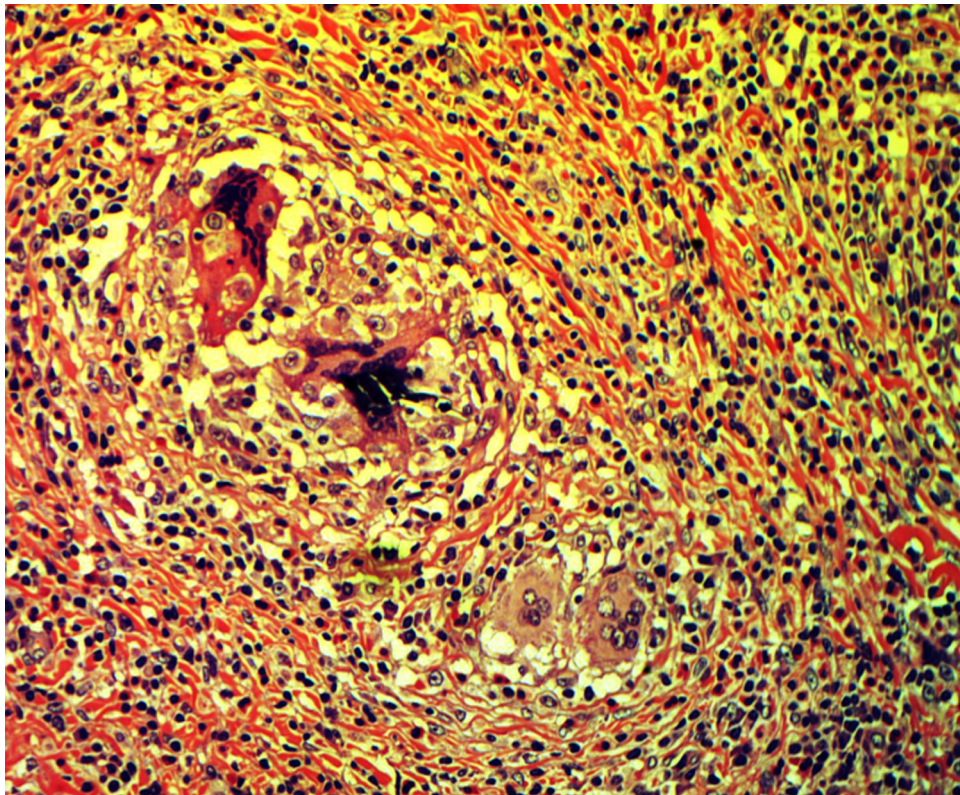


Fig. 5. microphotograph showing a foreign body granulomatous reaction.

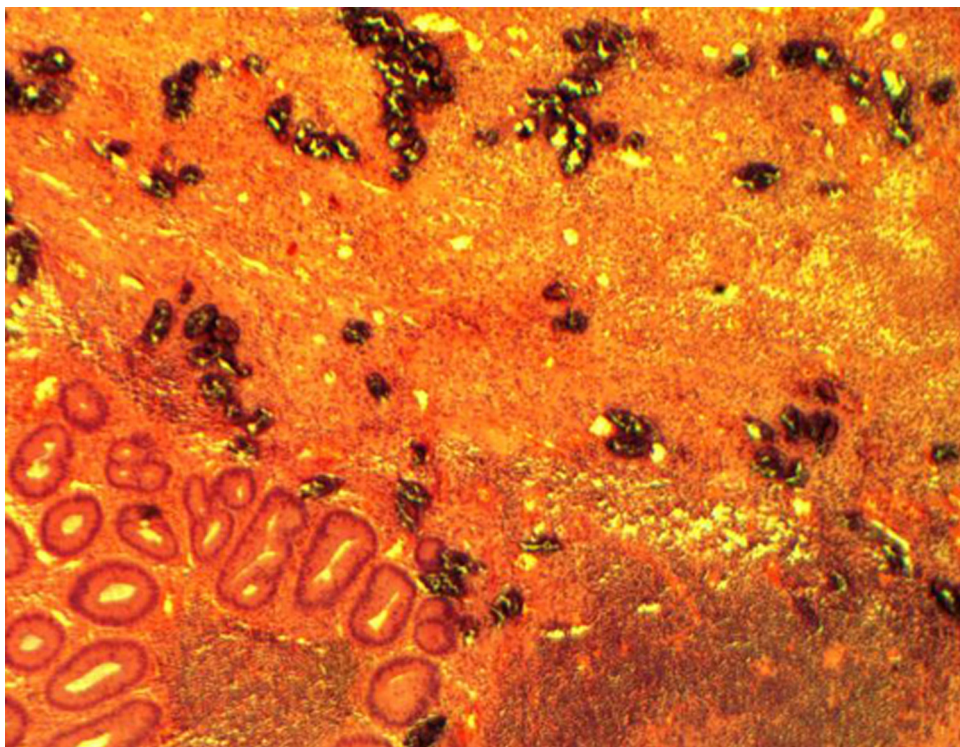


Fig. 6. Microphotograph of the appendix showing numerous schistosomal calcified eggs throughout the wall (H&E 40×).

Patients who are found to have a positive serology with no history of treatment should be assumed to have schistosomiasis and treated accordingly [1]. Nevertheless, family members of infected individuals, even if asymptomatic, should be screened and treated.

Several hypotheses have been adopted. One of the theories explains that the egg emboli causes ischemic changes in addition to the hemorrhagic damage while penetrating the full thickness of the appendiceal wall. Not only does this process decrease the

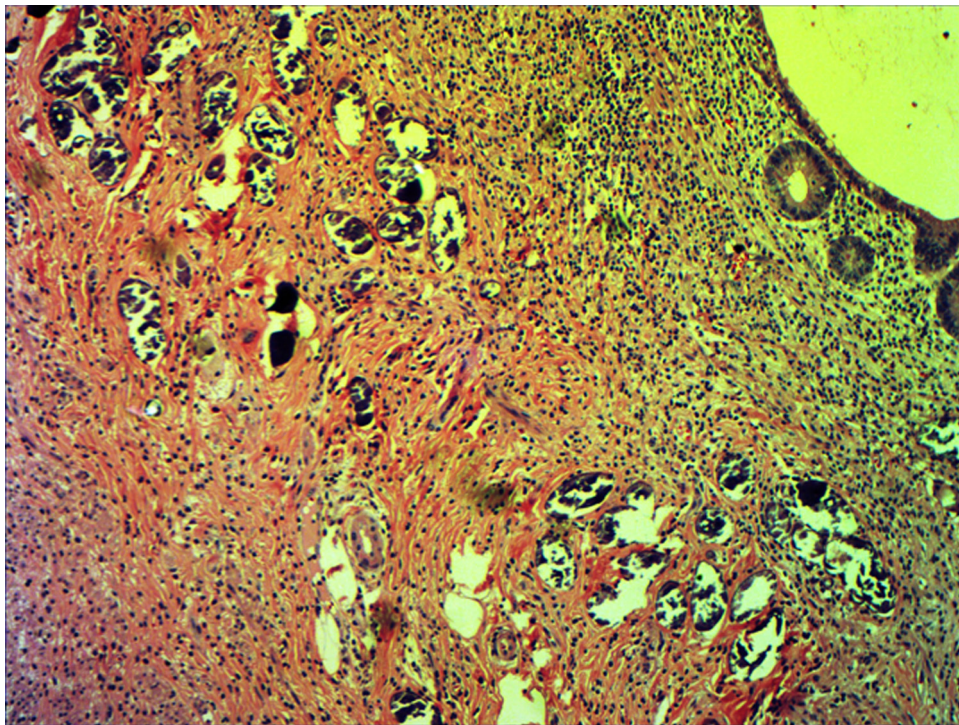


Fig. 7. Microphotograph showing abundant schistosomal calcified eggs in submucosa (H&E 100×).

Table 1

Summarizes the clinicopathological data of the patients diagnosed with schistosomal appendicitis between the period of 2007 to 2011.

Initials	Gender	Age	Duration	fever	Symptoms	Total WBC	Eosinophils	neutrophils	Surgery	Histology
S.A	M	28	2 days	negative	RIF, vomiting, no urinary symptoms, no diarrhea	11.07	0.63 (5.7%)	6.34 (57.3%)	Lap Appendectomy	acute appendicitis
R.B	M	24	1 day	negative	RIF pain, no vomiting,	13.32	1.29 (9.7%)	8.07 (60.6%)	Lap Appendectomy	acute suppurative appendicitis
A.Z	M	40	1 day	negative	RIF pain, vomiting, no urinary symptoms	13.4	0.04 (0.3%)	11.27 (84%)	Lap Appendectomy	acute suppurative appendicitis
N.M	M	42	1 day	negative	RIF pain, vomitive	10.47	0.07 (0.7%)	8.04 (76.7%)	open appendectomy	acute suppurative appendicitis
O.F	M	30	1 day	negative	RIF, no vomiting, dysuria	14.98	0.58 (3.9%)	10.18 (67.9%)	open appendectomy, perforated appendicitis + abscess	Healing Appendicitis
A.A	M	33	1 day	positive	RIF pain, vomiting, no urinary symptoms	12.66	0.01 (0.1%)	10.58 (83.5%)	open appendectomy, appendicular mass	acute purulent appendicitis
H.A	M	39	1 day	positive	RIF pain, vomiting, no urinary symptoms	12.69			lap appendectomy, appendicular mass	acute appendicitis
A.M	M	26	1 day	negative	RIF pain, vomiting, no urinary symptoms, no diarrhea	14.15	0.56 (4%)	10.29 (72.6%)	open appendectomy	acute appendicitis

mucosal immunity, it also triggers an allergic reaction leading to appendicitis [7]. Another theory suggests that the disease is a result of chronic schistosomal granulomatous inflammation leading to fibrosis. This leads to narrowing of the appendical wall, causing further obstruction and acute appendicitis [8,9].

In conclusion, physicians of non-endemic areas should be aware of this disease, as migration and travelling to endemic areas have resulted in tropical infections being imported to other parts of the world. Even though preoperative suspicion would not alter the management of schistosomal appendicitis, postoperative histological diagnosis should result in further management including praziquantel.

Conflict of interest

I affirm that we have no financial affiliation (e.g., employment, direct payment, stock holdings, retainers, consultant ships, patent licensing arrangements or honoraria), or involvement with any commercial organization with direct financial interest in the subject or materials discussed in this manuscript, nor have any such arrangements existed in the past three years. Any other potential conflict of interest is disclosed. All human studies have been approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All

patients reported in this paper were informed about the study and an informed consent was obtained for being included in the study.

Funding

None.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patients for publication of this case series and accompanying histopathology images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Fawaz Abo-Alhassan and Fatemah Faras: wrote the manuscript; including the table and the references, did the literature review. Did the data analysis. Yousef M. Malek: Helped in data analysis and data gathering. Munish Joneja: wrote the histopathology section including the figure legends. Retrieved and reviewed

all the histopathological slides. Piyaray M. Dhar: Supervised the manuscript including the figures and table.

Guarantor

None.

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