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An Unusual Localized Association of Schistosomiasis and Acute Appendicitis: Case Report and Literature Review

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Abstract: Bilharzia or schistosomiasis is a tropical disease caused by parasitic worms. Several infectious pathogens (viruses, bacteria or parasites) may be involved in appendicitis. Acute appendicitis can be seen at any age and is one of the most frequent emergencies in digestive surgery. After malaria, schistosomiasis is the second most socio-economically devastating parasitic disease in Africa. When parasites are released by certain types of freshwater snails, they come into direct contact with people and cause infestation either through the skin or through the mouth affecting the digestive tract. Appendicular involvement is very rare in endemic areas, even exceptional in Western countries. Appendicitis is not specific to a parasite, but many parasites can be found during an appendectomy: pinworms, the most common in Europe, but also roundworms, taenia, amoebae, schistosomes. The appendix is inflamed or gangrenous, but is often healthy, the parasites being discovered incidentally. The role of parasites in the genesis of acute appendicitis is not clear. Appendicular schistosomiasis was first described in 1909, but remains a rare condition, although it has been reported from endemic areas. Appendicular schistosomiasis is a rare condition with an exclusively histopathological diagnosis. The urogenital, intestinal and hepatosplenic localizations are the most frequent while the other sites are extremely rare. Appendectomy must be followed by treatment with praziquantel to avoid the occurrence of complications. Herein we report a case of 20-year-old young man with appendicular schistosomiasis from and living in a tropical area. We discuss also, the place of this parasitosis in the genesis of appendicitis with a review of the literature.

Keywords: Appendicitis, Bilharzia, Schistosoma Haematobium, Praziquantel

1. Introduction

Acute appendicitis is one of the first surgical emergencies in the world [1]. The radical treatment for this surgical pathology is appendectomy. Appendicular schistosomiasis was first described in 1909, but remains a rare condition in Western countries.

However, the fact remains that it has been reported in tropical areas or in patients who have traveled to endemic areas.

It is estimated that at least 90% of those requiring treatment for schistosomiasis live in the tropical region. In Africa, schistosomiasis is the second most devastating parasitosis socio-economically after malaria. These parasitosis pose a public health problem in the countries concerned. Certain types of snails living in fresh waters release the parasites which will be either ingested or in direct contact with the skin thus causing an infestation.

Urogenital, intestinal and hepato-splenic localizations are the most common, while other sites such as the appendix are extremely rare.

Appendicular schistosomiasis is a rare cause of appendicitis and exclusively histopathological diagnosis [2]. The schistosomiasis infestation of the appendix has evolutionary and therapeutic characteristics that are specific to it, hence the need for a systematic histopathological examination of any specimen of appendectomy, a fortiori in areas of schistosomiasis endemic such as the case we report.

The aim of our work is to highlight through a case of

appendicular schistosomiasis, the contribution of histopathological examination in the diagnostic and therapeutic strategy of appendicitis.

2. Case Presentation

A 20-year-old male patient, with no particular pathological history known, presented to the surgical emergency department of the tertiary hospital center of Kiffa in Mauritania, with 4 days history of abdominal pain, early postprandial vomiting and fever. On admission, a reported temperature of 37.8 with stable vital signs on physical examination and he was conscious and oriented but in severe pain. The general condition preserved, but tenderness and pain on palpation of the right iliac fossa with positive Blumberg and Rovsing signs. Hyperleukocytosis with 12,000/mm^3 was found on the blood count, 80% of which were neutrophils with no associated hyper eosinophilia. The rest of the biological assessment was normal. An abdominal ultrasound performed showed a thickened appendix, measuring 8 cm and not compressible. The diagnosis of acute appendicitis was retained and the indication of an appendectomy was performed. We proceeded to an appendectomy by incision of Mc Burney then ligation and section of the meso appendix, the appendix was increased in volume, as shown in Figure 1, swollen, hard on palpation with false membranes.

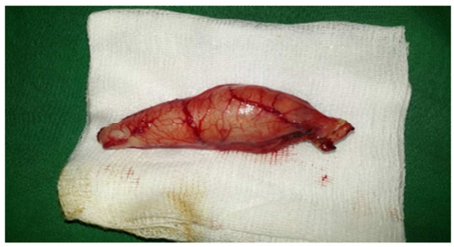

Figure 1. *Phlegmonous appendix, increased in volume, swollen.*

The immediate post-operative follow-up was simple. The patient underwent appendectomy with analgesics, antibiotics. Histopathological examination of the surgical specimen was requested. The laboratory examination focused on an appendix measuring 6.5 X 2.8 X 2 cm, covered with false membranes. The Figure 2 shows in section a thickened, hard appendicular wall with a constricted lumen.

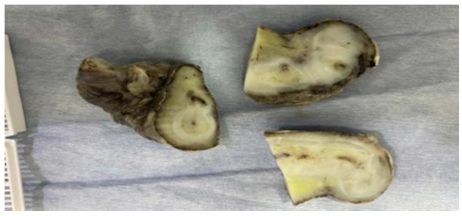

Figure 2. *Thickened and hard appendicular wall with a constricted lumen.*

Histopathological analysis in Figure 3 shows an appendicular mucosa with a preserved appearance, the appendicular walls contain an important epithelioid and gigantic-cellular granulomatous reaction, and numerous bilharzia eggs, often with terminal spur and calcified.

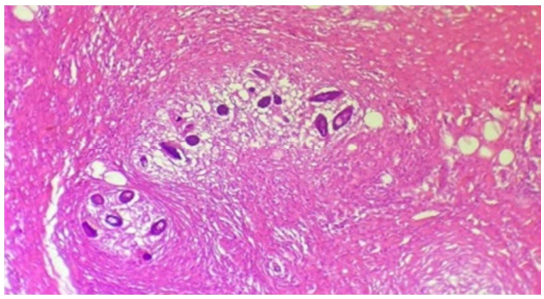

Figure 3. *Appendiceal tissue with dense suppurative inflammation and shistosoma eggs.*

The diagnosis of phlegmonous appendicitis of bilharzia origin was retained. The management was completed with a single dose of 1600 mg of praziquantel (at a single dose of 60 mg/kg).

3. Discussion

Bilharzia is a parasitosis caused by a trematode that affects more than 300 million people worldwide [3-6]. Schistosomiasis infestation of the appendix was first described by Turner in 1909 during an autopsy study [7-9]. This type of appendicular disease is a rare condition, even in endemic areas. Its frequency is very low and the prevalence rate varies according to the studies published in endemic areas or not.

In a study of 1920 appendectomies performed by Al-Kraida et al. [3] in Saudi Arabia, only 15 (0.8%) cases of schistosomiasis appendicitis were reported. In endemic areas, appendicular localization of schistosomiasis is very rare: 0.2% reported by study of Doudier et all [2] in Hong Kong, 2.4% found by two studies realized in Nigeria by Hodasi WM [9] and Sanusi and Bello [10]. In Ghana 2.9% were reported by M. Harouna Coulibaly [1], Bourée P et al. [7] and Callisto Madavo [8]. In Western countries, cases of schistosomiasis appendicitis are exceptional and are mainly seen in patients from endemic areas.

Most often, the clinical symptomatology is poor, and the clinical picture looks like a banal appendicular syndrome [11, 12]. The patient has no hepatosplenomegaly, no jaundice, no urinary disorders such as hematuria or dysuria that could lead to suspicion of the parasitic diagnosis preoperatively,

However, it has been reported by Mateus Zacarias et al [12] and I Thiam et al. [13] that, the infestation by Schistosoma japonicum and mansoni are responsible for frequent damage to the liver and spleen, and may even partly condition the prognosis by the resulting portal hypertension.

Schistosome eggs are frequently found in the wall of the digestive tract, but the parasitosis most often remains asymptomatic. Involvement of the digestive tract can be manifested by lower digestive hemorrhage secondary to the crossing of the intestinal wall by the eggs, by diarrheal manifestations or be incidentally discovered during an endoscopic or parasitological examination of the stool

performed for any other reason [6, 14].

The involvement of schistosome eggs in appendicitis is a source of controversy. Some authors believe that the prolonged passage of a large number of eggs through the mucous membrane would undoubtedly lead to ulcerations, the primary means of an inflammatory reaction. In addition, schistosomiasis granulomas can compress the vessels causing ischemic and necrotic lesions [4, 15]. Other authors prefer the term “bilharzia appendix” as opposed to true bilharzia appendicitis [12]. For the latter, the schistosomiasis infestation would rather lead to the obstruction of the appendicular lumen, thus facilitating a bacterial infection. It is therefore for these authors, a bacterial appendicitis favored by a parasitic infestation. These authors are even more convinced because the cases of schistosomiasis appendixes found in autopsy study far exceed the "supposed" schistosomiasis appendicitis reported in the literature [4, 9, 13].

Either way, the presence of Schistosoma eggs requires specific antiparasitic treatment with praziquantel at a single dose of 60 mg/Kg. Appendicular schistosomiasis is often associated with other visceral locations (bladder, intestinal, liver, spleen, etc.) [2, 14, 16].

In the absence of treatment, occlusive or neoplastic mechanical complications may occur [3, 7, 16].

No clinical, biological or radiological arguments can point to such an etiology. Only a pathological examination will confirm schistosomiasis involvement of the appendix, hence the importance of a systematic pathological examination of any appendectomy specimen [17].

The appendix may be the site of an infestation by the eggs of the parasites. With the exception of ascaris eggs which are large preventing any penetration into the appendicular lumen; other species such as Enterobius vermicularis, Trichiuris, Strongyloides Capillaria where schistosomes have been reported. Appendicular schistosomiasis is probably underestimated in Mauritania. It is responsible for a banal appendicular syndrome. The diagnosis of this condition is only histopathological. The treatment combines, in addition to surgery, drug treatment based on praziquantel. In the absence of this specific antiparasitic treatment, the evolution can be done towards an extension of the disease associated or not with mechanical or neoplastic complications.

4. Conclusion

In Mauritania, women who perform household chores in infested water, such as washing clothes, are at risk of infection. Bad habits and poor hygiene also make children particularly vulnerable to infection.

It is important to point out that to prevent populations from this disease, water used from rivers, lakes or reservoirs should be boiled to kill the parasites. It is therefore imperative to use drinking water.

In developed countries the incidence of schistosomal appendicitis is rare. On the other hand, in tropical or endemic zones, schistosomiasis appendicitis should not be neglected in the event of radiological calcifications of the appendix.

Clinicians should suspect schistosomiasis as the cause of acute appendicitis in patients who have stayed in endemic areas.

The diagnosis of schistosomiasis appendicitis can only be established after histological analysis of the appendectomy specimen, it is therefore necessary to analyze any appendectomy specimen in order to detect the presence of schistosomiasis eggs and to begin medical treatment by Praziquantel.

Conflicts of Interest

The authors declare no conflicts of interest.

Authors' Contributions

All authors contributed to the development and implementation of this work. AM Ded contributed to drafting the manuscript and data collections of patient and AM Idriss contributed to the critical revision by English translation of the manuscript and gave final approval of the manuscript. All authors read and approved the final manuscript. The authors also declare that they have read and approved the final version of this manuscript.

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