ISSN 1669-9106 MEDICINA (Buenos Aires) 2024: 84: 1235-1239

Case report

UNCOMMON PRESENTATION OF AN ACUTE APPENDICITIS CAUSED BY *ENTEROBIUS VERMICULARIS* IN AN ADULT

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Received: 2-V-2024
Accepted: 8-VII-2024

Abstract

Enterobius vermicularis is the most common parasitic infection in temperate and cold climates, affecting populations in both developing and developed countries. Although typically asymptomatic, it can lead to acute appendicitis, a relationship that has been a subject of debate. Therefore, we present a case of acute appendicitis induced by *E. vermicularis* in an adult patient and provide a comprehensive review of the current literature.

A 21-year-old man presented with acute right lower quadrant abdominal pain consistent with acute appendicitis. Laboratory and imaging studies supported the diagnosis. A subsequent laparoscopic appendectomy revealed the presence of *E. vermicularis* within the appendiceal lumen, confirming parasitic involvement. Postoperative management included anthelmintic therapy, resulting in an uneventful recovery.

Acute appendicitis is commonly recognized as a primary cause of lower abdominal pain, while the potential contribution of parasitic infestation remains subject to debate. E. vermicularis has been implicated in some cases. Diagnosis of acute appendicitis caused by pinworms typically involves direct visualization of the parasite or microscopic detection of eggs, with treatment consisting of surgical removal and anthelmintic therapy. It is essential to consider parasitic infestation in the differential diagnosis of appendicitis for proper diagnosis and appropriate management.

Association between parasitic infestation and acute appendicitis remains debated, recognizing the potential role of *E. vermicularis* in appendiceal pathology is crucial. This case serves to expand our understanding of appendicitis etiology and highlights the necessity for comprehensive evaluation and management.

Key words: Enterobius vermicularis, acute appendicitis, appendectomy, pinworms, Argentina

Resumen

Presentación atípica de apendicitis aguda causada por Enterobius vermicularis en un adulto

Enterobius vermicularis es la infección parasitaria más común en climas templados y fríos, afectando a poblaciones tanto en países en desarrollo como en países desarrollados. Aunque típicamente es asintomática, puede llevar a una apendicitis aguda, una relación que ha sido objeto de debate. Por lo tanto, presentamos un caso de apendicitis aguda inducida por *E. vermicularis* en un paciente adulto y ofrecemos una revisión exhaustiva de la literatura actual.

Hombre de 21 años se presentó con dolor agudo en fosa iliaca derecha, consistente con apendicitis aguda. Estudios de laboratorio e imagenología apoyaron el diagnóstico. Una posterior apendicectomía laparoscópica

reveló la presencia de *E. vermicularis* dentro del lumen apendicular, confirmando la participación parasitaria. El manejo postoperatorio incluyó terapia antihelmíntica, resultando en una recuperación sin complicaciones.

La apendicitis aguda es reconocida como una causa primaria de dolor abdominal. El diagnóstico de apendicitis aguda causada por oxiuros típicamente implica la visualización directa del parásito o la detección microscópica de huevos, con un tratamiento que consiste en la extracción quirúrgica y terapia antihelmíntica. Es esencial considerar la infestación parasitaria en el diagnóstico diferencial de la apendicitis para un diagnóstico adecuado y un manejo apropiado.

La asociación entre infestación parasitaria y la apendicitis aguda sigue siendo objeto de debate, reconocer el papel potencial de *E. vermicularis* en la enfermedad apendicular es crucial. Este caso sirve para ampliar nuestra comprensión de la etiología de la apendicitis y la necesidad de una evaluación y manejo integrales.

Palabras clave: *Enterobius vermicularis*, apendicitis aguda, apendicectomía, oxiuros, Argentina

Acute appendicitis (AA) is the most common cause of surgical acute abdomen, commonly occurring in young individuals, especially between the second and third decades of life. The pathophysiology is characterized by the obstruction of the appendicular lumen caused by various etiologies, including lymphoid hyperplasia, fecaliths, tumors, or foreign bodies; it can rarely be caused by parasitosis[1].

Infection by *Enterobius vermicularis*, also known as pinworms, is the most common parasitic infection in temperate and cool climates, affecting both developing and developed nations[2-3], with a global prevalence of 29.8%[2].

These pinworms have a length of approximately 10 mm and live with their heads embedded in the ascending colon and the adjacent intestine. The fecal-oral route is the most common infection pathway in humans, and the viability of the eggs on bedding and clothing is approximately two to three weeks, explaining its easy spread among family members[4].

Appendicitis induced by parasites accounts for 0.05 to 3% of cases[1]. It is suggested that the presence of parasites in the appendicular lumen causes an obstruction that promotes the accumulation of mucus and bacterial growth, triggering the process of distension and causing an increase in intraluminal pressure that progresses to the obstruction of venous flow, favoring perforation and complicating the condition[1-5].

Due to the rarity of AA caused by parasitic infestation, there is limited data in the literature. Therefore, we present a case of an adult patient with AA caused by *E. vermicularis*, and we provide a comprehensive review of the literature published up to the present moment.

Clinical case

A 21-year-old man presented to the emergency department with acute right lower quadrant abdominal pain persisting for 12 hours, accompanied by nausea.

Physical examination revealed tenderness in the right iliac fossa upon palpation with concomitant rebound tenderness upon release. Additionally, Rovsing's sign was positive.

The complete blood count revealed leukocytosis at 15.10 x 10^9/L, with differential counts showing 71% neutrophils, 1% eosinophils, and 23% lymphocytes. Urinalysis, blood urea, and serum creatinine were unremarkable.

An abdominal CT scan revealed slight thickening of the walls of the appendix, with no pathological findings reported in the rest of the abdominal examination.

Considering the suspicion of acute appendicitis, a laparoscopic appendectomy was conducted under balanced general anesthesia, with the patient in a supine position. The macroscopic appearance revealed a hyperemic, congested cecal appendix with edema and the presence of scant free fluid. Mesoappendix was released with monopolar energy and the artery was linked with hemoclips. The appendicular stump was secured using a Vicryl 0 endoloop. Unexpectedly, pearly white, round, and curved parasites were observed emerging from the appendix lumen during the appendicectomy (Fig. 1). Consequently, the appendix was removed through the suprapubic port using an endobag, and the specimen was sent to pathology for further evaluation.

After the surgery, our patient and their family members were prescribed a daily dose of mebendazole (100mg) with a repeat dose in 2 weeks.

The pathological examination revealed an edematous and vasocongestive appendix with the presence of *E. vermicularis* in the lumen confirming an appendicular inflammatory process secondary to parasitosis (Fig. 2). The patient had an uneventful recovery and was discharged on postoperative day 2. A month after the operation,he was free of symptoms.

Figure 1 | A series of intraoperative pictures showing *Enterobius vermicularis* emerging from the appendix lumen

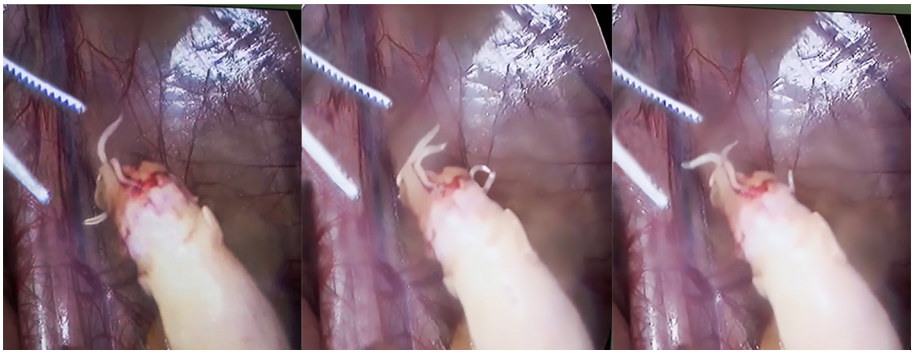
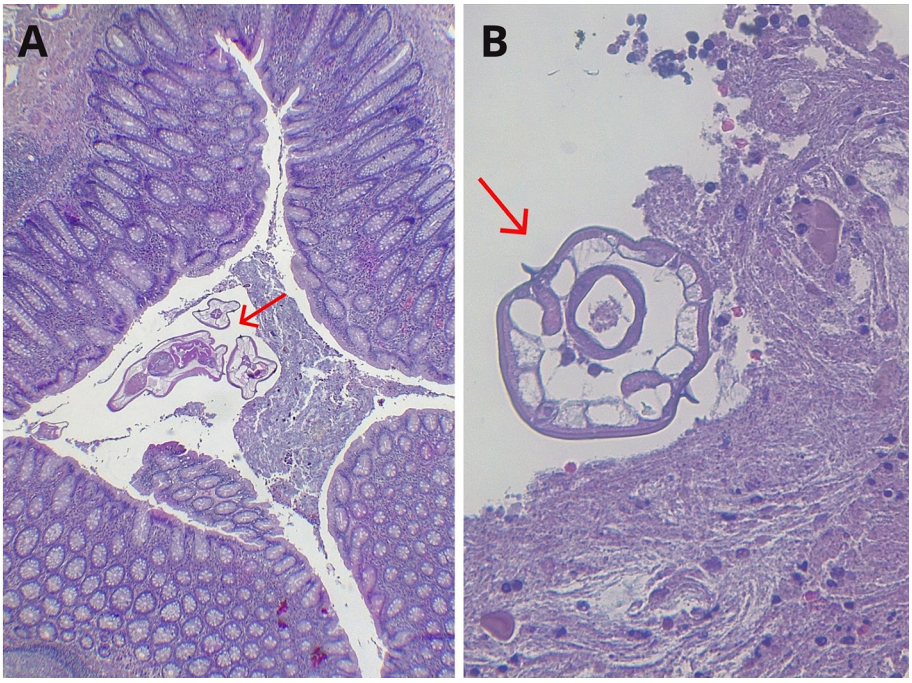


Figure 2 | A: A cross-sectional image of the appendix illustrating the presence of *E. vermicularis* within the appendiceal lumen (arrow). Hematoxylin and eosin stain, ×10 magnification. B: A cross-section of *E. vermicularis* in the appendiceal lumen (arrow). Hematoxylin and eosin stain, ×40 magnification



Written consent was obtained from the patient to use medical records and images in the publication of this manuscript.

Discussion

AA is a prevalent cause of lower abdominal pain, constituting 7–10% of all emergency department visits[6-7]. Its annual incidence varies from 5.7 to 50 cases per 100 000 inhabitants, notably peaking between the ages of 10 and 304. Worldwide, emergency operations for AA have a cumulative lifetime incidence rate of 9.0%[8]. The pathophysiology is characterized by the obstruction of the appendicular lumen, resulting from various factors, including lymphoid hyperplasia, fecaliths, tumors, or foreign bodies. Despite its rarity, parasitosis is recognized as a potential contributing factor[1].

E. vermicularis, commonly known as pinworm, stands as the most prevalent parasitic infec-

tion in both developed and developing countries with temperate and cool climates[2-3]. Reports indicate that 4% to 28% of children worldwide are infected, with manifestations typically being asymptomatic[4-9]. Prevalence of infection in Argentina is high, ranging from 10% to 60%, with the highest rates observed among children in socio-economically disadvantaged regions. A cross-sectional survey reports a median prevalence of 31.7% after analyzing 12 629 children across Argentina from 2010 to 2020. The most affected regions were the northeast and La Pampa, where median values approached 50%. In contrast, the Andean Northwest and Patagonia regions reported lower prevalence rates (20%)[2]. The prevalence of *E. vermicularis* is influenced by various individual and household-level factors. Individual factors include age, playing habits, and previous pinworm infections, while household-level determinants include family size and overcrowding conditions. Additionally, female children, those over six years old, and children living in homes without piped water or with incomplete parental literacy are at higher risk[2-3]. These pinworms are relatively small, measuring approximately 10 mm in length, with males ranging from 2 to 5 mm and females from 8 to 13 mm[4-10]. The most prevalent infection pathway in humans is the fecal-oral route[9]. These parasites predominantly inhabit the cecum of the large intestine. After being ingested, eggs hatch in the duodenum and take 1 to 2 months to develop into adult worms. During the night, the gravid female migrates to lay up to 15,000 eggs on the perineum[10-11]. Traditionally, it causes perianal pruritus and nocturnal restlessness. However, it may present similarly to AA, termed appendiceal colic, and occasionally advance to appendicitis[2-12]. Confirmation of the diagnosis involves directly visualizing the adult worm or detecting eggs under a microscope. The "cellophane tape test" can serve as a rapid method to conclusively establish the diagnosis[10].

Parasitic infestation is considered a controversial etiology for AA, and the relationship between them has been a subject of debate[1]. While several parasites have been linked to appendiceal infections, *E. vermicularis* accounts for 0.05 to 3% of cases globally[13]. The prevalence of *E. vermicularis* in appendicitis cases also demonstrates significant geographical variation, ranging from approximately 2% in the Americas to 8% in Africa. Socio-economic factors influence the distribution of this infection; low-income countries with a lower Human Development Index show higher prevalence rates compared to more developed nations[3].

According to the current hypothesis, the presence of parasites within the appendicular lumen transiently obstructs the appendix's lumen. This condition promotes the accumulation of mucus and bacterial overgrowth, initiating the process of distension. Consequently, it results in an increase in intraluminal pressure, ultimately progressing to venous flow obstruction[12-13]. Conversely, some authors presume that *Enterobius vermicularis* infestation of the vermiform appendix can mimic appendicitis. They use the term "appendiceal colic" to describe cases where clinical features suggest acute appendicitis despite the absence of macroscopic or histological evidence of inflammation, a condition known to resolve following appendicectomy. Surgeons should consider this differential diagnosis, especially when removing a macroscopically non-inflamed appendix, and take necessary precautions to minimize the risk of peritoneal contamination[13].

In cases where *E. vermicularis* is diagnosed intraoperatively, the treatment approach should follow several key principles. If any pinworms are identified, they should be managed through thermal ablation or endoscopic suctioning. A specimen bag must be utilized for the extraction of the appendix. Following the removal of the appendix, a thorough examination of the port sites, abdominal cavity, and pelvis is essential to identify and address any potential spillage before closure. These measures are crucial to prevent potential intraperitoneal contamination, which could lead to complications such as omentitis, pelvic inflammatory disease, and pelvic peritoneal granulomas[12].

It is mandatory to undergo pharmacological treatment after an appendectomy. There are a number of highly effective and well-tolerated anthelmintic agents for the treatment of enterobiasis, including mebendazole, pyrantel embonate, and pyrvinium embonate[14]. Nevertheless, there is a lack of larger controlled studies

regarding the individual drugs and treatment modalities. Some authors recommend treating patients with mebendazole, pyrantel pamoate, or albendazole at the time of diagnosis and administering another dose two weeks after treatment[12-15]. In other studies, mebendazole at a dose of 100 mg orally once is suggested as the primary choice. Alternatively, secondary options include pyrantel pamoate at 11 mg per kg orally once, with a maximum dose of 1 g, or albendazole at 400 mg orally once. Repeat treatment is advised after two weeks[10]. There is consensus across all studies indicating the necessity to treat household contacts, and it is recommended to thoroughly clean bedrooms and bedding.

In conclusion, the debated association between parasitic infestation and AA has prompted discussion within the medical community. In cases where *E. vermicularis* is diagnosed intraoperatively, specific principles should guide the treatment approach. Recognizing the potential link between *E. vermicularis* and appendicitis is crucial for accurate diagnosis and appropriate management. This underscores the importance of considering parasitosis within the broader spectrum of appendiceal pathologies.

Conflict of interest: None to declare

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