

CASE REPORT

Appendiceal syndrome associated with *Enterobius vermicularis*: a case reportFranklin Saavedra Idrogo¹, Luis Fernando Sánchez Rubio¹, Leslie Saavedra Paredes²¹ Surgery Service, Department of Surgery, Hospital II EsSalud, Cajamarca 06002, Peru.² Medicine Service, Department of Medicine, Hospital II EsSalud, Cajamarca 06002, Peru.

ABSTRACT

Background: Appendectomy is the most common emergency surgical procedure worldwide. The most widely accepted etiology is obstruction of the appendiceal lumen; however, parasitic etiology is uncommon. This report describes the case of a pediatric patient in whom active parasites compatible with *Enterobius vermicularis* were observed during transection of the cecal appendix.

Case description: A preoperative diagnosis of acute appendicitis was considered in a 2-year-old girl admitted to the Emergency Department for abdominal pain. On examination, muscle guarding and pain in the lower hemiabdomen were identified; the complete blood count showed leukocytosis. Laparoscopic surgery was performed, during which motile parasites compatible with *E. vermicularis* were found emerging from the appendiceal stump. The histopathological report confirmed the presence of *E. vermicularis* in the appendiceal lumen, without inflammatory involvement of the appendix.

Conclusions: Intraoperative visualization of motile parasites emerging from the appendiceal stump is unusual. In pediatric patients with abdominal pain and manifestations suggestive of acute appendicitis, it is important to obtain a more detailed medical and family history and to consider intestinal parasitosis as a relevant differential diagnosis in areas with deficient basic household sanitation and overcrowding.

Keywords: Appendectomy; Laparoscopy; Enterobius vermicularis; Enterobius (Source: MeSH)

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RESUMEN

Antecedentes: La apendicectomía es la cirugía de urgencia más común en el mundo. La etiología más aceptada es la obstrucción de la luz apendicular; sin embargo, la etiología por parásitos es poco frecuente. Se reporta el caso de una paciente pediátrica en quien se observaron parásitos activos compatibles con *Enterobius vermicularis* al seccionar el apéndice cecal.

Descripción del caso: Se planteó el diagnóstico preoperatorio de apendicitis aguda en una niña de 2 años que ingresó al Servicio de Emergencia por dolor abdominal. Al examen se determinó resistencia muscular y dolor en hemiabdomen inferior; el hemograma demostró leucocitosis. Se realizó una cirugía laparoscópica, durante la cual se encontraron parásitos móviles compatibles con *E. vermicularis* emergiendo del muñón apendicular. El informe anatomopatológico confirmó la presencia de *E. vermicularis* en la luz apendicular, sin compromiso inflamatorio del apéndice.

Conclusiones: La visualización intraoperatoria de parásitos móviles emergiendo del muñón apendicular constituye un hallazgo inusual. En pacientes pediátricos con dolor abdominal y manifestaciones sugestivas de apendicitis aguda, es importante ampliar los antecedentes médicos y familiares, considerando la parasitosis intestinal como diagnóstico diferencial relevante en zonas con deficiente saneamiento básico intradomiciliario y hacinamiento.

Palabras clave: Apendicectomía; Laparoscopia; Enterobius vermicularis; Oxiuros (Fuente: DeCS)

INTRODUCTION

Appendectomy is the most frequently performed emergency abdominal surgery worldwide. In children younger than 5 years, the estimated risk of undergoing this surgical intervention is 12% for boys and 23% for girls (1). From a pathophysiological perspective, acute appendicitis is attributed to obstruction of the appendiceal lumen by multiple causes, including intestinal parasitosis, which accounts for 0.05–3% of cases (2). The definitive diagnosis of appendicitis

is established through histopathological studies. However, the absence of inflammation has been reported in 10–20% of cases, corresponding to so-called negative or “blank” appendectomies, defined as the surgical removal of an appendix that is histopathologically confirmed to be healthy (3).

Enterobiasis or oxyuriasis is a household infection caused by *Enterobius vermicularis* that produces nocturnal anal pruritus, vulvar pruritus, and nasal itching, as well as gastrointestinal and neurological disorders that mainly affect the pediatric population (4). *E. vermicularis* is a small, filiform, whitish nematode of the class Secernentea and family Oxyuridae, also known as pinworm. It is responsible for the most common parasitic infestation worldwide, affecting more than 200 million people (5). It predominates in developing tropical countries (6). In Peru, the prevalence of helminthiasis remains significant, with a predominance in the jungle region, followed by the northern and central highlands of the country. Nevertheless, a progressive reduction has been observed in recent years, with an average annual downward trend of 13% (7). It occurs more frequently in males and in families with deficient basic household sanitation. Other associated risk factors include living with six or more people in the same household, onychophagia, and frequent contact (play) with soil or pets (8).

The life cycle of *E. vermicularis* is direct, and transmission occurs through the anus-hand-mouth route. Eggs enter through the oral or respiratory route; then, in the duodenum, the larvae are released and develop into adults. Adult parasites lodge in the ileum, cecum, ascending colon, and cecal appendix; in the latter location, they have been described as potentially playing a pathophysiological role in clinical presentations compatible with acute appendicitis. Finally, fertilized females migrate to the rectum, cross the anal sphincter, and lay eggs in the external environment (4).

Enterobiasis is the most common cause of parasitic acute appendicitis, with an incidence of 2% in the Americas. The association of *E. vermicularis* with this condition has been reported in 0.2–3.8% of cases (9), and it may present with symptoms similar to those of classic acute appendicitis; however, the pathophysiological mechanism does not involve parasite invasion or mucosal damage (10). Some studies describe the presence of *E. vermicularis* in histological specimens in up to 0.5% of cases (11). Nevertheless, in most of these anatomic pathology specimens, normal appendiceal histology is more frequently found (12), as occurred in the present clinical case.

The association between *E. vermicularis* infestation and acute appendicitis remains debated. It is important to consider the possible role of the parasite in the development of appendiceal processes or in the presence of similar symptoms (appendiceal syndrome), especially in regions where parasitosis is prevalent, to guide the prescription of complementary anthelmintic therapy.

We present the case of a 2-year-old girl with a preoperative diagnosis of acute appendicitis who underwent laparoscopic appendectomy. As an incidental finding, several active parasites were visualized emerging through the *ostium* of the appendiceal stump.

CASE DESCRIPTION

The patient was a 2-year-and-3-month-old girl from a household shared with two other families, with deficient basic sanitation, located on the eastern outskirts of Cajamarca, Peru. Her medical history included a urinary tract infection 2 months before admission and acute bronchitis 2 weeks before the current episode. She was admitted to the Emergency Department, accompanied by her parents. According to the maternal report, the girl had experienced abdominal pain for 40 hours. The pain was colicky, located in all four abdominal quadrants, and partially improved after semi-liquid bowel movements. Twelve hours after the onset of abdominal pain, abdominal distension developed, accompanied by semi-liquid stools of pasty and lumpy consistency, hyporexia, generalized weakness, and febrile peaks. In response to the symptoms, her parents administered syrups, which the mother could not identify, resulting in temporary symptom relief. Six hours before admission to the emergency department, the abdominal discomfort worsened and was associated with fever, drowsiness, and hyporexia (Figure 1).

On admission, she was evaluated in the Pediatric Emergency Department, where abdominal pain, liquid stools, hyporexia, and irritability were recorded in the medical history. Because of worsening abdominal pain and fever, she was transferred to the Surgery Department, where she appeared uncomfortable and irritable. Her heart rate was 98 beats per minute, her respiratory rate was 28 breaths per minute, and her axillary temperature was 39.4 °C. Her weight was 10.6 kg and height 80.6 cm. The respiratory system showed no abnormalities. Abdominal evaluation revealed distension, normal bowel sounds, and pain in the right iliac fossa on deep palpation, which exacerbated the patient's crying. In addition, muscle guarding was observed in the lower hemiabdomen. Notably, targeted history-taking to investigate parasitosis was not performed because the clinical picture suggested a surgical appendiceal condition.

Ancillary tests showed a leukocyte count of 16,520 cells/mm³ without a left shift, with 65.0% segmented neutrophils and 3.0% eosinophils. Hemoglobin was 13.4 g/dL, hematocrit 39.7%, glucose 73.6 mg/dL, and creatinine 0.36 mg/dL. Blood group and Rh factor were O positive. Coagulation tests showed a prothrombin time (PT) of 21.3 seconds, partial thromboplastin time (PTT) of 53 seconds, and international normalized ratio (INR) of 1.48. Urinalysis showed no abnormalities. Abdominal ultrasound showed the cecal appendix in the right iliac fossa, with an anteroposterior diameter of up to 7 mm, compressible, with increased echogenicity of the peritoneal fat, scant laminar fluid between bowel loops, and decreased peristalsis. Based on the clinical evaluation and laboratory studies, the Surgery Department established a presumptive diagnosis of acute appendicitis; therefore, informed consent was obtained from the parents, and preoperative preparation for laparoscopic appendectomy was initiated.

The surgical procedure was performed under general inhalational anesthesia, using three ports: an umbilical port for the optic device, one suprapubic port, and another in the left flank. Intraoperatively, a cecal appendix in pelvic position

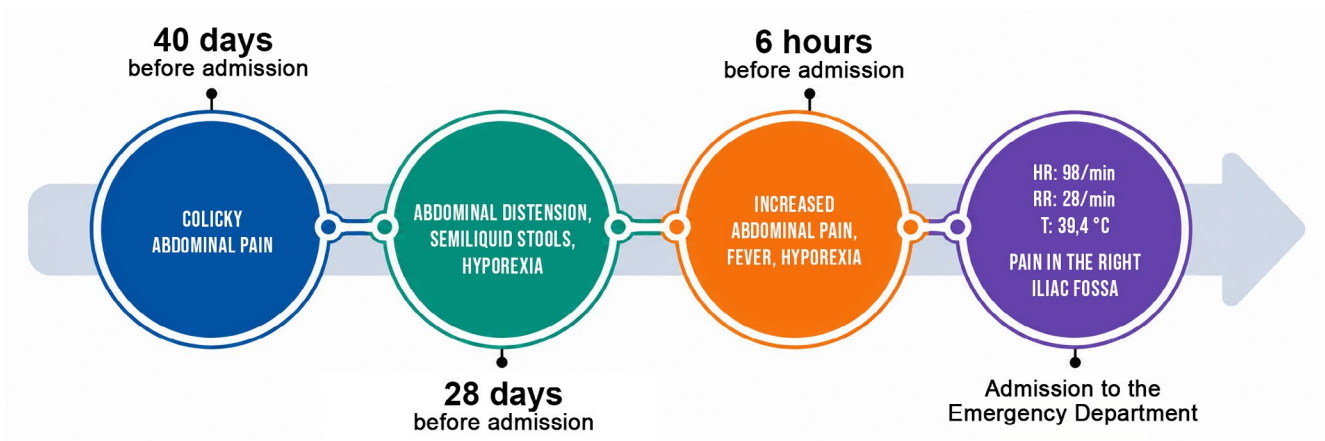


Figure 1. Timeline of the patient's clinical course

HR: heart rate; RR: respiratory rate; T: temperature.

was found, measuring 7.0×0.7 cm, apparently congestive in the distal third, with a base in good condition and no evidence of free fluid. During sectioning of the appendiceal base, four active fusiform parasites compatible with *Enterobius* spp. emerged through the *ostium* (Figure 2); these parasites were eliminated by thermal desiccation (Video 1). Subsequently, lavage and aspiration of the right iliac fossa with sodium chloride were performed to remove any remaining parasites. No sample of the parasites observed intraoperatively was obtained for parasitological study.

The histopathological report revealed a cecal appendix with mildly denuded epithelium and extensive lymphoid follicular hyperplasia; the muscularis propria showed no acute inflammatory infiltrate, so acute appendicitis was ruled out. In addition, a specimen of *E. vermicularis* was identified in the appendiceal lumen, containing multiple D-shaped fertilized eggs characteristic of this species (Figure 3).

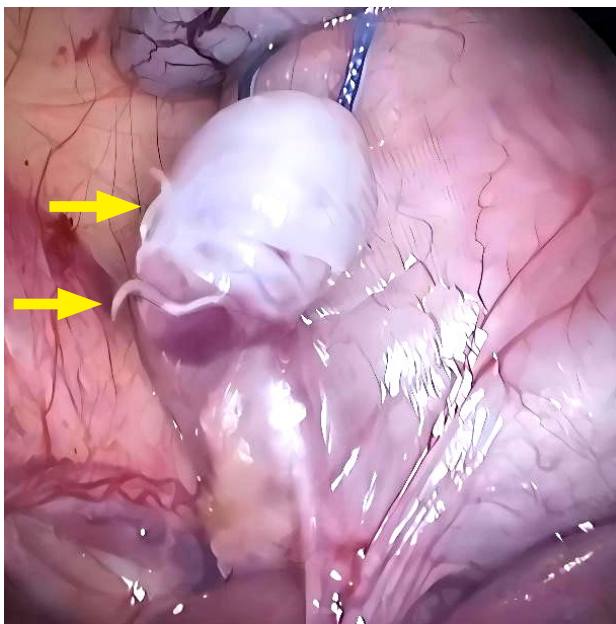
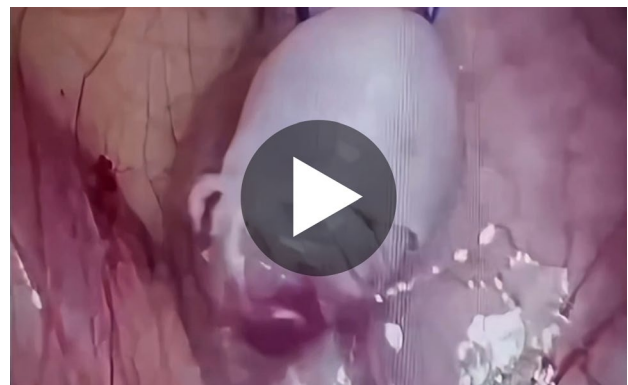


Figure 2. Intraoperative photograph showing parasites emerging from the appendiceal stump (yellow arrows)



Video 1. Active parasites emerging from the appendiceal stump

On the first postoperative day, the patient progressed favorably; she was active, had stable vital signs, and tolerated liquids, followed by a soft diet. The abdomen was soft and compressible, with mild pain at the laparoscopic port wounds. A single 400-mg dose of oral albendazole syrup was prescribed, with instructions to repeat the dose in 2 weeks. The parents were informed of the need for antiparasitic treatment for the entire family and adherence to appropriate hygiene measures

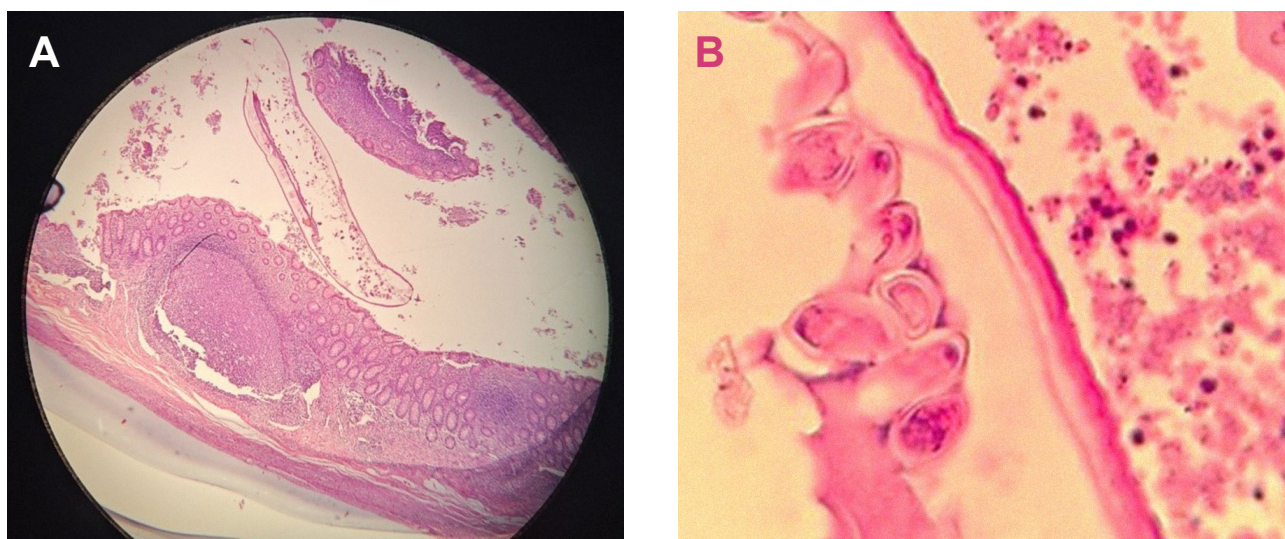


Figure 3. Histopathological findings of the cecal appendix showing the presence of *E. vermicularis*

A. The body of the female parasite is visible in the appendiceal lumen, with the appendiceal wall showing intact mucosa and lymphoid follicular hyperplasia; no acute inflammatory infiltrate is evident in the muscularis propria (H&E $\times 40$).
B. D-shaped *E. vermicularis* eggs inside the parasite (H&E $\times 400$). H&E: hematoxylin and eosin.

to prevent recurrence. At the seventh-day follow-up visit, the girl was well and had no symptoms; after 15 days, the family had completed the second dose of the antiparasitic treatment.

Written informed consent was obtained from the patient's mother for the scientific publication of the clinical data, images, and recorded video. The video was also shared with the mother during the postoperative report.

DISCUSSION

Acute appendicitis is defined as inflammation of the cecal appendix, characterized by migratory abdominal pain associated with other digestive symptoms such as nausea, vomiting, or hyporexia. Diagnosis is based mainly on clinical history, physical examination, laboratory tests, and, in some cases, imaging studies (13). Histopathological examination of the surgical specimen confirms the clinical diagnosis.

The etiology of appendicitis is not completely understood; however, several theories have been proposed to explain this condition, with obstruction of the appendiceal lumen being the most widely accepted. Described causes include lymphoid hyperplasia, fecaliths, tumors, foreign bodies, and intestinal parasites. Other studies have described local inflammatory processes secondary to infectious agents, including *Yersinia*, *Salmonella*, *Shigella*, mumps virus, *Actinomyces*, and others (3,13).

Acute appendicitis caused by parasites is uncommon; associated parasites include *Enterobius*, *Ascaris*, *Giardia*, and *Entamoeba histolytica* (14). The role of *E. vermicularis* in the

pathogenesis of appendicitis has been controversial for many years, since Fabrius discovered the presence of this parasite in the appendiceal lumen in 1634 (15). Other reports suggest that this parasite could irritate the appendiceal mucosa and cause microtrauma, thereby facilitating the entry of microorganisms into the appendiceal wall and triggering the inflammatory process characteristic of appendicitis (2,16). Conversely, cases involving *Enterobius* in the appendiceal lumen without inflammation have been described (17). The presence of these parasites in anatomic pathology specimens from appendectomies has been reported at rates of 1.53–4.2% (2,4,17,18). It has been proposed that parasitic elements may obstruct the appendiceal lumen and cause the so-called “appendiceal colic” without triggering an inflammatory response in the cecal appendix (17,18). Likewise, some reports indicate that appendiceal localization of *Enterobius* would favor fecalith formation in the lumen and proliferation of microorganisms in the wall, which could contribute to the development of appendicitis (4). In the present case, although *E. vermicularis* was identified in the appendiceal lumen, the histopathological study showed no inflammatory involvement of the appendix; therefore, it cannot be established that the parasite caused acute appendicitis.

The unusual nature of the case lies not only in the identification of the parasite in the anatomic pathology specimen, but also in the intraoperative visualization of motile parasites emerging from the appendiceal stump during laparoscopic appendectomy. This finding enabled direct documentation of the parasite's intraluminal presence during the surgical procedure.

The diagnosis of acute appendicitis is fundamentally clinical; however, despite the availability of modern diagnostic tools, it has been reported that 15–20% of appendectomies show no pathological evidence of acute appendicitis (17,19). Appendectomy remains the standard treatment, through either an open or laparoscopic approach. Laparoscopic appendectomy is widely accepted because it offers advantages, including a lower incidence of surgical wound infections, less postoperative pain, and faster recovery compared with open surgery (20). Although the clinical case presented showed that the clinical picture was not typical of acute appendicitis, laparoscopic appendectomy was decided because of the presence of pain and muscle guarding in the right iliac fossa, in addition to leukocytosis on the complete blood count, to reduce the risk of complications such as peritonitis (5).

The main limitation of this report was the inability to perform specific diagnostic studies in advance (such as the Graham test) due to the emergency medical context. Additional limitations included limited clinical information on the family and social environment and the absence of additional preoperative imaging studies, such as computed tomography, to complement the presumptive diagnosis. On the other hand, the main strengths were the clinical follow-up of the case, confirmation of the effectiveness of antiparasitic treatment in the patient's family, and the availability of anatomic pathology studies and images from the hospital's Pathology Service.

Regarding preoperative studies, the sensitivity of ultrasound for the diagnosis of appendicitis is variable (59–96%). In the present case, the ultrasound report described a compressible 7-mm appendix, a finding not suggestive of appendiceal inflammation. However, this examination should not be interpreted in isolation or replace clinical evaluation when considering the indication for emergency surgery.

The lack of tissue invasion may explain the absence of eosinophilia in this patient, given that the parasite was found in the appendiceal lumen. In addition, the eosinophil count in the complete blood count mainly increases in infections caused by tissue helminths (21). Regarding the abnormalities in the coagulation profile, no evident pathophysiological explanation was identified; therefore, the analyses were not repeated before her admission to the operating room.

It is important to mention that the patient had previous pediatric consultations for episodes of abdominal pain and urinary tract infection, which could have been related to undiagnosed enterobiasis. Likewise, given the family's socioeconomic conditions, overcrowding, and deficient environmental sanitation in the household, conditions frequently found in contexts of social vulnerability, complementary antiparasitic treatment should be considered in the pediatric population diagnosed with intestinal parasitosis.

CONCLUSION

Intraoperative visualization of motile parasites emerging from the appendiceal stump is unusual. In pediatric patients with abdominal pain and manifestations suggestive of acute appendicitis, it is important to obtain a more detailed medical and family history and to consider intestinal parasitosis in the differential diagnosis, especially in contexts of overcrowding and deficient basic household sanitation.

Author contributions

FSI: Conceptualization, Investigation, Data curation, Writing – original draft, Writing – review and editing. LFSR and LSP: Visualization, Writing – original draft, Writing – review and editing. All authors approved the final version of the manuscript.

Conflicts of interest

The authors have no relevant financial or non-financial interests to declare.

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Data availability

The data supporting the findings of this report are available from the corresponding author upon request.

Ethical aspects

Informed consent for the scientific publication of the clinical data, clinical images, and recorded video was provided by the patient's mother.

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