

Case report and review of the literature

Enterobius Vermicularis Related Acute Appendicitis: A Case Report and Review of the Literature

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Abstract: While the debate on the association between *Enterobius vermicularis* (*E. vermicularis*) and acute appendicitis has not been settled, a few case reports of this very rare encounter are beginning to come to light. *E. vermicularis* is one of the most common parasitic infestations around the world, acute appendicitis, on the other hand, is also a commonly encountered condition in general surgery. However, the association between the two remains controversial. Here we present a case report of a young woman with appendicitis associated with *E. vermicularis*.

Keywords: parasitological diseases; Oman; appendectomy; appendicitis; *Enterobius vermicularis*; Enterobiasis; case report; helminths; appendix; pinworms

1. Introduction

One of the most common emergency abdominal operations worldwide is for suspected acute appendicitis [1]. Appendicitis is an inflammation of the appendix characterized by abdominal pain. It is a malady that occurs when the insides of the appendix are blocked. The diagnosis of acute appendicitis is based on history, physical examination, laboratory evaluation and abdominal imaging. The classic symptoms include the aforementioned vague periumbilical pain as well as anorexia, nausea, intermittent vomiting, migration of pain to the right lower quadrant of the abdomen and low-grade fever. In approximately 90% of patients presenting with these symptoms, the diagnosis of acute appendicitis is made. The most common treatment is laparoscopic appendectomy [2]. A wide spectrum of cases have been cited as causes of appendicitis, and among the rarest are neuroendocrine tumor, serrated adenoma, low-grade appendicular mucinous neoplasm, hyperplastic polyp and intestinal parasite [3].

Enterobius vermicularis (*E. vermicularis*) has worldwide prevalence though crowded conditions are a risk factor for infection and they are more somewhat more common in developing countries [4,5]. *E. vermicularis*, also known as pinworms, is the most common human parasite, the most common helminth infection in the United States and are an infestation transferable only by humans [6]. Among the affected individuals, approximately 40% are asymptomatic. Pinworm infection is actually a self-limiting disease as the life expectancy of adult worms is so short, unless an autoinfection occurs. Perianal pruritis, is the most common symptom of this infection, tends to happen the most at nighttime while a person infected with the parasites is asleep. The itching and discomfort can disrupt sleep, and in some children, in about half of the cases, can enable a loss of bladder control. Scratching the perianal region can lead to ulceration and in some, repeated picking of this skin can result in lesions (excoriation), leaving an infected individual vulnerable to bacterial superinfection. Other potential indirect symptoms include perianal folliculitis, anal

dermatitis and ischiorectal abscess. Autoinfection or transmission may occur as a result of scratching because the action itself releases eggs which can easily stick to the fingers or under the fingernails and be passed on. In some rare cases, pinworms can be indirectly responsible for vulvovaginitis – irritation/inflammation of the area – if pinworms migrate to the vaginal area and this could also indirectly cause a urinary tract infection as other enterobacteria like *Escherichia coli* can adhere [7]. Pinworm infestation is also considered to be the most common helminthic infection found in the appendiceal lumen – the inside of the appendix – and has been said to cause acute appendicitis.

The role of *E. vermicularis* in relation to the pathogenesis of few cases of acute appendicitis has been the subject of controversy for many years, as pinworms have been discovered in the appendix as far back as 1634 by Fabrius [7,8] despite the fact that no causality has been reliably demonstrated. The majority of the cases reported has mainly been in the pediatric population. Here, however, we present a case report of an adolescent woman who presented to us with pain in the right iliac fossa, her abdominal examination revealed tenderness in the right iliac fossa with a positive rebound sign, blood tests showed normal inflammatory markers and abdominal imaging was suggestive of borderline tip appendicitis (a rare condition where the distal appendix becomes inflamed). Despite adequate conservative management she continued to remain symptomatic, so she underwent laparoscopic appendectomy. The histopathology of the re-sectioned appendix reported prominent lymphoid hyperplasia and *E. vermicularis* eggs were found within the lumen of the appendix.

2. Case Report

A 21-year-old female with no significant past medical or surgical history, presented to the emergency department at the Royal Hospital in Muscat, Oman, complaining of right iliac fossa abdominal pain of one week duration. The pain was progressive with associated anorexia. She denied any history of fever or change in her bowel habits but reported having regular clear vaginal discharge before menstruation. Her last menstrual period was a month ago. On examination, her vital signs were within normal limits. Her abdominal examination revealed tenderness in the right iliac fossa with a positive rebound sign. Urine analysis showed that no infection was present and pregnancy test was negative. Laboratory investigations showed hemoglobin of 13.3 g/dl, a white cell count of 4.1×10^9 . C-reactive protein was < 4.0 mg/L; coagulation profile and renal function tests were normal.

The patient's pelvic ultrasonography showed normal ovaries with no free fluid. A computed tomography (CT) scan was done given the low probability of acute appendicitis (Alvarado score of 4), which showed a retrocecal appendix with a short segment wall thickening of 7mm and no significant fat stranding suggestive of borderline tip appendicitis. When re-evaluation was done, abdominal tenderness was persistent despite adequate analgesia and hydration; therefore, a diagnostic laparoscopy and laparoscopic appendectomy under general anesthesia was performed. Intraoperative findings showed a mild hyperemic appendicular tip which was adherent to the lateral abdominal wall and cecum with minimal hemorrhagic fluid in the pelvis. Her postoperative period was unremarkable, and she was discharged home after 24 hours.

The histopathology report showed mild denuded epithelium and prominent lymphoid hyperplasia. In addition, *E. vermicularis* eggs were found within the lumen of the appendix. The patient and her family members received 400mg of albendazole once

weekly for three weeks. On her follow-up appointment, she was doing well with no complaints.

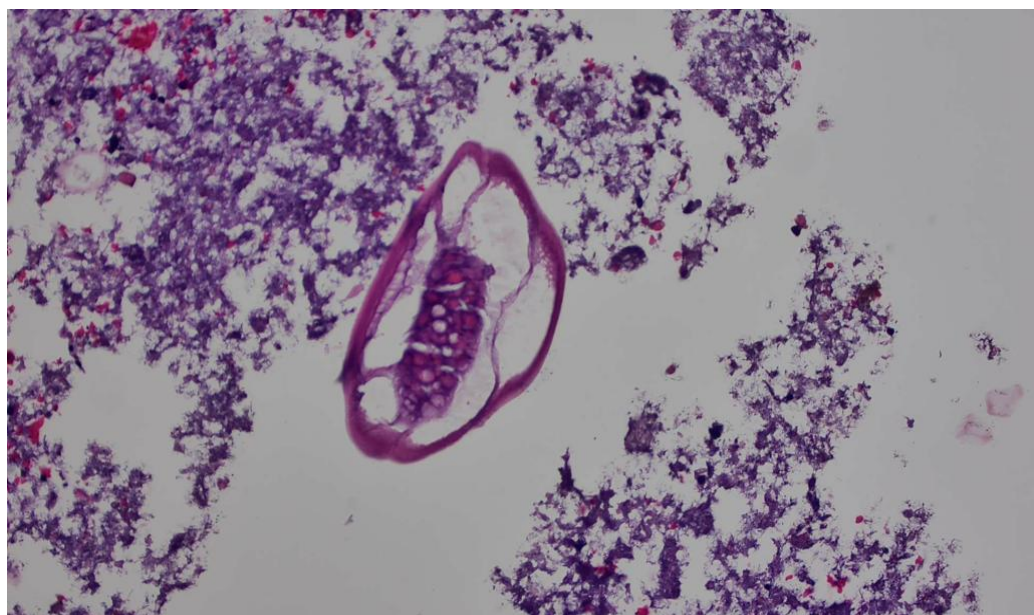
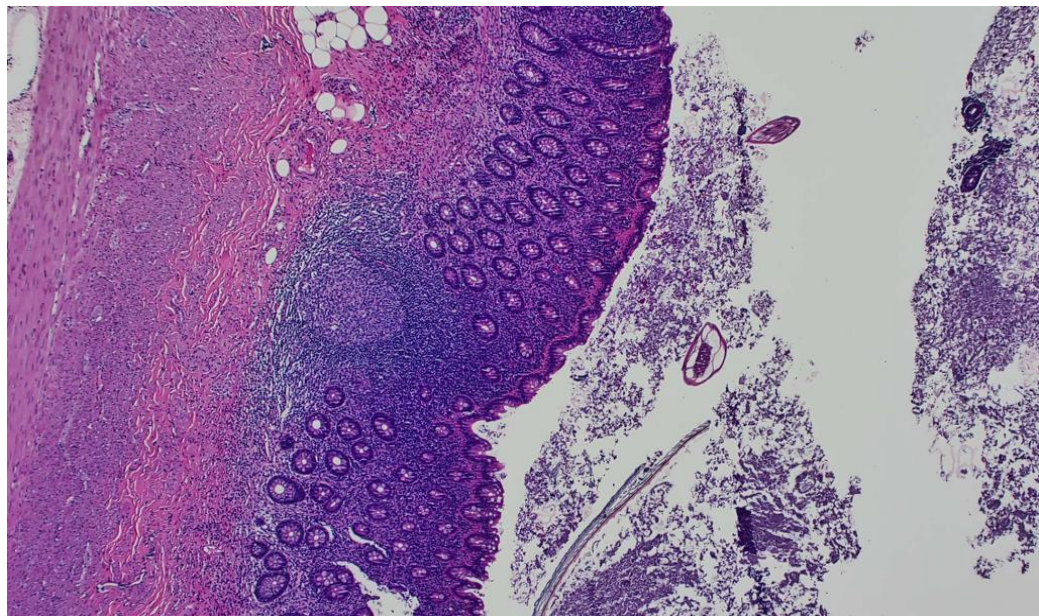
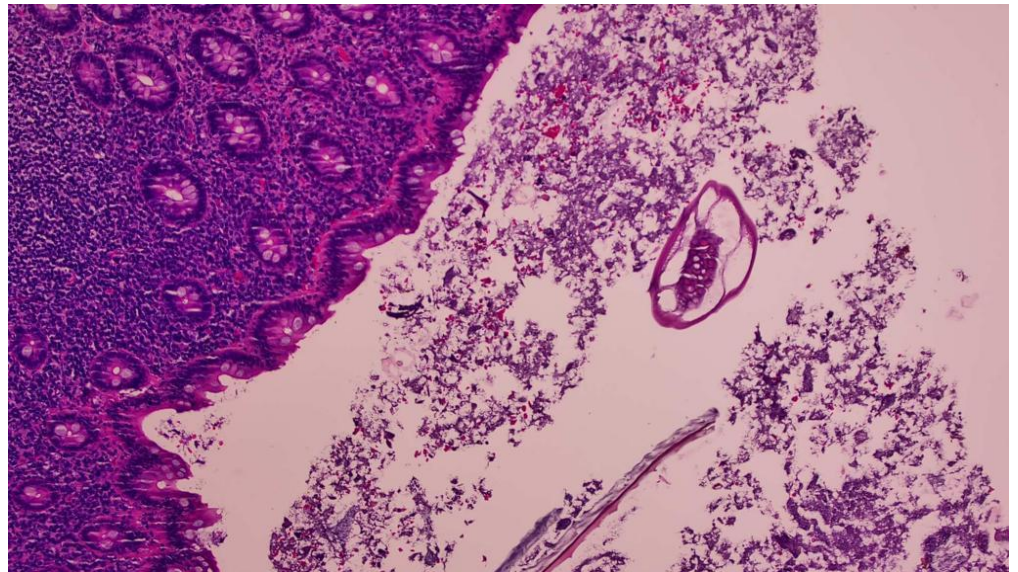


Figure 1. (a) Hematoxylin and eosin (H&E) stain show appendix mucosa involved by extensive



lymphoid hyperplasia and intraluminal *Enterobius vermicularis* egg; (b) High power view of H&E stain from the appendix lumen showing adult type worm of *E. vermicularis*; (c) High power view of H&E stain showing the extensive lymphoid hyperplasia of appendiceal mucosa which commonly is associated with *E. vermicularis* infection.

3. Discussion

Parasitic infestations are a global public health concern. More prominent with higher morbidity and mortality rates in developing countries, *E. vermicularis*, also known as pinworms or *Oxyuris vermicularis*, as they were traditionally called, is one of the most common nematode helminthiasis worldwide, especially among the pediatric group [10]. The infection becomes even more common among institutionalized individuals, and this includes children in daycare settings [6].

Prevalence studies conducted throughout the world in the past decade show the rate of *E. vermicularis* infection in children to be around 20% [4,6,11,12]. Rates have been noted as high as 38% in children 5-7 years old in Sri Lanka in 2013 [13] and as low as 0.21% in Taiwan following a 15-year reduction project [14] with 18% noted in a cohort of otherwise healthy Norwegian children [15] and 20% of American children who are said to have had at least one infection in their childhoods [6].

Even though this least harmful of the gastrointestinal nematode helminths is most common in children, that is not to say that infection does not occur in adults. Transmission to parents of infected children will occur and rates among adults who care for children are particularly high [6].

Symptomatic pinworm infection, referred to as enterobiasis, or oxyuriasis, the older term for the condition, was first described by a Swedish natural scientist named Carl von Linné. The thread-like appearance of these worms is the most distinguishing characteristic feature. The females are longer, at approximately 9-12mm in comparison to the shorter males at 3-5mm, and are visible to the naked eye. The whitish-beige color is also another striking feature, and they are typically round. The head section is rounded which contains the esophagus, which is muscular and the bulb, the tail section in the females is narrow and tapered sharply. The fertilized female worm has a very extensive uterine reproductive

system which when completely filled with eggs, can contain over 10,000 eggs per worm [7].

E. vermicularis is acquired typically by ingestion and occasional inhalation of eggs. The entire life cycle of *E. vermicularis* typically takes 2 to 4 weeks to mature to an adult worm. The time interval between ingestion of infective eggs and egg-laying, which is also termed as oviposition, is around 2 to 6 weeks. The adult female worm lays the eggs while the host is resting, which is predominantly at night while they sleep. The female worm leaves the anus and affix the laid eggs to tissue in the perianal area with an adhesive matrix, leading to the most common symptom of enterobiasis – perianal itch. *E. vermicularis* also commonly infects the cecum and rarely, extra-intestinal organs. The inhabitation of the gastrointestinal tract is considered more of an irritation rather than an infection. The most common manifestation is perianal itching which frequently happens at night, triggered by an inflammatory reaction to the presence of adult worms and eggs on the perianal skin. Itching leads to disintegration of the worm cuticle which leads to a massive release of eggs, which tend to get lodged beneath the fingernails, permitting further autoinfection and person-to-person transmission. Other presentations, such as abdominal discomfort, nausea and vomiting are generally related to a high worm burden. However, extra-intestinal organ involvement has been reported, including liver, kidney, peritoneal cavity, uterus, ovaries, mesentery, and lymph nodes [16-19].

The diagnosis of enterobiasis is usually made when a patient recounts intermittent perianal pruritis, this recurrent but not constant itching is typical in individuals with pinworm infections. Diagnoses can also be sought through inspection of the clothes and bed-sheets, as well as the anal verge which can demonstrate moving worm-like parasites. In cases of severe infestation, the worms may be expelled very visibly within the stool. The identification of the worms macroscopically, or the worms, and/or eggs, microscopically, is evidence of infection.

Since the oviposition takes place on the anal folds, the eggs are incredibly difficult to see with the naked eye. They can, however, be swabbed and identified under a microscope without much difficulty. A so-called Scotch tape test, also called an adhesive cellophane swab, can be performed, even by the infected individual themselves. The test, a sample collection, must be done first thing in the morning before any washing of the genital area or defecation. Using readily available adhesive tape, with buttocks spread apart, one can press a small strip several times to the anal and perianal region. Then the worm eggs are then able to be identified microscopically by affixing the tape, adhesive side down, to a slide [7].

Appendicitis is caused by obstruction of the appendiceal lumen which leads to the increase in the intraluminal pressure, followed by inflammation caused by bacterial overgrowth and probably ischemia and rupture without surgical intervention. Fecal stasis, fecaliths, lymphoid hyperplasia, vegetable residues, fruit seeds and tumors are considered the most common etiologies of appendiceal lumen obstruction [20,21].

Different intestinal helminths, namely *Ascaris lumbricoides*, *Trichuris trichura*, and *Taenia* species, have been reported as possible causes of acute appendicitis. The most common helminth reported among them is *E. vermicularis* [22]. Studies showed *E. vermicularis* was detected histopathologically in 0.3–4% of surgically removed appendixes [23]. In a recent retrospective study out of Iran, 20 out of 7628 appendectomy specimens contained *E. vermicularis*, a low rate but one that proves the occurrence [8]. The association between *E. vermicularis* and appendicitis is controversial. Its presence in the appendix can mimic appendicitis [24]. Nevertheless, the worm or its eggs can obstruct the appendiceal lumen leading to acute appendicitis which has been observed histologically in some cases. The most common histopathological finding in cases of appendicitis with *E. vermicularis* was lymphoid hyperplasia [25].

It has been well established that acute appendicitis is more prevalent in children and adolescents [26], like in our case of a 21-year-old female who presented with acute appendicitis. The histopathology report of the resected appendix showing *E. vermicularis* eggs within the lumen of the appendix was the only pointer toward the cause. Therefore, even

though rare, *E. vermicularis* should be noted as one of the differential diagnoses for the commonly presented condition of acute appendicitis as parasitic appendicitis requires a high index of suspicion and can easily be overlooked in low-prevalence regions [27]. Table 1 shows a few recent case reports on the association between *E. vermicularis* and acute appendicitis.

Table 1. The most important reports published in associating *E. vermicularis* to acute appendicitis.

Authors/references	Year of publication	No. of cases	Gender, mean age in years	Treatment	Country
Flores [28]	2022	1	Male, 10	Laparoscopic appendicectomy/anthelmintic	Mexico
Alshihmani [29]	2022	1	Male, 17	Open appendicectomy/anti-helminth	Iraq
Hooshyar [23]	2021	1	Female, 8	Open appendicectomy	Iran
Antilaky [30]	2021	1	Female, 5	Laparoscopic appendicectomy/anthelmintic	France
Lala [31]	2014	109	NA, 11.4	Laparoscopic appendicectomy/anthelmintic	New Zealand
Akkapulu [25]	2015	9	7 females, 2 males, 31	Appendectomy	Turkey
Fleming [32]	2013	13	NA, 11	Appendectomy	Ireland
Hamdona [33]	2013	30	17 male, 13 female, NA	Appendectomy	Palestine
Upadhyaya [34]	2015	7	4 females, 3 males, 25	Appendectomy	Nepal

Laparoscopic appendectomy remains the treatment of choice for acute appendicitis. However, evidence has been mounting toward use of broad-spectrum antibiotics, such as piperacillin-tazobactam monotherapy or combination therapy with either cephalosporins or fluoroquinolones with metronidazole, which has successfully treated uncomplicated acute appendicitis in approximately 70% of patients. The imaging findings has been sorted to categorize these patients, CT showing an appendiceal dilatation (appendiceal diameter ≥ 7 mm), or presence of appendicoliths, which is defined as the conglomeration of feces in the appendiceal lumen, identified patients for whom an antibiotics-first management strategy was more likely to result in failure. Therefore, for these group of patients who are fit for surgery, defined as having relatively low risk of adverse outcomes or postoperative mortality and morbidity, appendicectomy was recommended. But in patients without the above-mentioned high-risk CT findings, either appendectomy or antibiotics could be considered as first-line therapy. Even in those patients with high-risk CT findings, who were rendered as being unfit for surgery, the antibiotics-first approach was recommended, and surgery being considered if the antibiotic treatment were to fail [3]. Similarly, in cases involving enterobiasis infection, treatment with anthelmintic medication for the patient after appendectomy is highly essential. Enterobiasis is a very common and contagious infection, which has a high cure rate, but with very common recurrences. This infection can spread easily among family members due to long viability of its eggs on clothes which can linger on material for around two to three weeks. With the diagnostic challenges and

asymptomatic inhabitation of *E. vermicularis*, the need to treat close household contacts to prevent transmission and possible complications as stated above, is however, an unexplored field.

Hence, to conclude, though real, genuine appendicitis can still potentially be caused by *E. vermicularis* infection in the right circumstances where the patient has high worm burden that cause appendiceal lumen obstruction and eventually acute appendicitis, the mere presence of *E. vermicularis* alone in the appendix may otherwise only cause a local irritation which leads to lymphoid hyperplasia but not diagnosable appendicitis. Enterobiasis can mimic the clinical picture of acute appendicitis without the inflammation of the appendix. As observed in this case, most patients who presented with clinical picture of acute appendicitis where *E. vermicularis* was found in their appendix had normal inflammatory markers and their appendix showed lymphoid hyperplasia. This could imply that the presence of *E. vermicularis* might actually only mimic acute appendicitis. There is perhaps a need to assess for the presence of *E. vermicularis* prior to appendectomy, especially if the patient has normal inflammatory markers and is clinically stable. This could lead to the avoidance of unnecessary surgeries and their possible associated complications by simply treating *E. vermicularis* infection. However, this is just a possibility, and this theory requires further discussions in the future to come to a unified protocol on how to treat cases of suspected appendicitis.

4. Back Matter

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