

Case Report

The Rare Association of *Enterobius vermicularis* Infestation Causing Acute Appendicitis in Children: A Case Report and Review of the Literature

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Abstract: Acute appendicitis is the commonest causes of emergency surgery, having worldwide prevalence and affects all age groups. The etiology of acute appendicitis is multi-factorial and the pathology is most likely the obstruction of the lumen of the appendix. Acute appendicitis due to helminthes and protozoan infection is quite rare. Other unusual findings in appendectomy specimens are benign or malignant tumors. The presence of parasitic helminth *Enterobius vermicularis* (pinworm) in the appendix sometimes produces symptoms of acute appendicitis without any histological evidence, even though a patient might not have appendicitis. The causal relationship of appendicitis and pin worms' remains controversial. We present here a case of an 11-year old boy with enterobiasis of appendix presented with clinical features of acute appendicitis. *E. vermicularis* was discovered inadvertently after laproscopic appendectomy in a highly inflamed appendix. Prior to surgery, no parasites were revealed histopathologically. The relevant literature is also reviewed as accessed via PubMed and Google Scholar databases. Pinworm infestation can have various clinical presentations, including non-specific symptoms of decreased appetite, peri-anal pruritis, and loss of sleep. In acute condition, it can produce symptoms of 'appendiceal colic', or acute appendicitis in a non-inflamed appendix; or induce chronic inflammatory eosinophilic infiltrate or no tissue reaction in a histological appendicitis. In conclusion, the most common surgical condition in children is acute appendicitis. Parasitic infestations are a concern on a worldwide basis. The role of parasitic infestation in relation to appendicitis is controversial. Intestinal parasites may cause noteworthy morbidity and mortality.

Keywords: Appendicitis, Appendectomy, Helminthiasis, *Enterobius vermicularis*.

INTRODUCTION

The most common surgical cause of abdominal pain in children is the acute appendicitis (AA) [1]. The appendix is the most commonly intra-abdominal organ that is surgically removed or examined [2]. The causes of AA is mostly due to any obstruction, fecoliths, parasitic infestation in fecoliths, reduced blood supply, ischemic damage of the mucosa, and different infections. Any organisms colonizing the bowel can cause appendicitis, but parasitic infections also may cause a very small percentage of cases of appendicitis.

There is a long run relation between the roles of parasitic infection in the etiology of appendiceal diseases [3]. Reports also showed the presence of parasites in histologically normal appendices. These parasites may rarely change the histology during AA but can give the symptoms of acute appendicitis. Many

other cases reported the active penetration of these parasites into the wall of the appendix causing inflammatory reactions [4]. Thus the link of parasitic infestation with AA was confirmed in various reports.

The most common nematode parasite of humans in the developed world is the *Enterobius vermicularis* (pinworm) [5]. It is quite known to cause symptoms simulating the appendicitis. The occurrence of parasitic pinworm infestation with AA varies from less than 1% to nearly 4% [6]. As AA is generally a clinical diagnosis, not much confirmatory tests were done and thus the diagnosis of parasitic condition is made only after surgery [7].

These pinworm infestations may produce symptoms of AA, although the role of parasitic infestation in relation to appendicitis is controversial.

Pinworms may be found after colonoscopy or in the granulomas of the appendix [3]. These intestinal pinworm parasites may cause significant morbidity and mortality. We also presented a case report of a patient to give a glimpse on the concept. The patient was presented with symptoms of AA in whom the intra-lumen parasites were found during laparoscopic surgery. Histologically no parasites were revealed, the parasites were found in the lumen of the appendix. The dissected appendix appeared to be highly inflamed.

The most commonly examined intra-abdominal organ is the appendix [2]. The most frequent causes of appendicitis are due to ischemic damage of the mucosa, coincidence of luminal obstruction from a foreign body, faecolith or parasite; lymphoid hyperplasia; reduced blood supply and is not specifically associated with any pathogen. The precise diagnostic key for appendicitis for adults and children remains differential for wide and varied abdominal pain, but basically includes a critical review of history, right lower quadrant pain, leukocytosis and relevant physical findings.

AA was recognized since ancient times by the Egyptians, and remains almost unknown nearly 500 years ago. But now, in the 20th century, the rise in the incidence of AA is a greater problem [8-11].

Its incidence and prevalence may vary due to environmental, less refined dietary intake; allergic reactions, ethnic, association with privileged social class, age, urbanization, low-fibre diet having more of refined sugar, high-fibre diet, people migrating from rural to urban environment, seasonal variation, cultural and different demographic factors [9]. Due to changes in the pattern of dietary intake, the incidence of AA may increase thus pose a threat to the society as an emerging disease. Strikingly, worldwide the most frequent cause for emergency operations in visceral surgery is suspected AA. It is less frequent or particularly pertinent due to a reported low incidence in the developing countries. In developed countries, though its incidence has fluctuated, it is the most common intra-abdominal surgical emergency encountered in both children and adults [9, 12]. The lifetime risk of having an appendectomy is 25% for women and 12% for men [13, 14]. Interestingly; appendicitis is more common in males than females as revealed in some reports [15].

In children, the most common surgical cause of abdominal pain is AA [16]. Rapid and clinical diagnosis of appendicitis in children reduces the morbidity of the common cause of pediatric abdominal pain. Obstruction during AA may be due to ascariasis [17], actinomycosis [18], amebiasis [19], balantidiasis [20], *Blastocystis hominis* [21], enterobiasis [22], endometriosis [23], granulomatous diseases [24], trichuriasis [25], tuberculosis [26], taeniasis [27],

schistosomiasis [29], different kinds of tumor [23], carcinoma and adenoma [29], lymphoma [28], eosinophilic granuloma [24], and neurogenic appendicopathy [30].

Intestinal parasitic infestations are ubiquitous on a worldwide basis, may cause significant paediatric morbidity and mortality. *Enterobius vermicularis*, commonly known as pinworm (or oxyuris), causing widespread nematode parasitic infection is estimated to affect nearly 200 million people worldwide [31]. The association between pinworm and appendicitis dates back to late 19 century, but the role of parasitic infestation in producing symptoms of AA is controversial. In some recent study, in histologically normal appendices, the parasites were most frequently seen and can give the symptoms of AA. *E. vermicularis* infestation can also be revealed during appendectomy [3, 9-10, 32-33].

These parasites cause inflammatory reactions by invading or active penetration of the wall of the appendix [4], causes pathologic changes like lymphoid hyperplasia, acute phlegmonous inflammation, ruptured appendicitis [34] and also life-threatening complications like peritonitis and gangrene [34]. In the developed world, *E. vermicularis* is the most common parasite of humans. It is the most prevalent nematode in the United States [5]. *E. vermicularis* also causes symptoms simulating appendicitis and the occurrence of AA due to *E. vermicularis* infestation in every socioeconomic group with varied geography varies from less than 1% to nearly 4% worldwide [34-37] with also a higher reported incidence of granulomas in the appendix in presence of pinworms [38].

AA being a clinical diagnosis, basically based on past history, strong clinical suspicion, and physical findings. So, further diagnostic studies generally are not needed for confirmation and thus in most of the reported cases, the diagnosis or presence of the parasite was revealed only after surgery [7]. Although fecoliths and lymphoid hyperplasia are the usual causes of obstructions in AA, enterobiasis is a rare cause in causing acute appendicitis in children and its associated inflammation [1, 8, 10, 32, 39]. Though appendicitis operation and helminth infections is quite common among children [20], we also highlighted just one case report from Kolkata to visualize the concept of this review i.e. helminths (pinworms) infestation causing appendiceal luminal obstruction and presenting as appendicitis is uncommon [33]. Parasites are rarely found associated with inflammation of the appendix [11]. The aim of this study is to establish the possible role of *E. vermicularis* in the pathogenesis of appendicitis and to recall the need for medical treatment after appendectomy.

CASE REPORT

In this paper we also report an 11-year old male patient from Kolkata, India, with symptoms of acute appendicitis with absence of histological inflammation, where pinworm parasites were found during laparoscopic surgery. Prior histological examination revealed a normal appendix with no parasitic infestation in the patient.

This previously healthy boy was presented with a 3-days history of abdominal pain, mainly in the right lower quadrant. His pain was sometimes colicky and sometimes spasmodic. The pain score was 8/10. When he was examined, his Rovsing's sign, Obturator sign were positive. He also had fever on the day of presentation. The fever was continuous without rigor and was relieved temporarily after taking Paracetamol (650 mg/dose). He had symptoms of nausea and vomiting. He had loss of appetite one day prior to pain in abdomen. There were no sick contacts and no history of uncooked food consumption. No history of diarrhea or travel history recently known. On arrival, he was conscious, alert but with a toxic face. Physical examination revealed tenderness in the right iliac fossa with rebound tenderness and guarding. Right lower quadrant tenderness and rigidity were found on deep abdominal palpation. Due to the combination of the classic symptoms and a typical progression of symptoms coupled with right lower quadrant tenderness, AA was clinically suspected and confirmed by investigations. Surgical removal was decided. The chest was clear with equal breath sounds bilaterally. The remainder of the physical examination was unremarkable. Laboratory evaluation included a complete blood count, with elevated total white blood

count (Table 1). On urinalysis, no abnormality was seen. All other tests were within normal ranges, included in Table 1. Ultrasound examination showed non compressible, non-motile hyperechoic tubular texture in the right iliac fossa with thickening of the appendix wall.

Based on the findings of the physical and clinical examination, a clinical diagnosis of appendicitis was made. The patient was taken to the operating room for laparoscopic appendectomy. During the laparoscopic surgery the appendix was found to be elongated (length 160 mm and diameter 7.2mm), but multiple mobile helminthes were noted in the lumen of the appendix (Figure 1B). Appendix was inflamed. The cytopathology report of the helminthes identified them as *E. vermicularis*. The patient was treated with anti-helminthic drug and recovered completely from abdominal symptoms. He was discharged on the 3rd post-operative day.

Using the PubMed and Google Scholar databases, 128 studies published between August 2014 and December 1952 were compatible with our criteria. Fifty of these were written as original articles, 19 were review, 37 as case reports, and ten as case series. When we looked at the countries in which the case reports were prepared, 7 were from Europe, 6 from Asia, 6 from the Americas, 8 from Africa, and one were from Australia. In total, 252 cases were discussed in these articles, and all the patients were operated on the presumption of acute appendicitis. Unusual findings were detected in 65 of the cases.



Fig. 1: *E. vermicularis* discovered after laparoscopic appendectomy

Postoperative photograph showing an appendix together with pinworms (marked with a circle) found in the lumen of the appendix. The dissected appendix

was highly inflamed (length 160 mm and diameter 7.2mm).

Table 1: Clinical history and laboratory parameters

Parameters	Clinical & Laboratory Parameters
Physical Parameters	
Age (years)/ sex(M/F)/ Weight (Kg)	11/Male/45
Chief Complain	1. Pain abdomen for 3 days 2. Vomiting for 2 days. 3. Fever for 2 days 4. Loss of appetite for same duration
Birth and Vaccination History	1. Normal delivery 2. Immunization up to date
Past Medical History	Medical jaundice at 6 years of age
Past Surgical History	Not significant
Drug Allergy	Name not known
On Examination	
A. General examination	
	Patient conscious & alert
GCS	15/15
Pulse/min	112
Temperature (°F)	99.6
Pallor	Nil
BP (mm of Hg)	100/60
Hydration status	Mild dehydration
Clubbing	Nil
B. Systemic examination	
Chest (Air entry)	1. Adequate 2. Respiratory rate: 20/min
CVS (S1 and S2)	Audible, no gallop
Abdomen	1. Soft 2. On superficial palpation- there is no tenderness 3. On deep palpation- there is muscle guard and tenderness over right iliac fossa
C. Local examination	
	1. No skin rash 2. No blisters 3. No petechie
D. Laboratory investigations	
Hb (gm/dl)	13.2
D-1. Inflammatory and Infective Markers	
TLC (cells/cu.mm)	16,800
DLC (%)	N (89), L (6), M (2), E (3)
ESR (mm/first hour)	36
CRP (mg/L)	42
D-2. Sugar Level Tests	
Blood Sugar (mg/dl)	RBS-118
D-3. Renal Function Tests	
Urea (Mg/Dl)	24
Creatinine (mg/dl)	0.9
D-4. Electrolytes	
Sodium (Na+) (mEq/L)	136
Potassium (K+) (mEq/L)	4.2
D-5. Coagulation Markers	
Liver Function test 1. Albumin (gm/dl) 2. Other liver enzymes	Within normal range
D-6. Chest X-Ray	
Straight X-Ray Abdomen (Erect)	Normal profile seen
D-7. USG of Abdomen	1. Appendix is enlarged and advised to correlate

	clinically 2. Other profile normal
D-8. Cardiac Functions Tests	
ECG	Within normal limit
D-9. Urine Routine Examination	
	Pus cells 1-2
E. Treatment Summary	
<ul style="list-style-type: none"> • Patient was initially managed by 900 ml normal saline (i.v.) as bolus dose • 0.45 dextrose normal saline (500 ml) + 5 ml KCl (i.v.) 6 hourly • Analgesic injection Drotaverine (1 ampoule/SOS/i.m. stat) • Injection Pantoprazole (i.v.) (40 mg/O.D.) • Injection Ceftriaxone (1 gm/i.v./ twice daily) (after proper skin test) • After collecting all the reports, patient was thoroughly evaluated clinically and then laparoscopic appendectomy done under general anesthesia. Intra-operatively when appendix was separated then large numbers of pinworms were seen within operative field. • Huge numbers of infested pinworms were seen inside the surgically dissected appendix. • Specimen was send for histopathology and to parasitological evaluations. • Reports were inflamed appendix with <i>Enterobius vermicularis</i> infestation. • Anti-helminthic drug was given 	
F. Output of Patient	
	Successfully discharged

#GCS= Glasgow Coma Scale, min=minutes, °F= degree Fahrenheit, BP= Blood pressure, CVS= Cardiovascular system, S₁= first heart sound and S₂=second heart sound, TLC=Total Leucocyte count, DLC= Differential leucocyte count, ESR= Erythrocyte Sedimentation rate, CRP= C-reactive protein, N=Neutrophil, M=Monocyte, L=Lymphocyte, E=Eosinophil, ND=Not Determined, ECG=Electrocardiogram, µg=microgram, mg=milligram, pg= picogram, i.v= intravenous, i.m=intramuscular, SOS= as and when required, NA=Not applicable, mEq= milliequivalents, dl= deciliter , USG =Ultrasonography.

DISCUSSION

The association of this parasitic infestation with AA varies worldwide. *E. vermicularis* is a parasite that inhabits the human digestive tract. We present an 11-year-old patient with symptoms mimicking AA that were found to have *E. vermicularis* infection. Intra-operative findings included an inflamed appendix with lumen infested with *E. vermicularis*.

E. vermicularis infection of the appendix can present with a clinical picture similar to AA or accounts for appendiceal related pain in the absence of histological inflammation [40]. In at-risk populations, it should be included in the differential diagnosis for children with right lower quadrant abdominal pain [41]. Patients with parasite-associated appendicitis should receive anti-helminthic treatment because appendectomy eliminates the complication but not the cause of intestinal disease [42, 43].

CONCLUSION

The continuing challenge of AA remains a considerable problem in both the developing and developed world. With modern management the mortality is low, but morbidity still exists. Without clinical suspect, preoperative diagnosis of pinworm infestation is almost impossible. Parasitic infestation may resemble symptoms of AA but rarely parasitic infection causes it. This is also important in the differential diagnosis cases that mimic Crohn's disease.

Further study of possible etiological factors may yield further clues as to its pathogenesis.

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Patient Confidentiality

For the purpose of maintaining patient confidentiality, the personal details of the patient have not been given in this review article. Informed consent form obtained from patient is submitted together with this manuscript.

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Appendix

Informed consent form