CASE REPORT

Enterobius vermicularis (pinworm) infestation in a child presenting with symptoms of acute appendicitis: a wriggly tale!Louise Dunphy,¹ Zoe Clark,² Mazhar H Raja¹

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ABSTRACT

Background The European Society for Medical Oncology (ESMO) has developed the ESMO Magnitude of Clinical Benefit Scale (ESMO-MCBS), a tool to assess the magnitude of clinical benefit from new cancer therapies. Grading is guided by a dual rule comparing the relative benefit (RB) and the absolute benefit (AB) achieved by the therapy to prespecified threshold values. The ESMO-MCBS v1.0 dual rule evaluates the RB of an experimental treatment based on the lower limit of the 95%CI (LL95%CI) for the hazard ratio (HR) along with an AB threshold. This dual rule addresses two goals: inclusiveness: not unfairly penalising experimental treatments from trials designed with adequate power targeting clinically meaningful relative benefit; and discernment: penalising trials designed to detect a small inconsequential benefit.

Methods Based on 50 000 simulations of plausible trial scenarios, the sensitivity and specificity of the LL95%CI rule and the ESMO-MCBS dual rule, the robustness of their characteristics for reasonable power and range of targeted and true HRs, are examined. The per cent acceptance of maximal preliminary grade is compared with other dual rules based on point estimate (PE) thresholds for RB.

Results For particularly small or particularly large studies, the observed benefit needs to be relatively big for the ESMO-MCBS dual rule to be satisfied and the maximal grade awarded. Compared with approaches that evaluate RB using the PE thresholds, simulations demonstrate that the MCBS approach better exhibits the desired behaviour achieving the goals of both inclusiveness and discernment.

Conclusions RB assessment using the LL95%CI for HR rather than a PE threshold has two advantages: it diminishes the probability of excluding big benefit positive studies from achieving due credit and, when combined with the AB assessment, it increases the probability of downgrading a trial with a statistically significant but clinically insignificant observed benefit.

was reassured and sent home. On this presentation, her pain increased in intensity. She denied a history of nausea or vomiting. Her bowels were opening regularly, once a day, with no evidence of blood or mucous. There was no history of anal pruritus. There were no urinary symptoms. She weighed 34 kg. Her medical history was unremarkable, and she had no known drug allergies. On examination, she was tender in the right iliac fossa with guarding. She was Rovsing's sign positive. There was no evidence of peritonism, and bowel sounds were present on auscultation. There was no palpable lymphadenopathy. Respiratory and cardiovascular examinations were unremarkable.

INVESTIGATIONS

Her observations were as follows: temperature 37.3°C, respiratory rate 26, SpO₂ 98% on air, heart rate 73 beats/min and blood pressure 99/65 mm Hg. Her urine dip was positive for leucocytes but negative for nitrites. Urine digital microscopy demonstrated white cell count 5/L. It was negative for red cells, epithelial cells and casts. Haematological investigations confirmed a normal white cell count with an eosinophilia (table 1). Her CRP was elevated at 8.7 mg/L.

As her clinical signs and symptoms were suggestive of acute appendicitis, further investigation with an ultrasound scan of her abdomen was not deemed necessary. Her Alvarado appendicitis score was 5/10, suggesting the possibility of appendicitis. A decision was made to proceed to laparoscopic appendicectomy.

TREATMENT

Under general anaesthesia, a laparoscopic appendicectomy involving an open pneumoperitoneum with 2×5 mm ports in the left iliac fossa was performed. Intraoperatively, the appendix did not look acutely inflamed, although it was slightly congested and was unusually wriggly. The appendix was dissected midway, and multiple worms were macroscopically visible adherent to the mucosa (figure 1, supplementary video 1). There was no contamination of the peritoneal cavity with *Enterobius vermicularis* on dividing the appendix. The appendix was delivered with the use of a specimen bag. Macroscopically, the external surface of the appendix had a pale grey colour, and no other gross abnormality was detected. On sectioning, the lumen

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CASE PRESENTATION

A 10-year-old girl presented to the emergency department with a 48-hour history of acute abdominal pain radiating from her umbilicus to her right iliac fossa. She reported to her attending general practitioner a diffuse abdominal pain 24 hours previously, and her white cell count and C reactive protein (CRP) were unremarkable. She

Table 1 Haematological investigations confirmed an eosinophilia

Complete blood count	
White cell count	8.8×10 ⁹ /L
Differential count	
Neutrophils	2.73%
Lymphocytes	3.94%
Eosinophils	1.618%×10 ⁹ /L
Monocytes	0.67%×10 ⁹ /L
Basophils	0.11%×10 ⁹
Haemoglobin	130 g/dL
Mean corpuscular volume	83.1 fL
Platelets	345×10 ⁹ /L
C reactive protein	8.7 mg/L
Liver function	
Alanine aminotransferase	9 IU/L
Globulins	28 g/L
Bilirubin	4 µmol/L
Total protein	74 g/L
Albumin	46 g/L
Renal	
Sodium	139 mmol/L
Potassium	4.2 mmol/L
Urea	3.4 mmol/L
Creatinine	41 µmol/L

of the appendix contained soft faecal material. Numerous small, thin white worms were observed. Microscopically, the appendix showed no evidence of acute inflammation. *E. vermicularis* (pinworms) were seen.

OUTCOME AND FOLLOW-UP

Her postoperative recovery was unremarkable, and she was discharged 24 hours later. She was treated with mebendazole 100 mg orally with a second dose 2 weeks later to treat possible reinfection. Close family members also received prophylactic treatment. There was complete resolution of her symptoms at her 6-week follow-up. Her eosinophil count was normal (0.22%).

DISCUSSION

E. vermicularis (pinworm), formerly known as *Occiur vermicularis* or oxyurides, is a small, white obligate nematode that affects 1000 million individuals worldwide.¹ It has also been referenced in the ancient writings of Hippocrates, as far back as 430 BC. Pinworm infestation is the most common helminthic

Figure 1 The appendix was dissected midway, and multiple worms were macroscopically visible adherent to the mucosa.

infection in children with an estimated prevalence of up to 50% in children and 20% in adults.² In 1919, Symmers first described inflammation in the ileocolic region caused by *E. vermicularis*, with further reports showing an association with inflammation of the appendix.^{3,4} As far back as 1898, Wiebe described a case of *E. vermicularis* affecting the vermiform appendix.⁵ One year later, in 1899, Still was the first to describe a case in the lumen of the appendix of children under 12 years.⁶ In 1901, Metchnikoff brought up the question of the relationship between the parasites and inflammation of the appendix.⁷ In subsequent papers, Aschoff⁸ and Hueck⁹ stated that oxyuria may evoke symptoms resembling appendicitis. Parasites are rarely associated with appendiceal inflammation, with reports from developed countries stating that 1.5% of cases are associated with parasites.¹⁰ It is usually an incidental finding in the appendix of children aged 7–11 years.

The most common clinical presentation of pinworm infection is perianal itching, predominantly occurring at night-time and is caused by the migration of the female pinworm to the anus to lay eggs. The presence of pinworms in the appendix has been shown to cause symptoms mimicking appendicitis or appendiceal 'colic' but frequently without any histological evidence of acute inflammation. Appendiceal colic consists of intermittent chronic right lower quadrant and pelvic pain and can be explained by the hypothesis of appendiceal lumen obstruction. Infestation may present with ileocolitis, enterocutaneous fistula, urinary tract infection, mesenteric abscesses, salpingitis and appendicitis. Macroscopic and microscopic blood may be evident in the stool. Its host is the human. Autoinfection occurs by scratching the perianal area and transferring infective eggs to the mouth with contaminated hands.

After ingestion, the eggs hatch, and the larvae are released into the small intestine. The adult worms mainly establish themselves in the caecum and the appendix. Interestingly, one-third of affected individuals are asymptomatic. Obstruction of the appendiceal lumen can occur with parasites and their eggs, thereby increasing the intraluminal pressure. Pathologically, *E. vermicularis* can lead to lymphoid hyperplasia, acute phlegmonous inflammation dissecting the muscularis propria and peritonitis. The finding of *E. vermicularis* in appendectomy histopathological specimens is a rare incident. The reported rates of inflammation in specimens from appendices infested with *E. vermicularis* range from 13% to 37%.

E. vermicularis should be considered in children presenting with signs of acute appendicitis.¹¹ Arca *et al* reported it in 1.4% of 1549 appendectomies in the USA.¹⁰ It can also result in complications such as intestinal obstruction, perforation and colitis. *E. vermicularis* has also been implicated in pelvic inflammatory disease, pelvic pain and ectopic infections.

The diagnosis of appendicitis remains clinical and can be challenging in the young, elderly and those individuals of reproductive age. Indeed, physical examination is generally not specific enough to differentiate between parasitic and ordinary appendiceal pain. A history of perianal itching should be elicited. The Cellotape test, which involves pressing a cellophane tape to the perianal skin to detect eggs, has a low diagnostic sensitivity. Stool examination is not necessary as worms and eggs are not passed in the stool. The Alvarado score, the Raja Isteri Pengiran Anak Saleha Appendicitis score and the acute inflammatory response were developed to help the clinician establish the diagnosis, but they are not universally acceptable.¹² In equivocal cases, the diagnostic certainty can be improved by the addition of radiological imaging. In cases of suspected appendicitis, ultrasound and CT have proven to be beneficial. Since McBurney introduced open

appendicectomy in 1891, it has been considered the standard approach. Admittedly, the worms can be strongly adherent to the mucosa by their heads; therefore, partial cutting of the appendix with traction will enable inspection of the lumen. If the worms are clearly visible within the luminal cavity, careful thermal dissection with the tip of the scissors directly onto the worms or endoscopic suction should be performed. However, since the advent of laparoscopic appendectomy by Semm in 1983, it has been gaining acceptance.¹³

However, it runs the risk of contamination of the peritoneal cavity with worms.

Pharmacological eradication with antihelminthics such as mebendazole is also required.

Education about hand hygiene should also be provided.

In conclusion, the presence of *E. vermicularis* infection usually accounts for appendiceal-related pain in the absence of histological inflammation. This case illustrates the importance of considering *E. vermicularis* infestation in a child presenting with right iliac fossa pain as the clinical signs of intestinal parasitic infection may mimic acute appendicitis. A careful evaluation

of symptoms such as pruritus ani or eosinophilia on laboratory examination, especially in children, could prevent unnecessary appendectomies.

Anthelmintic therapy is required even after appendicectomy, as appendicectomy only eliminates the consequence of the disease but not its cause.

Contributors All authors contributed to the writing of this manuscript. LD wrote the case report and discussion. ZC performed the literature search. MHR edited the paper and final approval.

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Learning points

- *Enterobius vermicularis* infestation should be considered in the differential diagnosis of right iliac fossa pain in children, but who do not have a significantly raised white cell count or high Alvarado scores.
- A history of perianal itching should be sought, predominantly occurring at night-time and is caused by the migration of the female pinworm to the anus to lay eggs.
- Appendicectomy should proceed with caution if *E. vermicularis* is suspected as there is a risk of peritoneal contamination.

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