CASE REPORT

Intestinal spirochaetosis mimicking acute appendicitis with review of the literature

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SUMMARY

Human intestinal spirochaetosis is a well-established micro-organism existing in the colon. It is less commonly seen in the appendix, and rarely presents as acute appendicitis. We present a case of a man presenting with symptoms consistent with acute appendicitis. The literature on spirochaetosis presenting as acute appendicitis is also reviewed.

BACKGROUND

Human intestinal spirochaetosis is a well-established micro-organism existing in the colon. It is less commonly seen in the appendix, and rarely presents as acute appendicitis. We present a case of man presenting with symptoms consistent with acute appendicitis and review the literature of this relatively rare pathogen. The literature on spirochaetosis presenting as acute appendicitis is also reviewed below.

CASE PRESENTATION

A 24-year-old man presented with a 2-day history of abdominal pain. This was associated with vomiting, headache and a mild fever. Significant medical history included asthma, and at the time of presentation, he was on a course of prednisolone for a recent attack.

On examination, there was right iliac fossa tenderness and percussion tenderness with a positive Rovsing's sign. The white cell count was $20.3 \times 10^9/L$, neutrophils $17.0 \times 10^9/L$ and C reactive protein 2 mg/L.

Figure 1 Appendiceal mucosa colonised with spirochaetes (arrow points to mucosa lined by spirochaetes staining pale purple with H&E histological stain).

The clinical diagnosis was acute appendicitis and the patient underwent a laparoscopic appendicectomy. Despite the symptoms being suggestive of acute appendicitis, on visualisation, the appendix appeared normal. Histologically, there was no acute inflammation; however, there was widespread colonisation by spirochaetes (figures 1 and 2).

OUTCOME AND FOLLOW-UP

The patient recovered well postoperatively and was discharged.

DISCUSSION

Intestinal spirochaetosis is a histological diagnosis, confirmed by the presence of spirochaete micro-organisms on the luminal surface of colorectal mucosa.¹ In animals, particularly in pigs, it is an established cause of diarrhoea, malnutrition and poor growth.² In humans, the clinical significance remains unclear.

In humans, *Brachyspira pilosicoli* and *B. aalborgi* are the two micro-organisms of the spirochaetes phylum which predominate, typically seen in poor socioeconomic regions, HIV individuals and homosexuals.¹ The prevalence depends on the method of detection. Rectal biopsies have demonstrated a prevalence between 2% and 7% in the Western world, compared with much higher levels seen in India and other Asian countries.^{3–6} Faecal analysis reports detection rates of between 1.1% and 5% in developed countries,¹ compared with rates as high as 64.3% in Indian villages.¹⁷ It has been postulated that the variation between different geographical locations is in part due to varying levels of hygiene and antibiotic use.³

As discussed above, the clinical significance of intestinal spirochaetosis remains up for debate. There are studies which argue that the bacteria exist as commensal organisms and are unlikely to be of pathological significance. These studies report the majority of infected patients as being asymptomatic.¹³⁸ However, other studies report symptoms in up to 86% of colonised individuals.⁹ In patients where the micro-organisms has invaded the epithelium, rather than simply existing on the surface of the luminal epithelium, it is more commonly symptomatic, presenting with chronic watery diarrhoea and abdominal pain, with no other obvious underlying cause.¹⁹ Other rare cases of abdominal pathology should be ruled out.¹⁰ The degree of microscopic invasion into the mucosa, loss of microvilli and microscopic burden of spirochaete

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Figure 2 Immunohistochemical staining for spirochaetes (arrow points to spirochaetes, staining positively with specific immunohistochemical stain).

organisms on the luminal surface often correlates with the severity of the symptoms.¹¹ In symptomatic individuals, coinfection with other pathogens including *Shigella flexneri*, *Helicobacter pylori*, *Enterobius vermicularis* and *Neisseria gonorrhoeae* further clouds the pathological significance of spirochaete infection.¹

Response to treatment with antibiotic therapy, typically metronidazole targeting the anaerobic spirochaete bacteria, is varied. Some patients have an excellent response, with complete resolution of gastrointestinal symptoms; others remain symptomatic with or without the spirochaete micro-organisms remaining on the luminal epithelial layer.^{1 12} Despite the varied response, initial treatment with antibiotics is still widely accepted as the initial treatment. Metronidazole is typically used; however, there are reports of macrolide use as well.¹³

The spirochaete micro-organism has been widely reported as existing in the colorectum^{8 14 15}; however, colonisation of the appendix is less reported, and even less have reported cases of intestinal spirochaetosis presenting as acute appendicitis. To our knowledge, there are only a few publications in the literature which have found patients with spirochaetosis presenting clinically as acute appendicitis. Between 1971 and 1997, three studies were published reporting the presence of spirochaetes in 0.7%–4.8% of confirmed cases of appendicitis. In 0.9%–12.3% of cases, appendicitis was clinically suspected, but the appendix was histologically normal, except for spirochaete colonisation. This latter group has been termed ‘pseudoappendicitis’ by Henrik-Nielsen *et al.*^{5 16 17}

More recently in 2010, 113 cases of confirmed intestinal spirochaetosis were reported, 16 of which were in the appendix. Of these 16 cases, 5 had confirmed appendicitis.⁸ In 2012, a case report was published on a histologically confirmed case of appendicitis with spirochaetosis.¹⁸

It is evident from the review of the literature, that in individuals with histologically confirmed acute appendicitis, the appendix is rarely concurrently infected with spirochaetes. In

comparison, individuals who have pseudoappendicitis, as in the case we present above, the incidence of spirochaete colonisation is slightly higher. It has been postulated that this group present with symptoms of appendicitis due to a rapid and massive colonisation of the appendix by spirochaetes.¹⁶

Learning points

- ▶ The clinical significance of intestinal spirochaetosis is uncertain.
- ▶ If symptomatic, intestinal spirochaetosis commonly presents with watery diarrhoea and abdominal pain.
- ▶ Intestinal spirochaetosis rarely presents as acute appendicitis.
- ▶ Colonisation of the appendix with spirochaetes may result in pseudoappendicitis.

Contributors JG: drafted article, collated information and images for case report. CB: critically revised article. DA: critically revised article, chose histology images and drafted histological entry into case report discussion. CP: chose patient for case report, was overall supervisor and critically revised article. All authors have approved final article and are accountable for the article.

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