Serendipitous discovery in a fistula-in-ano

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CLINICAL QUESTION

A woman in her early 30s presented with perianal purulent discharge. A rectal examination showed a high complex fistula-in-ano. A fistulotomy was performed wherein two fistulous tracts at the 4 o'clock and 8 o'clock position involving the external sphincter muscle were identified, and the granulation tissue was submitted for histopathological evaluation.

WHAT IS YOUR DIAGNOSIS?

A. A dense infiltrate of macrophages with an artefact amidst the granulation tissue.
B. Macrophages exhibiting emperipolesis along with a cotton fibre adjacent to them.
C. Macrophages exhibiting emperipolesis along with a parasite in the adjacent tissues.
D. Macrophages exhibiting emperipolesis with a slender vegetable cell adjacent to them.

DISCUSSION

Amidst abundant granulation tissue, without eosinophils, were numerous multinucleated macrophages showing emperipolesis (figure 1). The engulfed cells were lymphocytes, plasma cells and occasional neutrophils. Because the emperipolesis was striking, we examined the section carefully and saw a single larval form of *Enterobius vermicularis* (figure 2). The 0.15-mm-long worm was depleted in deeper sections of the paraffin block. Review of the patient's preoperative laboratory data showed no peripheral eosinophilia. Stool examination performed after treatment was negative. The patient and all members of the family were treated with a course of albendazole-ivermectin and are asymptomatic two and half months post-procedure.

Enterobius vermicularis is a parasite specific to humans.[1] Infection is acquired by ingestion of eggs which hatch in the ileum. The larvae develop into adults which live in the caecum. Gravid females migrate to the rectum, where oviposition occurs. A characteristic symptom, pruritus ani, results in autoinoculation of the eggs via hands that have scratched the perineum. Rarely, retrograde infections occur where the larvae migrate from the perineum to the rectum.[1] There are reports of various ectopic sites of infection, particularly the female genital tract.[1]

There are earlier reports of *E. vermicularis* infestation of fistula-in-ano.[2 3] We hypothesise that in our patient, the newly hatched larva travelled up the existing fistula-in-ano. The larval form shares the characteristic pin-shaped tail and differs from the adult worms in that they are smaller and coiled. *E. vermicularis* at ectopic sites rarely show peripheral eosinophilia[4] or tissue eosinophilia, as illustrated in our patient and in others.[4] Stool examination shows parasites or eggs in only 5%–15% of cases.[4] The more sensitive 'Scotch tape' test could be used to demonstrate eggs on the perineum in an active infection.[5]

'Emperipolesis' is inextricably linked with sinus histiocytosis with massive lymphadenopathy (SHML)/Destombes-Rosai-Dorfman disease, an unusual non-neoplastic lesion of histiocytes.[6] It has also been reported in viral infections (acute hepatitis B), bacterial infections (Rhinoscleroma)[7] and in haemoparasites[8] (Leishmania-associated and malaria-associated haemophagocytic lymphohistiocytosis). As we demonstrate, emperipolesis may manifest even in non-blood parasitic infections.

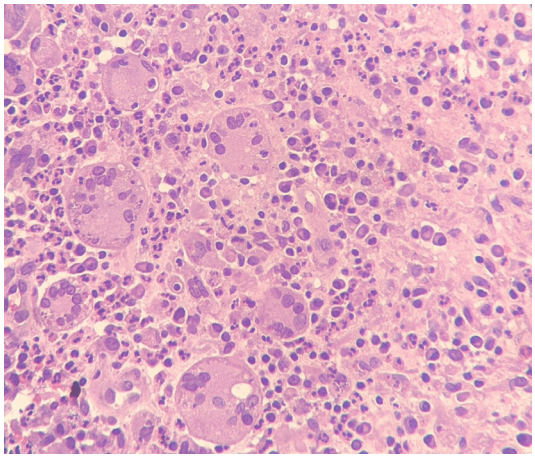

Figure 1 Sheets of multinucleated macrophages exhibiting emperipolesis.

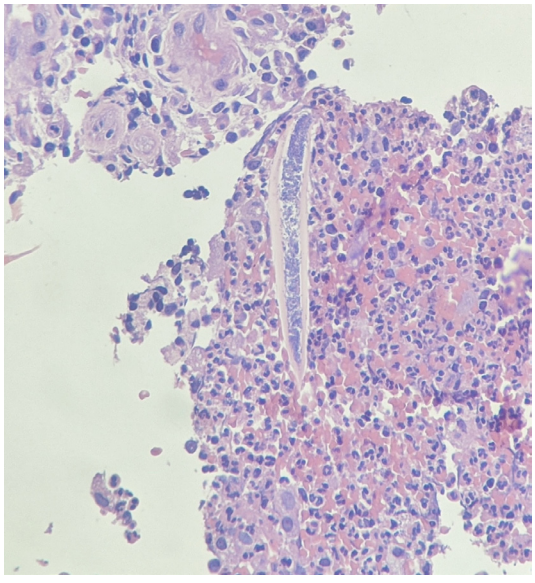

Figure 2 Larval form of *Enterobius vermicularis* amidst the granulation tissue.

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CORRECT ANSWER

C. Macrophages exhibiting emperipolesis along with a parasite in the adjacent tissues.

Take home messages

- Great care must be taken even while evaluating even 'mundane' specimens such as granulation tissue/fistulae-in-ano.
- *Enterobius vermicularis* may be detected at histological evaluation of ectopic sites of infection.
- Emperipolesis is not specific for sinus histiocytosis with massive lymphadenopathy but can be seen associated with parasitic (as well as bacterial and viral) infections.
- The entire household needs to be treated for *E. vermicularis* infection.

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