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Enterobius vermicularis (pinworm) infection of the liver mimicking malignancy: Presentation of a new case and review of current literature

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ABSTRACT

INTRODUCTION: Enterobius vermicularis or "pinworm" infection of the liver is an extremely rare condition with only five cases previously reported in literature. It is characterized by the presence of granulomas in the liver with a necrotic core, containing adult helminthes or their ova. Because of the relatively mild symptomatology associated with this disease, prior to the arrival of modern imaging methods hepatic enterobiasis was an incidental intra-operative finding during abdominal surgery for other conditions. In recent years however, with high-resolution abdominal imaging readily available and the improved safety of hepatic resection, a lower threshold for treating suspicious hepatic nodules aggressively with surgery is being adopted.

PRESENTATION OF CASE: We present the second case in international literature, where *E. vermicularis* of the liver was mistaken for malignancy and led to hepatic resection and perform a literature review of the five previously documented cases of hepatic enterobiasis.

DISCUSSION: Our report identifies certain trends in this condition's aetiology and clinical behaviour, but due to its rarity definitive answers cannot yet be established.

CONCLUSION: We do not advocate a change in the current approach of suspicious hepatic nodules, but we do feel that better understanding of the mechanisms involved with hepatic enterobiasis could, in the future, prevent unnecessary surgery.

1. Introduction

Infection with *Enterobius vermicularis* (pinworm), also known as enterobiasis, is the most common helminthic infection in the United States of America and Western Europe, mostly affecting children aged 5–10 years. The worms live and reproduce in the ileum, cecum, colon and appendix and the nematode female migrates to the anus to deposit its eggs and die, usually at nighttime. Transmission is believed to be via the fecal–oral route.[1] Despite enterobiasis of the gastrointestinal tract being very common, ectopic localization of the infection is rare. While a number of ectopic cases of the female genital tract have been reported, enterobiasis of other sites like the omentum, lung and liver remain extremely rare.[4,5]

We present a case of hepatic enterobiasis mimicking malignancy. This is the sixth case in international literature[2–6] and only the second one where modern imaging of the lesion is available.[6] The extreme rarity of such cases, the unclear pathogenesis of the condition and the distinct scarcity of imaging available have prompted this case report.

2. Presentation of case

A 46-year old female presented to our unit with a one-month history of right subcostal pain that was exacerbated during nighttime and at exertion. Past medical history was unremarkable, apart from a total hysterectomy that she had undergone for high-grade cervical dysplasia five years previously. Physical examination revealed localized tenderness on palpation of the right subcostal area. All laboratory findings were normal, including a normal eosinophil count of 0.12×10^9/l.

Initial imaging consisted of an abdominal ultrasound scan, which revealed a $4 \times 4 \times 3$ cm hypo-echoic lesion in the diaphragmatic surface of segment VIII of the liver. This was followed up by a CT-scan which revealed a round, hypo-dense lesion, with no obvious communication to the surface of the liver (Fig. 1).

Due to the inconclusive findings of both ultrasound and CT scanning, MRI-imaging of the abdomen was performed (Fig. 2); this showed a 2.9 cm round lesion in segment VIII of the right lobe of the liver, with necrotic/cystic appearance and small contrast enhancement, suggestive of malignancy, either primary or metastatic. Subsequent imaging and laboratory investigations for primary malignancy were negative, while ultrasound guided core biopsy of the lesion was performed twice and revealed necrotic tissue suggestive of malignancy, as well as hepatic parenchyma with signs of cholestasis and periportal inflammation.

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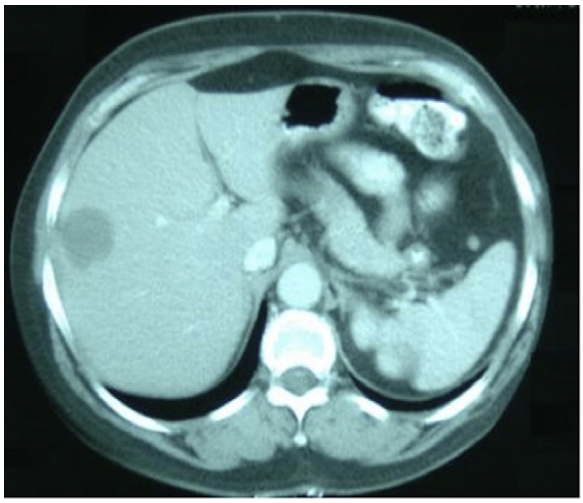

Fig. 1. An abdominal CT-scan revealed a low-density, round lesion in segment VIII of the liver.

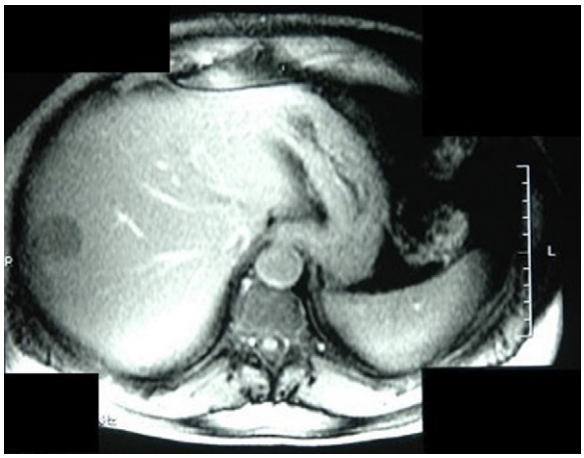

Fig. 2. A contrast MRI of the abdomen revealed a 2.9 cm round lesion in segment VIII of the liver, with cystic/necrotic appearance.

Following the suspicious imaging and pathological findings, open liver surgery was performed. The tumour was resected completely and the patient had an uneventful recovery. Histopathology surprisingly revealed multiple necrotic nodules containing structures consistent with *E. vermicularis* eggs. No evidence of malignancy was identified, nor where there other infectious microbes found in multiple stains of the specimen. The patient and all household members received appropriate treatment with mebendazole and are now well.

3. Discussion

Hepatic enterobiasis is extremely rare. Most of the previously reported cases were published before 1989,[2–5] which accounts for the distinct absence of imaging available. Furthermore, the "pinworm" nodules in those cases were incidental intra-operative findings during abdominal surgery for other diseases. This is easily explained, as pinworm liver infection appears to be mildly symptomatic at worst and because modern imaging techniques were obviously unavailable at the time.

Ng et al. have provided the most recent case report of hepatic enterobiasis in 2004.[6] Compared to that, our case shows interesting similarities: in both patients the presenting complaint appeared to be pain associated with the lesions that was dull and non-specific. Both lesions were located in segment VIII of the liver and both led to hepatic resections, as they raised suspicions of malignancy. CT-imaging of the lesions also revealed certain similarities: they were hypo-dense, located in segment VIII and rather close to the surface, however in the case of Ng et al. the lesion was multilobulated as opposed to round in our case. Ultrasound imaging was inconclusive in both cases and MRI imaging of our patient revealed cystic/necrotic characteristics, small contrast enhancement and was suggestive of malignancy.

Comparing data from all previous publications on hepatic *E. vermicularis* (Table 1), it appears that the helminth shows a preference to the right lobe of the liver, as documented in four out of five cases[2–6] (Little et al. do not state the location of the lesion). Furthermore, in all five cases the lesions were in very close proximity to the liver's surface,[2–5] or indeed "communicating" with it.[6] In all patients except ours, and that of Ng et al., extensive unhealthy colonic tissue was present. Our patient had a total hysterectomy 5 years prior to presentation. Ng et al. do not report any previous abdominal or urogenital surgery.

Regarding the exact mechanism of hepatic involvement, two main hypotheses have been suggested so far: haematogenous spread or direct migration, either through unhealthy intestinal tissues in both males and females or via the urogenital tract specifically in females. The migratory potential of *E. vermicularis* has been previously documented, especially through the female urogenital tract from the vagina to the ovaries, but the helminth is not believed to be able to penetrate healthy tissue.[4,5] Šlais, who was the first to report hepatic enterobiasis, presented a case where a nematode was found in a branch of the portal vein in a patient who died from "retroperitoneal phlegmon and diffuse peritonitis".[2] Based on that and their own findings, Little et al. were stronger supporters of the haematogenous spread; they suggested the nematode could have entered a mesenteric vein from a diseased part of the colon and from there carried to the liver by means of portal circulation.[3] In a later report however, Daly et al. considered direct migration as more likely than haematogenous spread, based on the "unnatural migration of an approximately 1 cm long, stout, adult helminth moving through such a narrow and circuitous channel to reach the liver".[4] In contrast, Mondou et al. did not believe that direct passage of the helminth through intestinal wall was likely, despite their patient having undergone surgery for resection of a large colonic villous adenoma.[5] Ng et al. reported that the aetiology of their patient's hepatic enterobiasis was "entirely unclear".[4] In their case, the patient had no history of gastrointestinal or urogenital surgery.

In the present report, our patient had no history of surgery of the gastrointestinal tract, which does not support the theory of migration through unhealthy colonic wall. However, she did have a total hysterectomy five years previously; if, at that time, colonic enterobiasis was present and the nematodes had already migrated to the urogenital tract, then the hysterectomy could have provided the helminths with a portal into the peritoneal cavity, where they could have established an asymptomatic, ectopic deposit in the liver, although we believe this hypothesis to be less likely than the haematogenous spread of the disease. Indeed, based on our clinical findings and review of available literature, we believe that in our patient the haematogenous route appears to be more likely, unless the ability of *E. vermicularis* to penetrate healthy tissue is greater than currently believed.

Table 1
A comparison of all previously published data on hepatic *E. vermicularis* infections.

Authors	Year published	Patient age/gender	Presenting symptoms	Imaging: USS	Imaging: CT	Imaging: MRI	Location of lesion in liver	Histopathology	Suggested transmission route
Arkoulis et al. (current article)	2011	46, female	One month history of mild right upper quadrant pain	Round, hypoechoic lesion	Low-density, round lesion, in segment VIII, no communication to surface of liver	Spherical lesion, cystic/necrotic characteristics, segment VIII, suggestive of malignancy	Segment VIII, right lobe	Multiple necrotic nodules containing *E. vermicularis*' eggs	Haematogenous more likely
Ng et al.	2004	51, female	One month history of right inframammary chest pain	Ill-defined lesion	low-density, multilobulated lesion, segment VIII, possible communication to surface	n/a	Segment VIII, right lobe	Multiple necrotic nodules containing fragments of *E. vermicularis*' larvae	Not given; unclear
Mondou et al.	1989	74, male	n/a – incidental finding during resection of colonic mass	n/a	n/a	n/a	"Right anterior segment of the liver"	Female *E. vermicularis* nematode in lesion, with numerous ova within the helminth	Direct migration considered unlikely
Daly et al.	1984	62, female	n/a – incidental finding during resection of colonic cancer	n/a	n/a	n/a	Not mentioned	Oval, partially hyalinized granuloma containing remnants of a pinworm with numerous eggs	Haematogenous or through direct migration through unhealthy colonic tissue
Little et al.	1973	54, male	n/a – incidental finding during resection of colonic cancer	n/a	n/a	n/a	"Near the edge of the right lobe of the liver"	Nodule surrounded by thick fibrous outer wall and amorphous necrotic centre with multiple *E. vermicularis*' eggs	Haematogenous most likely; direct migration unlikely
Slais et al.	1963	57, male	n/a – patient died from retroperitoneal phlegmon and peritonitis	n/a	n/a	n/a	"Ventral edge of right lobe of the liver"	Nodule with *E. vermicularis* in centre, necrosis of surrounding tissues, embolism of the peripheral branches of the portal vein	Haematogenous but no mechanism given

4. Conclusion

In this age of readily available high resolution imaging techniques and reduced post-operative morbidity and mortality following segmental hepatectomies, solitary tumour resection appears to be an easy option for both surgeon and patient. However, identifying a hepatic nodule as an *E. vermicularis* granuloma is very important, as it could prevent unnecessary surgery. Unfortunately, there do not seem to be any clinical, imaging or laboratory findings that are pathognomonic for this condition. The existing case reports are very scarce and comparison between them cannot produce statistically significant results; certain trends can be identified, but more data will need to be published to allow for better understanding of this condition.

Conflict of interest statement

No conflicts of interest.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions

1. Mr. Nikolaos Arkoulis (lead author): data collection, literature review and analysis and composing the paper
2. Dr. Helen Zerbinis: data collection and assistance with the paper revision
3. Dr. Georgios Simatos: supervisory and supporting role
4. Dr. Athanasios Nisiotis: Chief of Surgery, general supervision of task

Contributors

Dr. Maria Demonakou, Consultant, Department of Pathology, Sismanogleion General Hospital of Athens: responsible for pathological diagnosis of condition

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