

Case Report

A large adnexal tumor caused by *Enterobius vermicularis* mimicking malignancy

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Summary

Enterobius vermicularis usually causes trivial infections in the juvenile population. However, its extragenital presentation in adults is relatively rare. We present the case of a 64-year-old female suffering from poorly controlled diabetes and lower abdominal pain. CT scan showed a large tumorous expansion of the lower abdomen, mimicking malignancy. Perioperative findings revealed a large adnexal tumor adhering to the rectum. In addition, the histological examination uncovered a mixed inflammatory infiltrate with multiple surrounding eggs of the parasite and granulomatous reaction in the left fallopian tube and left ovarian cortex. As reported in our article, the rare ectopic sites of *Enterobius vermicularis* in postmenopause may become a diagnostic challenge.

Keywords: *Enterobius vermicularis*; extraintestinal; abdominal pain, adnexal tumor; postmenopause, malignancy

Introduction

Enterobius vermicularis is the most prevalent ubiquitous human extracellular endoparasite. The oldest paleoparasitological findings of *E. vermicularis* eggs in coprolites are from around 7837 BC and were found in different parts of the world (Paknazhad *et al.*, 2016). Oxyuriasis occurs primarily in children, and reinfections are common. Recent data from Poland showed a 4 – 32 % prevalence in young school children using molecular analysis and PCR, where 43 % of the infected children were asymptomatic (Kubiak *et al.*, 2017). Symptomatic infection is associated with considerable psychological distress for children and their caregivers (Wendt *et al.*, 2019).

The life cycle extends up to 2 months. After ingestion, the eggs hatch in the stomach and small intestine; the larvae migrate to the ileum, caecum, or appendix and take 15 days to mature. When fertilized worm migrates to the anus, up to 11 000 eggs are expelled.

Transmission methods are direct spread, scratching, autoinfection, or exposure to viable eggs from environmental objects. (Caldwell, 1982) Humans are the exclusive hosts; no evidence of domestic animals or canine reservoirs exists. Extraintestinal oxyuriasis can cause vaginitis and may extend to the uterus and fallopian tubes, reaching the ovaries and peritoneum in females (Cook, 1994). *E. vermicularis* is often histologically observed in the lumen of the appendix; however, it is usually not a cause of acute inflammation, as described by Mendoza *et al.* (1987).

Case Report

A 64-year-old female was referred to the outpatient department of the Bulovka University Hospital for dull lower abdominal pain. She had not undergone any gynecological examinations within the last six years. Clinical history revealed no risks for hereditary oncological diseases. However, she had poorly controlled type II

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diabetes mellitus requiring insulin therapy. On physical examination, the left abdomen was filled with a solid mass, partially fixed to the pelvis. Ultrasonography revealed tumorous infiltration of the left lower abdomen of dimensions 85x40x56 mm. According to the international ovarian tumor analysis–assessment (IOTA-ADNEX) the calculated risk malignancy was 34.5 % with possible infiltration of related organs (Albramowicz J. *et al.*, 2017). An abdominal CT scan (Fig. 1) showed a solid cystic, septated, and expansive lesion in the left adnexal region that measured 71x66x54 mm, with irregular margins and left uterine horn infiltration. The mass was located ventrally and caudally on the urinary bladder, where a lateral border was determined by the external iliac vessels. The left ureter laied dorsally to the tumor. Another lesion of similar characteristics was located cranially in the presacral region, measuring

29x20x23 mm and attached to the larger mass. Lymphatic nodes in the retroperitoneum were multiplied. Laboratory parameters showed hyperglycemia and elevated glycosylated hemoglobin. There were no signs of eosinophilia or immune system impairment; tumor markers were negative.

After the evaluation by the oncogynecologic team, a hysterectomy with bilateral salpingo-oophorectomy, tumor extirpation, and appendectomy was performed.

The definitive histopathological analysis confirmed an atrophic uterus and atrophic right adnexa. The left fallopian tube measured 7 cm, and the fimbrial part was distended with multiple adhesions to the enlarged left ovary. Histology images showed multiple *E. vermicularis* eggs, both solitary and in clusters, present in the lumen and wall of the left tube and the ovarian cortex (Fig. 2). The

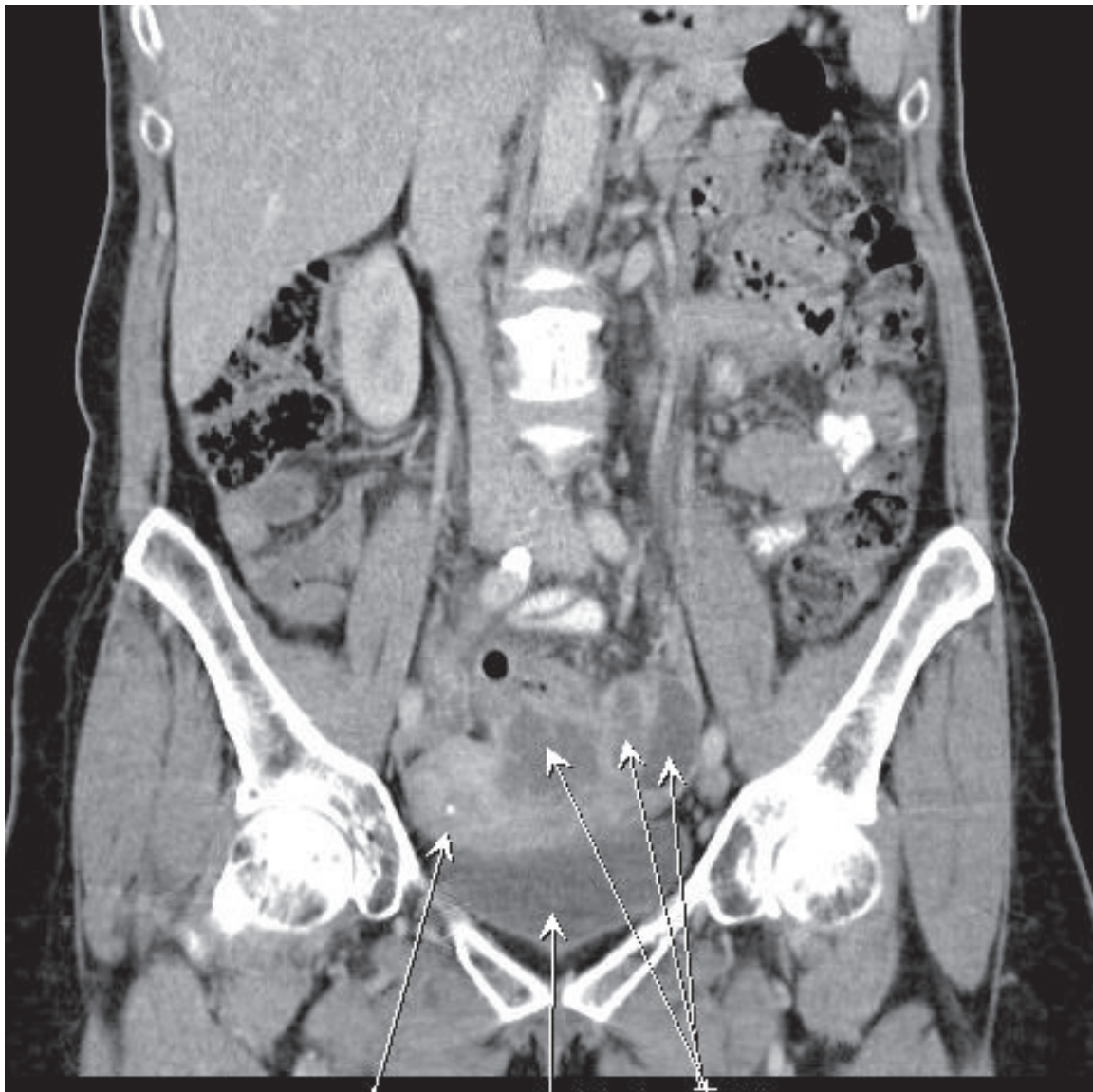


Fig. 1. CT scan revealed solid cystic, septated, and expansive lesion in the left adnexal region, with irregular margins and left uterine horn infiltration.

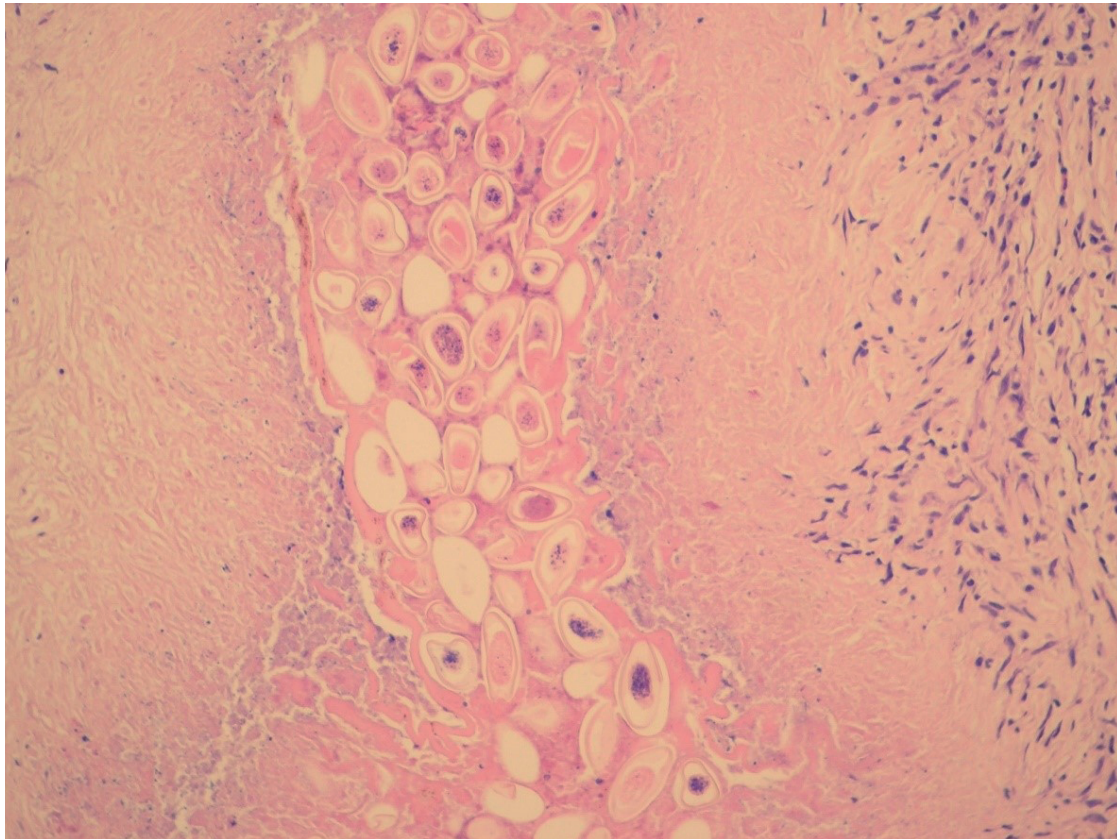


Fig. 2. *Enterobius vermicularis* visible inside the ovarian cortex; multiple eggs surrounded by granulomatous reactions and chronic inflammatory infiltrate.

eggs were surrounded by granulomatous reaction and chronic inflammatory infiltration with the presence of numerous eosinophils, erythrocytes, and fibrosis. An adult *E. vermicularis* and multiple eggs in the lumen were found on the cross-section of the appendix. The patient was discharged from the hospital 10 days later with a final diagnosis of inflammatory parasitic pseudocyst of the left adnexa and enterobiasis of the appendix. The stool sample was sent to the laboratory before the patient's discharge with a negative result.

Ethical Approval and/or Informed Consent

Consent was obtained from the patient for publication of this report and all accompanying images. The host institution ruled that the approval of the Ethics Committee was not required for this manuscript.

Discussion

Enterobiasis is Europe's most common parasitic disease that predominantly affects the juvenile population. The extraintestinal presence of *E. vermicularis* is well-reported. Recurrence of vaginal enterobiasis, despite complete treatment or absence of any gastrointestinal involvement, suggests that the vagina is a potential

reservoir for *E.v.*, which supports the theory of rare ectopic enterobiasis through the ascending pathway in the female genital tract (Ngu *et al.*, 2014). Nevertheless, we believe that ovarian cortex involvement is sporadic.

In symptomatic cases, *E.v.* causes vaginal bleeding, infertility, and peritoneal adhesions can lead to diffuse peritonitis and tubal ovarian abscesses responsible for the hysterectomies or ovariectomies (Mendoza *et al.*, 1987). The *E.v.* has been reported to form nodules in the fallopian tube and lead to the setting of ectopic pregnancy (Pampiglione & Rivasi, 2009). The presence of two *E. vermicularis* parasites invading macerated embryo was found in the tissue from an endometrial curettage performed for missed abortion in a pregnant woman (Mendoza *et al.*, 1987). Reipen *et al.* (2012) published a case report regarding cyclical lower-abdominal endometriosis-like painful symptoms in a woman of reproductive age. There is a paucity of literature regarding *E. vermicularis* complications in postmenopause. Al-Rufaie *et al.* (1998) described pinworm infestation of the uterus as postmenopausal bleeding. An accidental finding of the *E. vermicularis* in the endometrium of a 90-year-old patient was also reported by Pigac. *et al.* (2017). Another case described a pinworm granuloma associated with the uterine cervix squamous carcinoma. Additionally, bilateral tubal-ovarian abscesses with diffuse peritonitis were found in a 57-year-old woman (Pampiglione & Rivasi, 2009). Invasive systemic infections do not

occur even in severely immunosuppressed patients. Therefore, we presume that the hyperglycemia in our patient was not related to extragenital enterobiasis. Anthelmintics, such as mebendazole, pyrantel embonate, pyvinium embonate, albendazole, or ivermectin, are all capable of treatment against *E. vermicularis*. Additionally, young pinworms may resist some drugs (Wendt *et al.*, 2019). In our case, mebendazole 100 mg twice daily for 3 days, with a subsequent repetitive regimen in 14 and 28 days, was recommended, and all household members were treated. There are currently no other human anthelmintics available in the Czech Republic. Unfortunately, the prevalence of *E. vermicularis* in the adult population is not widely studied. A recent study by Stoyanova *et al.* 2019 showed that the overall prevalence of enterobiasis in Bulgaria was 0.91 %. In children, enterobiasis was significantly more prevalent – 1.49 % (CI:1.30 – 1.71), than in adults, 0.25 % (CI:0.17 – 0.36). Even though most infections appear harmless or asymptomatic, there are still some clinical challenges related to their extraintestinal presentation. Clinicians must be aware of the differential diagnosis, as unrecognized pinworm infections may impede the diagnostic process mimicking malignancy and lead to severe prognostic implications.

Conflict of Interest

The authors have no conflict of interest.

Acknowledgment

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