Genital schistosomiasis mansoni: tubal tumor and parietal peritoneum involvement diagnosed during laparoscopy

Esquistossomose mansônica genital: tumor de trompa e envolvimento do peritônio parietal diagnosticados durante laparoscopia

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ABSTRACT

Female genital schistosomiasis is not uncommon in endemic areas for schistosomiasis, but there are few reports in the Brazilian medical literature. Here, we describe the case of a 31-year-old woman with lower abdominal pain who was diagnosed as presenting a fallopian tube tumor caused by Manson's schistosomiasis. The diagnosis was delayed because her symptoms were considered nonspecific. Involvement of the parietal peritoneum of the ovarian fossa was observed during laparoscopy and confirmed by histological analysis. The left tube and the tumor were excised and schistosomiasis was treated with praziquantel. She presented a full recovery and options for future reproduction are under evaluation.

Key-words: Schistosomiasis. Fallopian tube. Genital schistosomiasis. Tumor. Schistosomiasis mansoni.

RESUMO

Esquistossomose genital feminina não é incomum em áreas endêmicas para esquistossomose, mas, há poucos relatos na literatura médica brasileira. Descrevemos aqui, o caso de uma paciente de 31 anos com dor abdominal no baixo ventre que recebeu o diagnóstico de tumor da trompa de falópio causada pela esquistossomose mansônica. O diagnóstico foi retardado porque os sintomas foram considerados inespecíficos. Havia envolvimento do peritônio parietal da fossa ovárica durante a laparoscopia, confirmado à histologia. A tuba esquerda e o tumor foram extirpados e a esquistossomose foi tratada com praziquantel. A paciente apresentou recuperação completa e opções para reprodução futura estão sob avaliação.

Palavras-chaves: Esquistossomose. Trompa de Falópio. Esquistossomose genital. Tumor. Esquistosomose mansoni.

There are several reports indicating that genital lesions in *Schistosoma mansoni* infection are not uncommon[1 7 12 13]. It has been estimated that 6 to 27% of girls and women with intestinal schistosomiasis suffer from pathological conditions induced by eggs sequestered somewhere in their genital organs. Female genital schistosomiasis is also often misdiagnosed as a sexually transmitted disease[33]. Recently, the World Health Organization decided to include female genital schistosomiasis in the group of gender-specific diseases that deserve high priority research.

Data on the topographic distribution of genital lesions reported from Brazil and other countries have shown the involvement of the following organs, ranked in descending order of importance: ovaries, cervix uteri, uterus, fallopian tubes, vulva and vagina[14 32]. In contrast with infections due to *Schistosoma haematobium*, there are few data concerning *Schistosoma mansoni*-induced genital lesions. In Brazil, the literature abounded with case reports in the 1940s and 1950s, i.e. during the period when schistosomiasis received considerable scientific and public health attention for the first time. Thereafter, there was almost total silence in the literature in relation to this subject, but it seems to have again been attracting increasing attention more recently[6 12 25 28 29].

Genital schistosomiasis has been explained in terms of worm migration and embolization of passively transported eggs to vessels of this anatomical region. Evidence of migrating worms is clearly demonstrated by identifying copulating adult worms in histological sections from internal genital organs[13].

In a prospective study, Bland and Geldand[5] reported that 10 patients out of 104 from a country endemic for bilharzia showed definite evidence of bilharziasis in the fallopian tubes. The importance of schistosomiasis of the fallopian tubes is that it may predispose toward ectopic pregnancy and decreased fertility[4 6 11 20 30]. Armbrust[2] described a case in Brazil in which *Schistosoma mansoni* had caused severe tubal disease.

Here, we add another case of a patient with a tubal tumor and peritoneal involvement caused by Manson's schistosomiasis diagnosed during laparoscopy.

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CASE REPORT

A 31-year-old woman came to the hospital complaining of lower abdominal discomfort and hyporexia, which had started two months earlier. She had been using nonsteroidal anti-inflammatory agents for pain with partial relief. She visited three physicians during this period and was assured that her symptoms had no organic basis or explanation. In our hospital, a gynecological examination revealed tenderness during mobilization of the uterus and palpation of the left adnexus. There was no fever or tachycardia on admission. No pelvic or abdominal masses were palpable. Laboratory tests were negative for βHCG and normal for C-reactive protein, erythrocyte sedimentation rate, urine culture, hemogram and leukocyte count. Pelvic ultrasound showed a hypoechoic homogeneous mass of $3cm^3$ in the left paraovarian area. The uterus and ovaries were normal. With a probable diagnosis of tubo-ovarian abscess, she was treated for pelvic inflammatory disease with antibiotics (clindamycin and gentamicin). She improved over the next 24 hours but abdominal discomfort recurred thereafter. New laboratory tests did not help. However, another ultrasound revealed a larger adnexal mass of $22cm^3$ (**Figure 1**). During laparoscopy, a hard blackish tumoral mass containing blood with whitish spots, with a largest diameter of 4 cm, was observed (**Figure 2**). This mass adhered to the left fallopian tube, which was edematous and twisted. Both ovaries were normal. In both ovarian fossae, the peritoneum showed whitish granules (**Figure 3**). With a presumptive diagnosis of tubal pregnancy, the left tube and ovary were excised together with the tumoral mass. Biopsies of the ovarian fossae were also taken. She improved quickly after the procedure and was discharged from hospital two days later. Histological examination of the surgical material revealed the presence of *Schistosoma mansoni* eggs in the tumoral mass (**Figure 4**), uterine tube (with salpingitis) and ovarian fossae (**Figure 5**). Ultrasound of the liver did not reveal any evidence of schistosomal liver fibrosis or signs of portal hypertension. Stool examination and rectal biopsy resulted negative for *Schistosoma mansoni* eggs. One month later, she was treated for schistosomiasis with praziquantel (50mg/kg of body weight, in a single dose). She is being followed up at the outpatient clinic and, over the initial six months after hospital discharge, has had no further complaints.

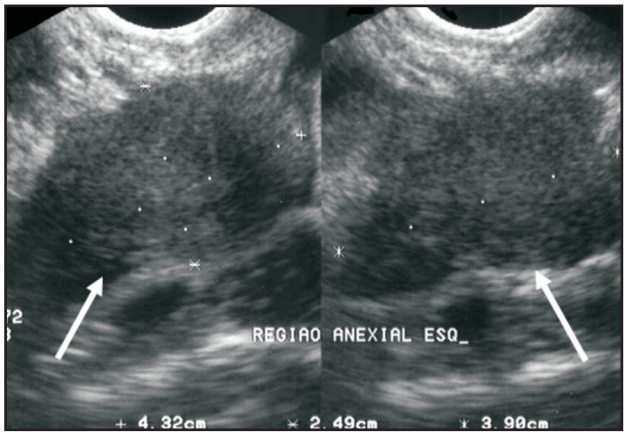

FIGURE 1

Ultrasound: a tumoral mass in the left adnexa (white arrows).

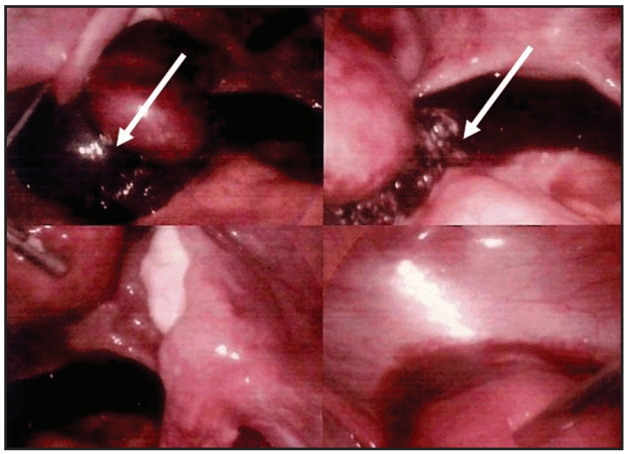

FIGURE 2

Laparoscopy: tumoral mass in left tube (arrows).

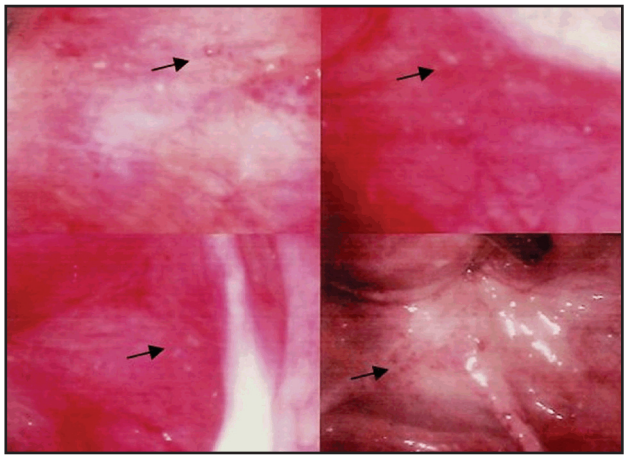

FIGURE 3

Laparoscopy: ovarian fossa with granules (arrows).

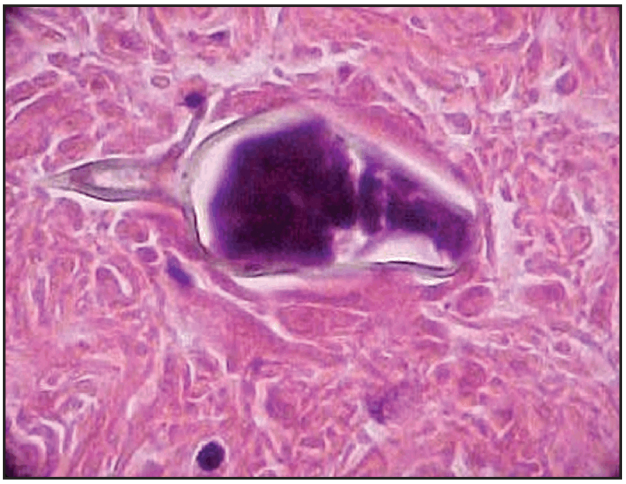

FIGURE 4

A *Schistosoma mansoni* egg in the left fallopian tube.

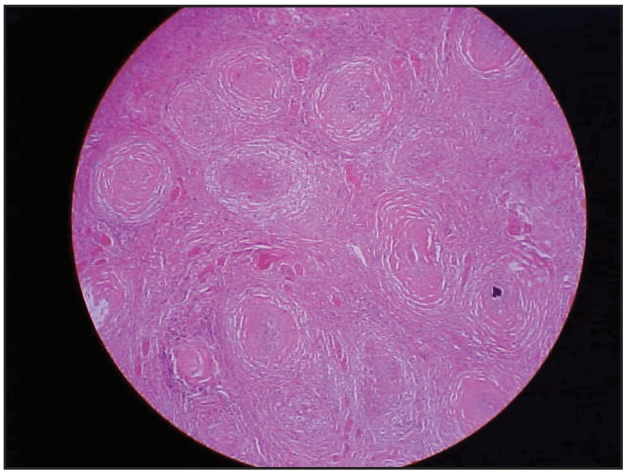

FIGURE 5

Granulomas in the peritoneum of the left ovarian fossa.

DISCUSSION

It took some time for physicians to investigate the hypogastric pain of this patient. She was anxious because her marriage was due to take place six months after the symptoms started. Anxiety was her first diagnosis. After genital schistosomiasis was diagnosed and treated with praziquantel, she was worried about her chances of having a baby because she had lost the left tube and schistosomiasis had spread to the right ovarian fossa. This patient was referred to a reproductive endocrinology clinic to discuss her options for future fertility. Infertility and ectopic pregnancy are real threats to such patients and have been acknowledged by investigators since the time of the first case descriptions[4 8 10 15 16 35].

In a recent review of 83 cases of genital Manson's schistosomiasis, it was observed that the ovaries were the main organs affected (28/83; 33%), followed by the cervix (20/83; 25%), fallopian tubes (14/83; 17%), uterus (15/83; 18%) and vulva (5/83; 6%)[12 26 27]. The primary involvement in our patient was in the left fallopian tube, but it is worth bearing in mind that granuloma had spread to the parietal peritoneum in this area. No worms were found in the vessels, but eggs of *Schistosoma mansoni* with and without granuloma formation were readily encountered, particularly in the ovarian fossa (**Figure 5**). The presence of worms in the genital area, including the skin, has been described both in male and in female patients[2 3 6 7 14 15 25 30 34].

Stool examination and rectal biopsy did not reveal any worm eggs in our patient. Such findings have been reported previously and this reinforces the need to maintain a high index of suspicion of schistosomiasis among patients who live in endemic areas or have recently traveled to endemic areas[18 21 26]. In our case, it was impossible to assess whether the schistosomiasis had been cured after treatment with praziquantel because stool examination and rectal biopsy were of no use for diagnosis.

The second ultrasound performed on our patient suggested the presence of an ovarian abscess. Ovarian abscesses are associated with pelvic inflammatory diseases caused by bacteria. The clinical picture frequently includes the presence of fever and laboratory abnormalities (leukocytosis and C-reactive protein augmentation in serum), but our patient had no evidence of bacterial infection[19 24].

The numbers of descriptions of cases of genital schistosomiasis in Brazil declined from the 1960s onwards. No sound explanation for this has been offered so far. However, it is well known that before the start of the Brazilian Program for Schistosomiasis Control in the 1970s, Brazilian scientists and the Ministry of Health decided that hepatosplenic schistosomiasis should be considered to be the most severe form of Manson's schistosomiasis. Thus, diminishing or eliminating this form of the disease was the main priority of the control program[9 20]. Many aspects of the disease, such as neuroschistosomiasis, glomerulonephritis, pulmonary hypertension, liver abscess, acute schistosomiasis and *Salmonella* bacteremia, among others, were left aside[17 21 23]. It is possible that genital involvement may have also been neglected.

The patient in the present study did not have any periportal fibrosis (as investigated by ultrasound) or any evidence of portal hypertension. Hence, she had hepatointestinal schistosomiasis and severe disease of the genital organs. Fortunately, the Brazilian Ministry of Health has changed its view about the disease and, at the latest meeting of the Schistosomiasis Control Committee, it was decided to encourage healthcare personnel to evaluate and report on the neglected aspects of this disease.

Summing up, we have presented here the case of a patient with tubal and peritoneal Manson's schistosomiasis who, over a two-month period, evolved with vague pain in the lower abdomen that represented a diagnostic challenge for the attending physicians. The left fallopian tube was excised and schistosomiasis was treated with praziquantel. Healthcare personnel should be aware of this condition and look for a diagnosis of genital schistosomiasis among patients from endemic areas.

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