Schistosomiasis of the abdominal cavity and infertility: a case report

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Abstract

Introduction
Schistosomiasis was first described in Cairo in 1852 by Theodor Bilharz. The disease commonly called bilharziasis in endemic zones, is caused by Schistosoma haematobium and affects over 150 million people in Africa and the Middle East. It is the fourth most prevalent disease in the world. The intermediate hosts are aquatic, sinistral snails, which breed prolifically in any still or slowly moving fresh water. It comprises a group of chronic diseases caused by schistosomes, a genus of digenetic parasitic worms. One species of these trematodes, S. haematobium, immigrates principally in the paravesical venous plexus in humans and causes urinary schistosomiasis (bilharziasis), and is now considered a major public health problem in most areas in which it is significantly endemic. Infection is acquired by contact with still or slowly flowing water contaminated by infected urine or faeces. The adults of S. haematobium are widely distributed throughout the pelvic and mesenteric venous plexuses and oviposition usually occurs in the pelvic organs especially in the urinary bladder, ureters, urethra, prostate and lower gastrointestinal tract. In female genital organs, ovaries and fallopian tubes sometimes have high egg burdens, but the mean eggs burdens in autopsy series are low. Egg burdens of the uterus and vagina are even lower. The aim of this study is to report a case of schistosomiasis of the abdominal cavity and infertility.

Case report
A 35-year-old British nulliparous woman was referred to our Gynaecology Clinic complaining of primary infertility having tried to conceive for the past three years. Several years ago she travelled extensively in Thailand, India, East and West Africa, South America and Caribbean. Of particular note is that she has spent one week swimming 3–4 times, at Lake Malawi. She has been married for two years and temporarily living in New Zealand and returned to the United Kingdom. She has got a past history of eczema and she describes also a recent fundal infection in the groin and axilla and one of her eyelids treated by her GP with steroids. No other important problem was noted from her previous medical or surgical history. Especially, she had no history of pelvic infection and came off the contraceptive pill nearly four years ago. She has had a BCG vaccination in the past. Her abdominal and pelvic examinations were normal and in particular there has been no vaginal discharge, abnormal uterine bleeding, dyspareunia, haematuria or urinary frequency. Her periods were regular with bleeding for 3–5 days out of every 25–26 days but have been quite irregular since her last operation in August 1999. On a pelvic ultrasound, several follicles were present in both ovaries (two on the right and three on the left), and were normal in size and appearance and in conclusion this was unremarkable. Standard infertility hormone investigations, LFTs, U and Es, semen analysis all proved to be normal. Her repeat blood counts were normal and especially her eosinophils count. Gonococcal and chlamydial cervical cultures were negative. Colposcopy showed a white deposit on the cervix. This was biopsied and showed a granuloma. Within this granuloma was an ovoid foreign body although no embryo was identified which may represent a degenerate parasite egg. The Pap test that was performed was
negative. No colposcopic abnormalities were found at a 6-month follow-up examination.

She had a hysteroscopy, laparoscopy and dye test. Inside the uterine cavity were no abnormal findings but looking inside the peritoneal cavity there were multiple 1–2 mm raised white spots and also peritubal, ovarian and large bowel dense adhesions (Figure 1). These patches were related to the right fallopian tube and ovary (Figures 2 and 3) and up the right paracholic gutter and on the liver and omentum (Figures 4–8). Several of these deposits were removed laparoscopically and sent for histology. Peritoneal washing results revealed no pathognomonic findings. Our first impression was that this would be suggestive of intraperitoneal tuberculosis. A dye test was performed and showed bilateral tubal blockage which was probably distal. A Mantoux test was done and the lesion was about 2 cm in diameter not ulcerated and indurated. C-reactive protein and ESR tests were normal. Her chest X-ray was normal. The histology result of these deposits showed that there were chronic granulomae and not caseation or acid-fast bacilli but within one of these granulomas there appeared to be a S. haematobium parasite (Figure 9a and b).

Figure 1: Laparoscopic picture of raised white spots and also peritubal, ovarian and large bowel dense adhesions due to chronic scistosomal tissue alterations.

Figures 2 and 3: Deposits at the right fallopian tube and ovary.

Figures 4–8: Deposits at the paracholic gutter, liver and omentum.
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The recent blood serology result also came back positive for schistosomiasis. The schistosoma enzyme-linked immunosorbent assay (ELISA) level was four. No Schistosoma ova were seen in a late morning urine specimen neither in a sample of faeces. The proven schistosomiasis was treated with 20 mg/kg Praziquantel taken twice daily for one day only and repeated schistosomal serology was suggested to her in one year’s time. After this treatment, a hysterosalpingogram and then laparoscopic adhesiolysis and salpingostomy were arranged to continue our management of her primary infertility (Figures 10–13). The hysterosalpingogram revealed that the right tube filled with dye but due to a blockage at the end, there was no spill. At the surgery, the left fallopian tube was checked and was normal and patent but at the right tube hydrosalpinx was found and a distal neosalpingostomy done. Liver and bowel adhesions on the right side to the anterior abdominal wall and right ovary were lysed and tubal adhesions divided. The right tube was reopened, normal fimbriae were observed but no fill or spill of the dye was seen. The uterus was normal and interestingly no white deposits were noted throughout the peritoneal cavity. Finally, during the cystoscopic evaluation, the bladder mucosa was smooth and pink and ureteral orifices normally positioned.

A repeat semen analysis was also normal and in view of the patient’s irregular periods she commenced Clomiphene 100 mg from Day 2–6 for a period of 6 months. The results of Day 21 progesterone level on three consecutive cycles showed she was ovulating. A sonohysterography at Day 5 of her menstrual cycle took place 8 months after the laparoscopic surgery and revealed an early menstrual endometrium (single hyperechoic line). There were no sonographic findings of the ovaries and myometrium but unfortunately no fluid in the pouch of Douglas was found after this. The transvaginal sonogram of the bladder during the same session did not show any focal thickening or any calcifications of the wall and the margins of the bladder wall appeared intact.

Discussion

Occasional cases of asymptomatic schistosomal oophoritis and salpingitis like our case probably represent an atypically distributed oviposition rather than generalised schistosomal infection. The fallopian tube obliteration may be caused by a concentric tubal muscle fibrosis or hypertrophy or sandy patches proximal to an obstruction. The adult worm pairs characterise active schistosomiasis, oviposition and a vigorous granulomatus host response. Intensity of infection is assessed directly by quantitating the worm burden at pathology examination and indirectly by determining the tissue egg burden and egg count. The abnormal cervical stroma in our patient was also suggestive of schistosomal infection. The granulomas seen in the cervix (Figure 9) may represent an atypically distributed oviposition rather than focal/degenerative processes. Schistosomal adhesions and tubal fibrosis may be responsible for infertility in our patient. Cysts seen on ultrasound may represent schistosomal oviposition. The adult worm pairs characterise active schistosomiasis, oviposition and a vigorous granulomatous response. Intensity of infection is assessed directly by quantitating the worm burden at pathology examination and indirectly by determining the tissue egg burden and egg count. The abnormal cervical stroma in our patient was also suggestive of schistosomal infection.

Figure 9: Single granuloma with an apparent focus of calcification representing a schistosoma, (a) low magnification; (b) high magnification.

burden which is a product of intensity and the duration of infection. The active stage is epidemiologically important because of its role in transmission. It is clinically important because during this stage this destructive process can be dramatically altered by applying chemotherapy converting active into inactive disease. Adult worms may live for up to 12 years. Inactive schistosomiasis, which occurs when adult worms have died, is characterised by the absence of viable eggs in tissues. Marked adhesion formation and salpingitis as observed in our case report, may be the effect of the early-stage disease ova deposition in the terminal veins of the fallopian tubes and ovaries and is associated with decreased blood supply as ischaemia plays an important role in adhesion formation. Endometrium is an uncommon host for the parasite because the ova with the characteristic long terminal spine are unable to pass through the tortuous vein system in the myometrium but eggs can be found in the subserosa of the uterus. The disease also affects the cervix. Reports suggest the cervix is not an uncommon site of the disease and when present it can be diagnosed by cytology. The cervical smear shows that among inflammatory cells there are a number of S. haematobium ova in various stages of development. Colposcopy usually reveals cream-coloured nodules within the squamous and metaplastic epithelium of the ectocervix. Acetic acid application makes the nodules centrally dark with a surrounding pale halo and also Shiller’s solution uptake is reduced at these sites. Punch biopsy from cervical nodule contains S. haematobium ova surrounded by granulomatous reaction and also sometimes a high-grade cervical intraepithelial neoplasia. Different types of tissue reaction except the previous one have been described reflecting the chronic natural history of the disease from granulomatous to fibrotic and acellular condition. We think as the eggs deposit and after years they die and calcify, more metaplasia of the squamous cells leads to a squamous carcinoma but cancer development probably requires the additional presence of the human papilloma virus. In the presence of pelvic schistosomiasis cervical biopsies should be obtained as bilateral cervical pathology was found in more than 50% of the patients. The vulva and vagina must be carefully inspected as these are also commonly infected. The patient’s history of visits in endemic areas as a back-packer; swimming repeatedly in lakes and streams underscores the importance of a complete history, especially if we are searching for an infectious cause. Sometimes the patient’s illness has two faces, one urologic and the other, gynaecologic. The bladder is the most affected organ in schistosomiasis and lesions are mainly occurring in the trigone and bladder’s neck. The most common complication of urinary schistosomiasis is obstructive uropathy, which usually results from involvement of the interstitial portion of the ureters. Antischistosomal chemotherapy during active disease may result in remarkable decrease of the lesions and their local effects. Two stages of this are well recognised: (a) the active stage, during which, after the worms penetrate the human skin (swimmer’s itch), there is abundant egg deposition and (b) the inactive stage, at this point there are very few or no live/dead eggs in the most common areas. The proportion of patients with inactive disease increases with age and since the time of primary infection. We suspect that our patient was infected during her visit to Africa. Haematuria does not usually occur during the inactive phase of infection. Even during the active phase haematuria has been reported in only 35% of patients with documented infection. The level of proteinuria correlates with the intensity of infection. Our patient’s urine specimen was negative for protein but it is well documented that secondary bacterial infection is common in schistosomiasis and also the absence of eggs in the urine does not stipulate an inactive phase of the infection. It is essential to obtain 24-hour urine specimen or at least urine collection must be done between 10 am and 2 pm, because of the diurnal periodicity of egg excretion. Histopathologically, biopsies obtained from sandy spots or nodules show a characteristic active or inactive lesion. Active lesions are yellow and are characterised by Schistosomal ova surrounded by giant cells, epithelioid cells, eosinophils and lymphocytes. Inactive (healed) lesions are greyish white, the ova are calcified and fragmented and cellular reaction is replaced by different degrees of fibrosis. Additionally, biopsies obtained from the surface of the ovaries did not reveal schistosomiasis. Schistosomal involvement of the female genital tract is manifested by granulomatous inflammation and fibrous adhesions which can lead to infertility. Thirteen cases of pelvic schistosomiasis were diagnosed by laparoscopic examination in Egypt who presented with infertility, and fertility was restored in 46% of these women after medical treatment with Niridazole. Tubo-ovarian adhesions, like in our case, also increases the risk of ectopic pregnancy. Although adhesions were also found in our patient’s liver and bowel, pathophysiologically important hepatic fibrosis or obstructive enteropathy is not associated. Urinary schistosomiasis has been linked to male subfertility. Prostate oviposition is generally low but the ejaculatory ducts passing through the prostate may bear heavy burdens. Seminal vesicle egg burdens correlate well with intensity of infection causing chronic lower urinary male tract disease. Several epidemiologic studies in regions where schistosomiasis is
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endemic have shown high frequency of squamous cell carcinoma and adenocarcinoma which has been associated with squamous metaplasia of the urothelium. Genital schistosomiasis in males has been observed among travellers complaining of haematuria, haematospermia and other changes in their ejaculate. In a community-based study of genital schistosomiasis, men showed detection of S. haematobium eggs in 43% of semen samples suggesting that the genital organs of men are frequently affected with schistosomiasis. During the inactive phase of infection haematuria in both sexes does not usually occur, accounting for our patient’s absence of haematuria and blood in her urine. Even during the active phase, haematuria has been reported in only 35% of patients with documented infection. ELISA is based on the detection of antigens circulating in the serum or excreted in the urine. Unless the serological-ELISA techniques prevalence in assessing schistosomiasis is higher than the parasitological they give no indication of the intensity of infection, do not distinguish between past and present infection, are not species-specific and require high and expensive technology. Improvement in sensitivity is needed for diagnosing mild infections and monitoring the efficacy of chemotherapy. Immunodiagnostic tests capable of detecting the presence of circulating antigens are more useful. Reports on travellers from such regions have been suggesting the last two decades that the disease might be increased in West-European countries, and therefore have been advising all people travelling in areas of endemic schistosomiasis to undergo schistosomiasis ELISA so that subclinical infections can be detected on time. However, do not distinguish between past and present infection, are not species-specific and require high and expensive technology. Improvement in sensitivity is needed for diagnosing mild infections and monitoring the efficacy of chemotherapy. Immunodiagnostic tests capable of detecting the presence of circulating antigens are more useful. Reports on travellers from such regions have been suggesting the last two decades that the disease might be increased in West-European countries, and therefore have been advising all people travelling in areas of endemic schistosomiasis to undergo schistosomiasis ELISA so that subclinical infections can be detected on time.

Case report

Although we were afraid for this woman having at least right-sided tubal disease, her best option for future fertility would be in vitro fertilisation; at 10 months following surgery, she conceived and gave birth to a healthy 3.1 kg male infant with normal delivery at full term.

Conclusion

Although we were afraid for this woman having at least right-sided tubal disease, her best option for future fertility would be in vitro fertilisation; at 10 months following surgery, she conceived and gave birth to a healthy 3.1 kg male infant with normal delivery at full term.

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.

References

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