

CASE REPORT

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Case report of tubo-ovarian Bilharziasis presented with pelvic pain and secondary infertility

Tonleu Linda Bentefouet, Marietou Thiam, Abdoul Magib Gaye, Amine El Wardi, Lamine Gueye, Mamadou Lamine Cisse

ABSTRACT

Introduction: We present a case of a patient with infertility in the context of tubo-ovarian schistosomiasis discovered at histology. This case supports the necessity to include schistosomiasis as a potential etiologic factor in infertility patients from the disease endemic regions. **Case Report:** A 22-year-old woman presented for gynecology consultation at the department for secondary infertility and pelvic pain lasting four years. Pelvic ultrasound revealed a right ovarian cyst. Right adnexectomy was then performed with a smooth postoperative period. The macroscopic examination revealed a large sized whitish ovarian cyst. The totality of ovary and the fallopian tubes was encrusted by the bilharzia eggs more or less calcified with terminal spines typical to *Schistosoma haematobium* species at histology. **Conclusion:** We hypothesize a link between the patient's infertility and the presence of *Schistosoma haematobium*. The latter is highly associated with the presence of the metabolite

catechol-estrogen/DNA adducts in patients' serum.

Keywords: Infertility, Serum biomarkers, Urogenital bilharziasis

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INTRODUCTION

Bilharziasis remains a public health problem, especially in tropical zones [1]. With the development of water management strategies, such as dams and irrigation, an increase in the incidence rate of this pathology is expected in the coming years. *Schistosoma haematobium* is the agent causing urogenital bilharziasis. However, *Schistosoma mansoni* and *Schistosoma japonicum* can also infect the genital tract [1]. A number of studies have incriminated genital Bilharziasis in sterility [2–4]. In addition to the medical situation, there is also a socio-cultural burden; procreation is critical in the African family and in the past the number of children was viewed as a sign of wealth and abundance and the absence of children as a misfortune. To the best of our knowledge, there has been no known study on the link between schistosomiasis and infertility, in Senegal. This

paper presents an infertility case due to tubo-ovarian schistosomiasis.

CASE REPORT

A 22-year-old woman with a regular menstrual cycle and no serious previous medical-surgical history, spent some time in the Senegal river region (Fouta). She is also prime gravida, prime para in her second marriage and mother to a healthy living child. She presented for consultation gynecology department by secondary infertility and pelvic pain for four years.

Clinical examination revealed a right sided tender pelvic mass not related to the uterus. Pelvic ultrasound revealed a right ovarian cyst measuring 7 cm in diameter (Figure 1).

Preoperative checks were within normal range. Right sided adnexectomy was then performed with a smooth post period. The specimen was sent for histological examination. The macroscopic examination revealed a large sized whitish ovarian cyst with an 8 cm grand axe length, a smooth external surface, and a renitent consistency. The dissection of the cyst revealed a unilocular cavity, an internal regular surface, a gelatinous content, and a thick inner wall. The whitish fallopian tubes measured 6x3 cm size with a 4-mm width thick wall (Figure 2).

Histology was performed using an optic microscope after staining the glass microscope slides with Hematoxylin and Eosin. The ovary and the fallopian tubes appeared very altered. The totality of the wall was encrusted by the bilharzia eggs more or less calcified with terminal spines specific to *Schistosoma haematobium* species, surrounded by a granulomatous inflammatory reaction rich in giant cells of the foreign body type (Figure 3). The parasitological examination of urine was normal.

DISCUSSION

Symmers was the first to describe genital schistosomiasis in 1906. In *Schistosoma haematobium* endemic zones, the genital schistosomiasis prevalence is 33–75% [5]. While the cervix is the most affected part of the genital tract in the majority of cases, but the ovaries, the fallopian tubes, the vagina and the vulva could also be affected [2, 4]. The link between schistosomiasis and male infertility has been studied in literature. Among males, testicle attacks have been the causes of secretory and excretory azoospermia [6, 7]. In these studies, the schistosomiasis histologic lesions were located on the testicles (82.9%), in the epididymis (14.3%) and 2.9% in the prostate.

As in this case study, because of no apparent signs of sickness, the genital schistosomiasis diagnostic is often the result of a fortuity situation and is generally made at a later stage; the patient initially consulted because of



Figure 1: Pelvic ultrasound showing a ovarian cyst.



Figure 2: Section removed following the hand-right adnexectomy showing unilocular ovarian cyst.

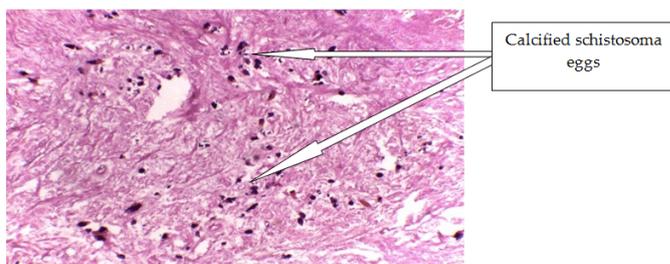


Figure 3: Calcified schistosoma eggs (H&E stain, x100).

non-specific genital signs that persisted for four years. The absence of screening and consistent care makes the genital schistosomiasis diagnosis difficult and delay effective treatment. In our case study, it is only after the histology examination that the genital schistosomiasis was suspected.

The histology lesions are linked to a toxic allergy and a mechanic action from the parasites and their eggs. At the macroscopic level, the lesions start like a papuloerythematous in disseminated granulation. At a later stage, the lesions cause polypoides tumefactions that are burgeoning, ulcerated, bleeding when touched

and often located on the cervix and vagina [8]. Other type of lesions such as ovarian cysts and tubo-ovarian adhesions has also been mentioned [4]. Similar to the case presented here, the thickness of the fallopian tube has been pointed out as a recurrent sign [8]. This sign of fallopian tube thickness should be considered as a warning sign and should raise the suspicion index to request histological examination of the salpingectomy specimen as well as a parasitology research for urinary schistosomiasis in endemic areas. The histologic lesions are dominated by the bilharzial granuloma with the presence of *Schistosoma* eggs in the tissue. The African pathologist should systematically look for bilharzia follicles and *Schistosoma* eggs when conducting the histological examination of the fallopian tubes. The lesions are located within the mucosa, under the mucous membranes and sometimes within the muscles of the urogenital organ [4, 8].

It is difficult to ascertain the infertility risk and eventually sterility caused by the schistosomiasis due to associated supra infection, and sexually transmitted diseases whose impact on infertility has been established. In this case, no other infections were found. As mentioned by other researchers, we speculate that presence of calcified eggs within the fallopian tube in additions to fibro-inflammatory changes could cause a partial narrowing of the fallopian tube which could then hamper the tubal peristalsis and natural evolution of the fertilized egg [4, 8].

Outside this consideration purely anatomic, recent studies from Nigeria and Angola have argued that the *Schistosoma haematobium* could be producing the catechol-estrogen/DNA adduct molecule detectable by mass spectrometry (LC/UV-DAD/ESI-MSn) [9, 10]. This metabolite isolated from the serum of infected persons could be at the origin of estrogen destabilization and infertility by playing on the alpha and beta receptors of estrogen sensitive cells in the genital sphere. Considering the statistically significant results obtained, we recommend the utilization of this molecule as a potential biomarker for infertility in women infected by *Schistosoma haematobium*.

CONCLUSION

Bilharziasis still constitutes a real public health problem in Africa, Asia, and South America, by the large numbers of individuals affected. In women living in endemic zones, one should not ignore bilharziasis in infertility cases. A detailed study in the association of bilharziasis and infertility needs to be carried out in our country.

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Author Contributions

Tonleu Linda Bentefouet – Substantial contribution to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Marietou Thiam – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Abdoul Magib Gaye – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Mamadou Lamine Cisse – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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