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Authors: Pauline Dioussé, Haby Dione, Mariama Bammo, Chérif Dial, Mariétou Thiam, Aissatou Amy Diamé, Bernard Marcel Diop, Mamadou Mourtalla Ka

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AUTHORS:
Pauline Dioussé ¹, MD
Haby Dione¹, MD
Mariama Bammo⁰, MD
Chérif Dial², MD
Mariétou Thiam³, MD
Aissatou Amy Diamé⁴, MD
Bernard Marcel Diop ³, MD
Mamadou Mourtalla Ka³, MD

AFFILIATIONS:
¹MD, Department of Dermatology-Venereology, Faculty of Health Sciences, Thies University, Thies, Senegal, paudiousse@yahoo.fr
²MD, Department of pathology, University Cheikh Anta Diop, Dakar, Senegal, dialcherif@yahoo.fr
³MD, Department of Health Sciences, Thies University, Senegal, bmdiopmi@yahoo.fr
⁴MD, Department of urology, regional hospital, Thies, Senegal, aadiame@hotmail.com

CORRESPONDING AUTHOR DETAILS
Pauline Dioussé
Complete Mailing Address: MD, Department of Dermatology-Venereology, Faculty of Health Sciences, Thies University, Thies, Senegal, Postal code: BP 34 A Thies, Senegal
Email: paudiousse@yahoo.fr
Short Running Title: Vulvar location is rare, the case of uro-digestive is even more so. We reported vulvar bilharzia, remarkable for its clinical presentation, concomitant urinary and gastrointestinal involvement and multidisciplinary management.

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ABSTRACT

Introduction
In Senegal, West Africa, the overall prevalence of schistosomiasis varies from 0.3 to 1%. A major focus of schistosomiasis urogenital is known around the banks of the Senegal River where the average infestation rate reached 71.8%. Vulvar location is rare, the case of uro-digestive is even more so. We report a case.

Case Report
It was a girl aged 9, from a village situated on the banks of the Senegal River, received for swollen labia lasting for 3 years. There was a notion of contact with the river water. At dermatological examination, she had an itchy swelling of the large labia, small labia, clitoris, the groin with a palpation papulo-nodular, fibrous. There was hyper eosinophilia. Dermal histology showed under an acanthotic epidermis, a granulomatous inflammatory reaction around the dermis of an infestation of viable eggs of Schistosoma species terminal spur characteristic of Schistosoma haematobium. The abdominal-pelvic ultrasound revealed bilateral ureteral hydronephrosis The CT urography without product injection noted an irregular parietal calcification of the bladder, ureters under pyelics, urethra, rectum, sigmoid, and the left colon with segmental dilatation of the ureters. She was put under praziquantel (40 mg / kg in two spaced taken 15 days). The outcome was favorable with a clear regression of swelling after 1 month. Resection of residual disease was made.

Conclusion
We reported a vulvar bilharzia, remarkable by a clinical presentation, an urogenital tract case and the multidisciplinary management.

Keywords: Schistosoma haematobium, vulvar, children
INTRODUCTION

Schistosomiasis or bilharzia is the second global parasitic disease after malaria, it affects 239 million people in 2009 and 80 to 90% of them live in Africa [1]. In Senegal, the overall prevalence of schistosomiasis varies from 0.3 to 1%. A major focus is known around the banks of the Senegal River where the average infestation rate reached 71.8% [2, 3]. Vulvar location is rare, few case series have been described in the literature [4, 5, 6, 7, 8]. It results in papules and nodular lesions. Treatment is primarily medical and for large lesions, it must always be followed by surgical resection of residual lesions. We report a case.

CASE REPORT

It was a girl aged 9, from a village situated on the banks of the Senegal River, received in dermatological consultation for swollen labia evolving for 3 years. She would get an urinary schistosomiasis 4 years ago. It was found in her personal history, frequent bathing in the river water. She was treated by unspecified drugs according to her parents. She had a good condition with a weight of 22 kg for a height of 1,26m with a BMI 13.90 kg / m2, corresponding to normal but close to the area of underweight build.

At dermatological examination, she had an itchy swelling of the big labia, small labia, clitoris, and groin folds of the aspect of papulo-fibrous and nodular on palpation (Figure 1). No abnormalities found in the gastro-intestinal exam. The rest of the examination was unremarkable. Diagnostic hypotheses were discussed: cutaneous calcinosis, vulvar tuberculosis, cutaneous sarcoidosis and cutaneous schistosomiasis. The blood count showed anemia (Hb = 8g / dl), normocytic (MCV = 80,9fl), normocytic (MCHC = 33,2g / dl) and hyper eosinophilia 21.1%. The erythrocyte sedimentation rate was accelerated, CRP was positive. The parasitological examination of urine was normal. The skin histology showing under acanthotic epidermis, inflammatory granulomatous reaction around bilharzia viable eggs of Schistosoma (S) haematobium, grouped into small clusters of a few elements surrounded by lymphocytes plasma cells, eosinophil’s polynuclear,
histiocytes, épithéloïdes and multi-nucleated. Images of micro abscesses were visible around schistosoma eggs in entering the squamous epithelium to the keratin layer (Figure 2, 3). The diagnosis of vulvar skin bilharzia was set. The abdominal pelvic ultra sound showed a bilateral ureteral hydronephrosis, uroscanner without product injection noted an irregular parietal calcification of the bladder, ureters under pyelic, urethra, rectum, sigmoid colon and left with segmental dilatation of the ureters and expansions moderate bilateral pelvicalyceal (Figure 4).

The remaining tests were normal: IDR tuberculin, fasting glucose, HBsAg, TPHA / VDRL, transaminases, creatinine, the KAOP stools, chest X-ray.
Praziquantel (40 mg / kg in two spaced taken 15 days) was prescribed. The outcome was favorable with a clear regression of the swelling after a month. Resection of residual lesions was performed by urologists from the hospital. The outcome was favorable with skin scarring of recurrence-free lesions with 6 months of decline.

DISCUSSION
Vulvar localization of S. haematobium is rare. Some case series have described six cases in Burkina [6] and 8 in Mali [4]. In the Malian series, the mean age was 11 years but cases have been reported in adults [9, 10]. The consultation period ranged from 7 to 13 months in the Malian study while it was a year in one of Diallo and colleagues [5] and 3 years in our case, in line with late cutaneous schistosomiasis.
The pathophysiologic mechanism is not completely clarified. The symptoms of urinary S. haematobium is related to injuries caused by migration or embolization of eggs and granuloma formation indicating a defensive response of the host [11, 12, 13]. The diagnosis of the cutaneous form is provided by the histology of the granuloma, consisting of three concentric areas with the center of ovular debris then a crown of macrophages, eosinophils and giant cells and finally an outer zone fibrosis [11, 12, 13].
Urogenital symptoms are usual manifestations of S. haematobium. The digestive signs are characterized by a rectal involvement, often asymptomatic, with granulomas found in the endoscopic exam. Those granulomas are numerous whitish granules on a hyperaemic mucosa, there are confluent giving an aspect of pellets or candle stains. Inflammatory adematous pseudopolyps, sessile or stalked can
sometimes been observed. These polyps can be responsible of occlusive syndrom [14].

Cutaneous schistosomiasis may be associated with obstructive consequences of urogenital apparatus which result in a ureteral hydronephrosis in some cases, hence the importance of abdominal ultrasound. These complications were described by Garba in Niger and N'guessan in Ivory Coast (West Africa) [15, 16]. They found about 329 students, 2.74% of cases of dilatation urinary tract and bladder lesions of 40.43%; among bladder lesions, the wall thickening was noted in 14.89%. Computed tomography (CT) scan without injection is the preferred exam to confirm the calcification of the bladder, ureter and urinary tract [17]. Due to its cost in our context of limited resources, the application is not systematic. In our case, it helped to highlight the digestive and urinary calcifications.

S. haematobium is more found in black Africa: Madagascar (west), the Nile Valley (Egypt), Middle East (Saudi Arabia, Yemen). This parasitic disease directly related to agricultural development and increased irrigation (water), raging in homes on an endemic epidemic way [18]. In Senegal (West Africa), due development works in Senegal River by the construction of two dams (Diama Manantali), we are seeing an emergence of waterborne diseases in a Sahelian environment [19, 20].

Chemotherapy with praziquantel prescribed orally at a dose of 40 mg / kg causes a sterilization visceral foci [21, 22]. The treatment of nodular lesions requires a large surgical excision of residual disease [23]. Clinical tests in phase III vaccine against schistosomiasis or Bilhvax® (composed of a glucathion S-transferase 28 000 Daltons) in infected children, vaccine developed by the Pasteur Institute of Lille promoted with the french National Institute of health and medical research (Inserm) would hope in combating this public health disease [24].

CONCLUSION

We reported vulvar bilharzia, remarkable for its clinical presentation, concomitant urinary and gastrointestinal involvement and multidisciplinary management.

List of abbreviations

Computed tomography: CT
CONFLICT OF INTEREST

Authors declare no conflict of interest

AUTHOR’S CONTRIBUTIONS

Pauline Dioussé
Group 1 - Conception and design, Acquisition of data, Analysis and interpretation of data
Group 2 - Drafting the article, Critical revision of the article
Group 3 - Final approval of the version to be published

Haby Dione
Group 1 - Conception and design, Acquisition of data, Analysis and interpretation of data
Group 2 - Drafting the article,
Group 3 - Final approval of the version to be published

Mariama Bammo
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published

Chérif Dial
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published

Mariétou Thiam
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published
Aissatou Amy Diamé
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published

Bernard Marcel Diop
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published

Mamadou Mourtalla Ka
Group 1 - Analysis and interpretation of data
Group 2 - Critical revision of the article
Group 3 - Final approval of the version to be published

REFERENCES


FIGURE LEGENDS

Figure 1: Swelling of the labia, the groin in a girl 9 years old.

Figure 2: Dermal granuloma schistosomiasis. Magnification x 100. hematoxylin eosin

Figure 3: Dermal granuloma schistosomiasis with evidence of Shistosoma haematobium feature by the spur terminal Magnification X 300 (hematoxylin eosin).

Figure 4: Sagittal CT urography without injection of product showing an irregular parietal calcification of the bladder, ureter under pyelic, sigmoid with segmental dilatation of the ureter and pelvicalyceal expansion.
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