Unusual presentation of schistosomiasis as vulval polyp

S. M. Sahabi¹, A. F. Rabiu²

¹Dr. S. M. Sahabi, Department of Histopathology, Usmanu Danfodiyo University Teaching Hospital, Sokoto, ²Dr. A. F. Rabiu Department of Obstetrics and Gynaecology, Usmanu Danfodiyo University Teaching Hospital, Sokoto

Address for Correspondence: Dr. S. M. Sahabi, Email: smsahabii@gmail.com

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Abstract

Background: Schistosomiasis is a water borne disease caused by Schistosoma. It is the third most devastating tropical disease in the world, being a major source of morbidity and mortality for developing countries. Although the clinical manifestations on the urinary or gastrointestinal tracts are widely known, many clinical health-care professionals are unaware of the genital manifestations which are often ignored or underestimated. **Aim:** To report a rare clinical manifestation of schistosomiasis in the form of vulval polyp and also to increase its awareness its presentation and management. **Case Report:** An 11-year old pupil of presented to a peripheral hospital with 5-month history of progressive vulva swelling. She had no history of pain, itching, discharge or bleeding from the site. She had no urinary symptoms. The mass was clinically diagnosed as vulval polyp and surgically removed and sent for histology. She did well post operatively and was discharged to follow up in 2 weeks with the histology result. She was however lost to follow up. The histology results showed a granulomatous inflammation secondary to schistosomiasis. **Conclusion:** Vulval schistosomiasis though rare should be suspected in a child with vulvo-vaginal swelling. Biopsy should be obtained before definitive treatment is instituted.

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Keywords: Schsistosomiasis, Vulva, Polyp

Introduction

Schistosomiasis is a water borne parasitic disease caused by Schistosoma, the digenic trematode found in the blood vessels of man and livestock. Sometimes referred to as bilharzias, bilharziasis, or snail fever, it was discovered by Theodore Bilharz, a German surgeon working in Cairo, who first identified the aetiological agent as *Schistosoma haematobium* in 1851 [1].

It is the third most devastating tropical disease in the world, being a major source of morbidity and mortality for developing countries [2]. The six species of Schistosoma that cause disease worldwide, include *Schistosoma haematobium, S. mansoni, S. japonicum, S. intercalatum, S. mekongi and S. guineensis* [3].

More than 207 million people, 85% of who live in Africa, are infected with schistosomiasis [2], and an estimated 700 million people are at risk of infection in 76 countries where the disease is considered endemic, as their agricultural work, domestic chores, and recreational

Manuscript received: 18th June 2017 Reviewed: 24th June 2017 Author Corrected: 30th June 2017 Accepted for Publication: 05th July 2017 activities expose them to infested water [2,4]. Globally, 200,000 deaths are attributed to schistosomiasis annually [5].

In a study conducted in 2 districts in Sokoto among primary school pupil aged 7- 15, the result showed the overall prevalence of urinary schistosomiasis (*Schistosoma haematobium*) was 60.8% (228 positive cases in 375 samples); and for intestinal schistosomiasis (*Schistosoma manson*i) was 2.93% (11 positive in 375 samples) [3].

Female genital schistosomiasis (FGS) is a frequent complication in women with urinary or systemic schistosomiasis, particularly in geographic areas where the disease is endemic [6].

Schistosoma haematobium is the organism most frequently identified in these cases. Persistent, untreated infections may lead to increased susceptibility to sexually transmitted diseases, or even sterility [6]. We describe a case of FGS involving the vulva of an 11-year old girl.

Presentation of case- An 11-year old pupil of presented to a peripheral hospital with 5-month history of progressive vulva swelling. She had no history of pain, itching, discharge or bleeding from the site. She had no urinary symptoms. The mass was clinically diagnosed as vulval polyp; surgically removed and sent for histology.

She did well post operatively and was discharged to follow up in 2 weeks with the histology result. She was however lost to follow up. Microscopic examination revealed viable and calcified eggs of schistosomes in a typical advanced circumscribed granuloma with centrally located eggs, zone of epithelioid cells and outer zone of fibrous connective tissue infiltrated by lymphocytes. The histology result showed a granulomatous inflammation secondary to schistosomiasis.

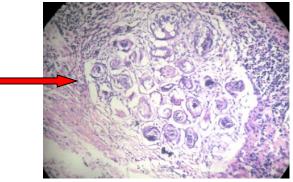


Figure-1: Photomicrograph showing a granuloma with aggregates of ova of S. haematobium (red arrow) (H&E Mag. X200)

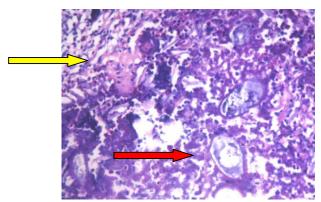


Figure-2: Photomicrograph showing ova of *S. haematobium* (red arrow) in a background with brightly eosinophilic crystalline material (Splendore-Hoeppli phenomenon) [yellow arrow] (H&E Mag. X400)

Discussion

Previous studies have also shown that FGS is a common manifestation of *S. haematobium* infection, with a prevalence ranging from 30 to 75% [3]. It has been estimated that approximately 9 to 13 million women may be afflicted with FGS in Africa alone [3]. Another important patient population to consider is composed of women who have visited endemic areas (especially if they have swum in freshwater lakes), or those who previously resided in those endemic areas [7]. FGS may be an important risk factor in the spread of sexually transmitted diseases including human immunodeficiency virus [8-10], and may lead to infertility secondary to ovarian fibrosis or tubal occlusion [11, 12]. FGS may indeed be a cofactor for the development of cervical cancer, but a clear link has yet to be established [13, 14].

The geographic distribution and etiology of schistosomiasis reflect the unique life cycle of

Schistosoma species. Schistosomes infect susceptible freshwater snails in endemic areas, usually with specific species of schistosomes infecting specific species of snails. The infected snails release cercariae 4-6 weeks after infection [15]. They can survive in fresh water up to 72 hours, during which time they must attach to human skin or to that of another susceptible host mammal or die [15].

Successful cercariae attach to human hosts, utilizing oral and ventral suckers. They then migrate through intact skin to dermal veins and, over the next several days, to the pulmonary vasculature. During this migration, the cercariae metamorphose, shedding tails and outer glycocalyces while developing double-lipid-bilayer teguments that are highly resistant to host immune responses [15]. The organisms, now called schistosomula, migrate through the pulmonary capillaries to the systemic circulation, which carries them to the portal veins, where they mature in male and female adult worms. Together they migrate along the endothelium, against portal blood flow, to the mesenteric (*S mansoni, S japonicum*) or vesicular (*S haematobium*) veins, where they begin to produce eggs [15].

The eggs, which are highly antigenic and can induce an intense granulomatous response, migrate through the bowel or bladder wall to be shed via feaces or urine. The cycle starts all over again. Eggs that are not shed successfully may remain in the tissues or be swept back to the portal circulation (from the mesenteric vessels) or to the pulmonary circulation (from the vesicular vessels via the inferior vena cava). Eggs can end up in the skin, brain, muscle, adrenal glands, and eyes [16].

Many case reports indicate that S. mansoni and S. japonicum may affect the genital tract [17]. However, only two community-based studies have explored this via biopsy of the uterine cervix [18]. Both studies were carried out in low-endemic areas, and further investigations are required. Blood vessel anastomoses between the pelvic organs are probably responsible for 'spill-over' of eggs into the genital tract, and the cervix has been suggested to be the predilection site for trapped eggs [19]. In clinical practice, the cervix, the Fallopian tubes, and the vagina are the most common gynecological sites found to contain Schistosoma eggs [20, 21].

Genital schistosomiasis is associated with stress incontinence and increased frequency of urination. There are case reports of severe acute disease such as ascites with ovarian schistosomiasis, ectopic pregnancy, and heavy egg infestation of the uterus in pregnancy [22]. Several case reports indicate decreased fertility and abortions in women with FGS, but this has only been explored in two community-based studies and needs further investigation [23]. A direct effect on hormones or ovarian function is controversial [24]. It is likely that the women with fatal complications, such as extra-uterine pregnancies, may have died or recovered receiving neither diagnosis nor adequate care [12]. The largest study to date, in Zimbabwe, did not find any association between abortion or menstrual irregularities with S. haematobium. In Madagascar, however, an association was found with abortion. There are reports of soft cervices by bimanual palpation as usually found in pregnancy, although the clinicians in the community-based studies have not discussed this [25].

Case reports on girls below the age of 15 are very few and are most commonly from the vulval regions [25]. A vaginal polyp was found in a 3-year old [26], in the upper third of the vagina was found in a 16-year old [27], vulval lesion in a 9-year old [28], and in an 11-year old girl [29] were the handful of cases available after literature search. Furthermore, there are reports of stunting and late pubertal development, suggesting hormonal disturbances, confirmed in animal models with decreased fertility and arrested development of corpora lutea [30].

Conclusion

The widespread lack of awareness of genital schistosomiasis leads to misdiagnosis and therefore, false and ineffective therapy. As female genital schistosomiasis is rarely diagnosed correctly, knowledge about the effect of treatment is also scanty. Incorrect diagnostic of genital schistosomiasis lesions frequently leads to debilitating and irreversible operations such as ovarectomy, salpingotomiy and hysterectomy. It is therefore of utmost importance to sensitize health workers and raise awareness of urogenital schistosomiasis, particularly in endemic countries.

The public health advantages of anthelminthic treatment with praziquantel go beyond the simple benefits of curing schistosomiasis and preventing its related genital morbidity. Regardless of the presumptive causal association with HIV infection, urogenital schistosomiasis is a disabling disease by itself, and it should be prevented with the currently available means. The WHO recommended policy of regularly treating school-age children with praziquantel [30] should be reinforced and extended, to involve collaborations with programmes for preventing HIV and other sexually transmitted infections.

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