Diagnosis and management of urogenital schistosomiasis in a young adult; a case report

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Schistosomiasis is rarely diagnosed in Western European countries. However, due to the popularity of exotic vacations, more and more western patients can get infected by schistosomiasis. Awareness of this disease is important, as an infection can lead to non-transitionnal cell bladder carcinoma in the long run (squamous cell carcinoma; SCC). In this article, we present a rare case of urogenital schistosomiasis in a 27-year old Belgian male. Extensive patient history together with eosinophil count and bladder biopsy, is the key to making the diagnosis. Medical treatment with praziquantel is often sufficient.

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Introduction
In most African countries, bladder cancer is the most frequent malignancy with a mean age of 46 years (as opposed to 72.9 years in patients with transitional cell carcinoma (TCC) of the bladder in the United States). Most of these tumours are squamous cell carcinomas (SCC), arising on a background of Schistosoma haematobia infection. Roughly 200 million people in 74 countries suffer from schistosomiasis, of whom 100 million people are affected by bilharziasis of the urogenital tract, caused by S. haematobium. A very rare disease in Europe, it is endemic and a common health problem in Malawi and other African countries. Due to increasing migration and tourism, European urologists are currently more frequently confronted with infected people travelling from these endemic zones. Therefore, awareness of the disease and thorough patient history are of vital importance.

Patient History
A 25-year old Belgian male consulted the outpatient clinic with complaints of dysuria, bilateral testicular pain and urgency. Ultrasound examination showed acute prostatitis and epididimitis while urine cultures were negative. He was treated with quinolones for six weeks but his symptoms did not completely disappear, resulting in the diagnosis of Chronic Pelvic Pain syndrome. At the age of 27, he presented with persisting urethralgia, frequency, microhaematuria, leukocyturia and slightly elevated PSA of 1.54 ng/mL. Because of his severe symptoms in combination with persisting microhaematuria, a thorough anamnesis with special attention to his travel history was performed, revealing that he went on a diving-trip in Lake Malawi in Malawi two years before. A few weeks after his return in Belgium his complaints had started. In Malawi, Schistosomiasis haematobium is
highly endemic.² Therefore, his travel history, together with his symptoms, were very suspicious for schistosomiasis. Blood analysis showed eosinophilia, but urine egg count and serology were negative. Cystoscopy showed atypical granulomata in the trigone, sparing the ureteral orifices. Transrectal ultrasound was repeated and showed thickening and hyperechogenicity of the irregular bladder wall, coexisting with a submucosal mass protruding into the lumen at the bladder base (Figure 1). A transurethral endoscopy with biopsies of the lesions - with the impression of egg-excretion - was performed. Postoperatively, one dose of praziquantel 3g (40mg/kg) was administered orally. The next day, the patient left the hospital pain-free.

Histopathological examination of the bladder biopsies confirmed the presence of granulomatous lesions with active form of schistosomiasis (Figure 2) and excluded (squamous) bladder carcinoma. Two months later, the patient’s symptoms were completely resolved and cystoscopy was negative. Eosinophil count was again within normal range. One month later, a second dose of praziquantel was administered. Yearly follow-up with cystoscopy and cytology (risk of bladder SCC), medical imaging of the higher urinary tract (risk of obstruction) and eosinophil count were planned.⁴⁻⁷,¹¹

Discussion and conclusions

Urogenital schistosomiasis is caused by the digenetic trematodes S. haematobium and is frequently the result of unprotected contact (eg. bathing, swimming, fishing,…) with contaminated fresh water in endemic areas.¹,²,⁴ Indeed, S. haematobium eggs are excreted in the water, hatch and release miracidia that penetrate their intermediate host, the aquatic bulinid snails. They excrete cercaria, which penetrate (non-injured) human skin during contact with contaminated water. Mature worms mate and migrate to the pelvic venous plexuses to begin oviposition, after which the eggs migrate into the surrounding organs causing micro-mucosal perforation. Non-excreted eggs remain in the submucosa of the pelvic organs, encapsulated in fibrous granulomas.

The ulcerative stage of the infection frequently presents with urethralgia, frequency, suprapubic pain and haematuria but the presentation may differ greatly depending on the stage of the disease, severity of the infection and localisation of the eggs of the parasite. It may cause local complications, such as upper urinary tract obstruction with consequent hydronephrosis and kidney damage, nephrotic syndrome and ectopic localisations due to aberrant worm migration.¹,⁴⁻⁹,¹⁰ Moreover, the association between schistosomiasis and the late (10-20 years post-infection) develop-
ment of non-transitional cell bladder carcinomas (SCC) is well known. Bladder SCC was the most common malignant disease in Egypt until a reduction of schistosomiasis in Egypt resulted in an important shift in the epidemiology of bladder cancer in Egypt, which is nowadays characterised by a lower incidence, a higher age at diagnosis, a lower male predominance and lower proportion of SC histology, but with increased incidence of transitional cell carcinoma. This shows the remarkable cause-effect relation between the eradication of schistosomiasis and the subsequent radical decline in SCC of the bladder. Over 90% of bilharzial SCC patients present with T3 and T4 tumours, due to the overlap of symptoms of bilharzial cystitis with early malignant disease. In regions where Schistosomiasis is rare, it is even more likely to miss these early stages if one is not aware of this disease.

Morphologically, SCCs are nodular, ulcerative, or infiltrative. Papillary lesions are rare. Lymph node invasion is rare (19%), even though the tumours are mostly diagnosed at advanced stages. Distant metastases are much lower in incidence than those reported for the transitional cell carcinoma. Management of SCC of the urinary bladder should be the same as that of advanced muscle-invasive TCC. Radical cystectomy remains the main treatment giving a 5-year survival rate of 50%.

The importance of a thorough anamnesis and travel history can therefore not be stressed enough. In the early phase of the disease, ultrasound may not show the typical lesions as seen in our case but when the classic therapy for prostatitis fails, schistosomiasis still has to be incorporated in the differential diagnosis if there is an anamnesis for travelling in endemic areas.

Diagnosis is based on the detection of S. haematobium eggs in the urine, which should be collected on several consecutive days between 11.00 and 14.00, because of the peak output of the eggs at this time. Urinalysis looking for microhaematuria and proteinuria and urine cytology in the screening for malignancy should be performed as well. When the egg count is positive, the viability of the eggs can be checked through the egg hatch procedure and diagnosis is confirmed for which medical treatment should be started. However, the sensitivity of urine filtration for parasitologic diagnosis is known to be lower in individuals with minimal infections. Such light infections are more common among adults, making the detection of their active infections more difficult. Thus, adults who score as “egg-negative” in urine filtrations may be minimally infected with S. haematobium, with ongoing tissue injury and inflammation.

Cystoscopy is indicated in cases where non-invasive semiological evaluation does not lead to a diagnosis. Thus, patients with negative egg-counts require a cystoscopy, which can show very characteristic lesions and generally is followed by transurethral biopsy of the bladders lesions and histopathological examination of the resected tissue. Cystoscopy may also be used to evaluate bladder-neck stenosis, bladder calcification, and ureteral orifices stenosis. Again, when these results are positive, medical treatment should be started.

Other diagnostic tests for urinary tract sequelae include ultrasonography for ureteral obstruction or to detect hydronephrosis, urography to evaluate bladder morphology and ureteral stenosis and CT in suspected cases of kidney tumour.

As medical treatment the two drugs proven to be active against S. haematobium are praziquantel and metrifonate. They are both effective treatments and
have few adverse effects.1,11 Our patient was given praziquantel. Surgical treatments are only necessary when complications or bladder cancer occur and are not further discussed here. Epidemiological studies carried out in Ghana showed that bladder damage seen on ultrasound is common in young adults as well as in older people, but it is not clear when cancer starts to develop. Also, it is not clear whether medical treatment of schistosomiasis can interrupt or prevent cancer development.8,10 The medical treatment is efficient, but therapeutic failure (with complications) is possible. Therefore, yearly follow-up with cystoscopy, cytology, medical imaging of the higher urinary tracts and eosinophil count is required, next to lifelong awareness because of the possible late development of SCC.

### References

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