CASE REPORT

PROSTATIC SCHISTOSOMA JAPONICUM WITH ATYPICAL IMMUNOPHENOTYPING OF INDIVIDUAL GLANDULAR TUBES: A CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract. There are few cases of prostatic schistosomiasis. Here we report a case of Schistosoma japonicum of the prostate, in which the immunophenotyping of individual glandular tubes was atypical. Whether the S. japonicum infection contributed to the lesion or not is unknown. We suspect the lesion was a sign of early precancerous hyperplasia. Follow-up of this patient may give clues about the relationship between schistosomiasis and prostate cancer. This is the first case report of prostatic S. japonicum in the English literatures. A review of the literature is carried out.

Keywords: Schistosoma japonicum, prostate, immunophenotyping, glandular tube

INTRODUCTION

Schistosomiasis is found in 75 countries worldwide including Asia, Africa and Latin America and is one of the most important tropical infectious diseases in the world (Yosry, 2006). Approximately 200 million people worldwide are believed to be infected, leading to the loss of 1.53 million disability-adjusted life years (Gryseels et al, 2006). The major species of Schistosoma infecting humans are Schistosoma mansoni, S. haematobium and S. japonicum. S. haematobium lives in the veins of the bladder and perivesical venous plexus (Fu et al, 2012). The adult worm spawns its eggs in the bladder wall, leading to chronic inflammation of the bladder, which is associated with hyperplasia of the bladder mucosa and squamous metaplasia (Von Lichtenberg et al, 1971). On rare occasions, it can be found in the uterus, vaginal wall, prostate

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and other genitourinary organs (Cheever, 1978). To our knowledge, there are no case reports in the English literature of *S. japonicum* infecting the prostate. This is the first case report of prostatic *S. japonicum*.

**CASE REPORT**

A 66-year-old man came to the outpatient department with a complaint of frequent micturition. He denied a past medical history of diabetes, heart disease, hepatitis or tuberculosis. He did have a history of hypertension for several years. The patient had grown up in Honghu, a city along the bank of the Yangtze River, a place where *S. japonicum* is prevalent. He had no complaints of dysuria, hematuria, fever, diarrhea, vomiting or other symptoms. General physical examination was unremarkable. Rectal examination revealed an enlarged, firm prostate with no nodes. Laboratory investigations showed a total prostatic specific antigen (TPSA) of 5.7 ng/ml and a free prostatic specific antigen (FPSA) of 0.949 ng/ml (reference values: TPSA <4 ng/ml, FPSA <0.934 ng/ml). Ultrasound examination of the urinary system revealed an enlarged prostate. Ultrasound guided transrectal biopsy of the prostate was performed. Twelve specimens were obtained from the four zones of the prostate and sent for histopathological and immunohistochemical examinations. The results revealed hyperplasia of both the stromal cells and gland of the prostate. Additionally, scattered eggs of *S. japonicum* were found in the middle section of the left lateral side of the prostate (Fig 1A). Immunohistochemical examination (examining antibodies to p504S, 34BE12 and P63) revealed benign hyperplasia of the prostate. However, immunophenotyping of the individual glandular tubes was atypical (Fig 1B). A diagnosis of prostate cancer could not be ruled out completely. A serum examination revealed IgG against *S. japonicum* was positive (+++). The patient was treated with praziquantel for prostatic schistosomiasis. At present, the patient is undergoing close follow-up.

**DISCUSSION**

Schistosomiasis of the prostate is uncommon. We searched PubMed for reports and found case reports of concomitant prostatic schistosomiasis and
Table 1
Published literature of patients with prostatic schistosomiasis.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Country of patient</th>
<th>Patient age (years)</th>
<th>Signs and symptoms</th>
<th>Medical history</th>
<th>Laboratory results</th>
<th>Pathological diagnosis</th>
<th>Schistosoma species</th>
</tr>
</thead>
<tbody>
<tr>
<td>Al Adnani</td>
<td>1985</td>
<td>Iraq</td>
<td>Not mentioned</td>
<td>Not mentioned</td>
<td>Not mentioned</td>
<td>Not mentioned</td>
<td>Squamous carcinoma of prostate</td>
<td>Not mentioned</td>
</tr>
<tr>
<td>Alexis and Domingo</td>
<td>1986</td>
<td>Puerto Rico</td>
<td>49</td>
<td>Urinary tract obstruction</td>
<td>Unremarkable</td>
<td>Alkaline phosphatase (↑), Acid phosphatase (↑)</td>
<td>PAC</td>
<td>S. mansoni</td>
</tr>
<tr>
<td>Cohen et al</td>
<td>1995</td>
<td>South Africa</td>
<td>27, 29, 29</td>
<td>Weight loss, malaise, back pain, urinary symptoms</td>
<td>Not mentioned</td>
<td>PSA (↑)</td>
<td>PAC</td>
<td>S. haematobium</td>
</tr>
<tr>
<td>Basilio-de-Oliveira et al</td>
<td>2002</td>
<td>Brazil</td>
<td>68</td>
<td>Routine examination</td>
<td>Unremarkable</td>
<td>PSA (↑)</td>
<td>PAC</td>
<td>S. mansoni</td>
</tr>
<tr>
<td>Lambertucci et al</td>
<td>2006</td>
<td>Brazil</td>
<td>56</td>
<td>Routine examination</td>
<td>Unremarkable</td>
<td>PSA (↑)</td>
<td>PAC has not been discarded.</td>
<td>S. mansoni</td>
</tr>
<tr>
<td>Bacelar et al</td>
<td>2007</td>
<td>Brazil</td>
<td>47</td>
<td>Diagnosis of PAC (Gleason score 3+3, cT2a)</td>
<td>PAC</td>
<td>PSA (↑)</td>
<td>PAC</td>
<td>S. mansoni</td>
</tr>
<tr>
<td>Mazigo et al</td>
<td>2010</td>
<td>Tanzania</td>
<td>50</td>
<td>Chronic hematuria and suprapubic pain</td>
<td>Unremarkable</td>
<td>Hemoglobin (↓)</td>
<td>PAC</td>
<td>S. haematobium</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>75</td>
<td>Frequent micturition</td>
<td>Unremarkable</td>
<td>Urea (↑), Hemoglobin (↓)</td>
<td>PAC</td>
<td>S. haematobium</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>41</td>
<td>Suprapubic mass, frequent micturition</td>
<td>Unremarkable</td>
<td>Hemoglobin (↓), Urine creatinine (↓)</td>
<td>PAC</td>
<td>S. haematobium</td>
</tr>
</tbody>
</table>

PAC, prostatic adenocarcinoma; PSA, prostatic specific antigen; ↑, elevated; ↓, decreased.
adenocarcinoma. Schistosomiasis has long been associated with malignant disease. In 1911, Ferguson published a case series of 40 Egyptian patients with simultaneous bladder cancer and S. haematobium infection and proposed a relationship between schistosomiasis and bladder cancer. Some studies have reported a link between S. japonicum infection and carcinoma of the colon and rectum (Ch'en et al, 1965; Ming-Chai et al, 1980). However, case reports of schistosoma infection and prostate cancer are rare (Table 1).

Al Adnani (1985) reported two Iraqi patients with schistosomiasis and prostatic carcinoma, squamous cell carcinoma of the prostate and calcified eggs in the seminal vesicles were found. Using immunohistochemistry, the author confirmed the tumor was a primary tumor of the prostate in both cases, rather than infiltration of bladder cancer (Bacelar et al, 2007).

Prostatic cancer and schistosomiasis of the prostate have also been reported in younger patients: a 49-year-old patient was diagnosed with simultaneous prostate cancer and S. mansoni (Alexis and Domingo, 1986), and another case was reported in 1992 (Godec et al, 1992). At the time of diagnosis, the patient already had spread of the cancer to his lumbar spine. Pathology showed a diffusely infiltrating adenocarcinoma with S. mansoni eggs surrounded by inflammatory cells. The authors suggested severe infection of the prostatic gland with S. mansoni eggs may be associated with the development of cancer in young patients. Cohen et al (1995) reported a case series of patients with simultaneous advanced prostate cancer and infection with S. haematobium in South Africa. The patients were diagnosed with advanced cancer at an unusually young age without a family history of malignancy. One patient was 27 years old and the other two were 29 years old. The young patients with simultaneous advanced prostate cancer and S. haematobium infection raise the possibility of a relationship between neoplasia of the prostate and schistosomiasis. The authors proposed infection with schistosoma could have been a causal factor for the development of prostate cancer at a young age.

The species of schistosoma in the case reports were S. mansoni and S. haematobium. As far as we know, there have been no case reports of S. japonicum of the prostate in the English literature; this is the first one. As to the possible relationship between schistosomiasis and carcinoma of the prostate, many authors have proposed the hypothesis that glandular atrophy associated with focal fibrosis of the prostate may lead to precancerous hyperplasia (Moore, 1936; Franks, 1973). Other authors have suggested the presence of carcinogens in the parasite and the enzyme beta-glucuronidase tends to act as cofactors for inducing cancer (Khalafallah and Abul-Fadl, 1964; Mazigo et al, 2010). In our case, fibrosis of the prostate was not clearly present, but the immunophenotyping of individual glandular tubes was atypical. Studies of animal models with prostatic schistosomiasis may provide clues to the mechanisms involved.

Even though there are several case reports of schistosoma infection of the prostate in prostate cancer patients, the causal relationship between adenocarcinoma of prostate and schistosoma infection is unclear. In our case, the immunohistochemical examination revealed immunophenotyping of individual glandular tubes was atypical. Whether schistosoma infection contributed to the atypical immunophenotyping of the glandular tubes is unknown. We suspect precancerous hyperplasia is the cause of the elevated
PSA level, even though the diagnosis of precancerous lesions lacks sufficient evidence because of the limited pathologic findings. The patient is undergoing close follow-up. Follow-up may give a clue to the relationship between schistosomiasis and prostate cancer. If the patient develops precancerous lesions or adenocarcinoma, our suspicions will be supported by the findings. The possibility of a causal relationship between schistosomiasis and prostatic cancer will be further supported.

In conclusion, the relationship between schistosomiasis and prostate cancer remains unclear. We report a case of *S. japonicum* of the prostate in which the immunohistochemistry findings showed atypical individual glandular tubes. We suspect the lesion is an early stage of precancerous hyperplasia. Follow-up will help us better understand the relationship between schistosomiasis and prostate cancer.

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REFERENCES


