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Autor: Badejo, O.A. / Soyinka, Femi / Laja, A.O.
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Faculty of Health Sciences, University of Ife, Ile-Ife, Nigeria

Ectopic lesion of schistosomiasis of the penis simulating an early carcinoma

O. A. BADEJO, FEMI SOYINKA, A. O. LAJA

Summary

A case of ectopic lesion of *Schistosoma haematobium* of the penis with extensive tissue destruction, simulating an early carcinoma of the penis and almost resulting in an autoamputation of the crown of the penis, is presented. The penis was surgically repaired and the patient treated with ambilhar. In schistosoma endemic area, it is important to think of ectopic schistosoma lesion by such a presentation. Existing theories to explain the presence of schistosoma eggs in locations outside the portal-caval system were reviewed and another one was advanced: its being sexually transmitted.

Key words: *Schistosoma haematobium*; genital lesion; differential diagnosis; case study.

Introduction

Adults of *Schistosoma haematobium* are normally found in the vesical plexus and/or in the pudendal and hemorrhoidal plexii. Occasionally, they may lodge in the portal vessels and pulmonary arterioles, and their eggs may be found in far distant places in the portal-caval venous circulation.

Tissue reactions of the body to the worms or their eggs occurring outside the portal-caval venous circulation has been called ectopic lesions of schistosomiasis.

Ectopic lesions of *S. haematobium* was said to have been reported as far back as 1936 (Faust, 1948) where the eggs of this parasite were identified in granulomas of spinal cord. Since then, there has been series of reports on cases of ectopic lesions of schistosomiasis in man. Faust (1948) summarized 82 cases of ectopic schistosomiasis. These cases included lesions in the conjunctiva and superior ophthalmic vein. The largest number of proven or presumptive sites

Correspondence: Dr. O. A. Badejo, Faculty of Health Sciences, University of Ife, Ile-Ife, Nigeria

of ectopic lesions are said to be the brain, next in frequency to this are the conjunctival lesions, and third in the series is the involvement of the spinal cord. Furthermore, isolated cases of skin and peripheral blood vessels have been reported.

The reported ectopic schistosomiasis lesions of the skin were on the scrotum, the lower and upper portions of the chest, and in the lumbar regions. These lesions were said to have presented as clusters of small abscesses or large indolent ulcers containing masses of eggs.

Makar (1955) reported 6 cases of bilharziasis of the penis, out of the 88 patients that were admitted for bilharzial urethral fistulae. The presenting symptom of the chronic stage of this disease was said to be urinary obstruction. According to Makar, bilharziasis of the penis and penile urethra is an extension of a bilharzial affection of the bladder, and as such signifies a severe form of the disease.

In view of the various existing reports, ectopic lesions in schistosomiasis can no longer be considered as "rare" or "very rare". However, the peculiarities of this case presented by us, together with the unforeseen consequences that might have befallen the patient, make us report it as another but yet uncommon presentation of ectopic lesion of schistosomiasis.

Case report

The 35-year-old driver presented in the surgical outpatient of the Ife University Teaching Hospitals Complex, Ile-Ife, on May 10th, 1977, with abrasion of crown of penis immediately after intercourse with a prostitute and appearance of bullae and papular rashes around the crown of the penis a week later. The bullae ruptured giving way to small ulcerations which merged together to form a large ulcer round the crown of penis. The ulcer progressed in size and depth eating up the whole circumference of the base of the crown within six months. Later hypospadias developed and micturation became a problem, but no haematuria was noticed. His marriage had to be suspended and native medicine was applied without success. Earlier the patient had been exposed to having baths in an open stream in his farm. This he continued to do after the onset of his illness, but ultimately stopped because the ulcer was getting bigger and he could not allow anyone to see.

Examination and laboratory findings

General examination of the patient revealed no abnormality. Full blood count, serum urea and electrolytes were normal. Routine urinalysis at first showed pus cells, but no ova of schistosoma. There was no blood in urine. Culture of urine for micro-organism also yielded no growth. Swab from the ulcer

yielded no growth. VDRL test and Kahn's test were negative. Stool examinations for parasites and ova were negative.

Because of the extensive tissue destructions on the penis, the diagnosis that was initially thought of, was that of carcinoma of the penis, though this occurrence is known to be uncommon in this environment. A biopsy was first taken from the edges of the lesion for quick histological confirmation. The histopathological report came back as "ulceration and chronic inflammation. No evidence of malignancy. Ova of *Schistosoma haematobium* noted in the dermis deep to the area of ulceration".

Intracutaneous test done on the volar surface of the patient's arm with 0.5 ml of schistosomiasis skin test antigen (Wellcome) gave a strongly positive reaction (2.5 cm), whereas the antigen control test on the same patient was negative. Repeated collection of terminal urine of the patient over 24 h gave positive *S. haematobium* ova on two occasions. The diagnosis of an ectopic lesion of *S. haematobium* with autoamputation of the penis was therefore made.

The penis, however, showed advanced ulceration and the so-called autoamputation of the glans penis. On the dorsal side, there was no connection between the glans and corpus. The urethral opening was located 12 mm from the proximal end of the torn off end of the glans. The ulcer which was 0.5 mm in depth in some places circumscribed the whole of the glans, leaving it hanging on a fibrous tissue on the ventro-medial aspect. The remaining part of the penis anatomy up to the bulb were normal.

Management of patient

The patient was started on full dose of niridazole, and the penis was repaired surgically. The operation was performed under general anaesthesia. The urethra was opened up to the middle of the corpus through a longitudinal incision. The secondary hypospadias edges were resected and a Foley's catheter inserted into the bladder. The massive fibrosis and granulating tissue in the glans penis still presented doubts as to the possibility of carcinoma of the penis, but this is rare in this part of the world. A circular excision round the glans penis was made. The urethra was closed up with 3-0 catgut and the skin with 3-0 astralen. The glans penis was reimplanted on the corpus using the inserted catheter as a bridge all the way.

The postoperative course was normal. No haematuria was noticed. The stool examination gave nothing significant. The operation wound healed primarily with no signs of excess keloid or painful erection. The postoperative urethrocystogram gave no signs of stricture or wall calcification of the bladder. There were no signs of schistosomiasis of the bladder.

The biting pain during micturation disappeared under ambilhar therapy, and the patient was discharged approximately four weeks after operation.

Discussion

The occurrence of ectopic lesions of *S. haematobium* has been adequately reported, however, a case of ectopic lesion of this disease on the penis, almost ending in an autoamputation of the penis is to our knowledge not yet reported.

In the six cases of bilharziasis of the penis reported by Makar (1955) in Cairo, involvement of the cavernous tissue with fibrosis and urinary obstruction were reported as the main symptoms. He mentioned induration of the penis as a possible complication of the disease. However, no case of autoamputation was reported. Makar stressed in his report that bilharziasis of the penis and penile urethra is an extension of bilharzial affection of the bladder.

The presenting symptom in our case was an indurated ulcer of the penis which already had 'eaten' its way through the urethra on the dorsal part of the penis and almost causing an autoamputation of the crown. The lesion according to the patient started on the skin of the penis. There was no urinary obstruction, and the penis was still capable of erecting or contracting. It is significant to note that there were neither signs of urethral stricture nor that of radiographical abnormalities of the bladder wall due to schistosoma. Thus, a primary ectopic lesion of the penis could not be ruled out.

Various theories have been advanced to explain the presence of schistosoma eggs in the several locations outside the portal-caval systems.

One theory was that cercariae enter the skin or mucous membranes and develop directly into adults in nearby veins without going through the incubative stage in the liver (Diamantis, 1928; Shimidzu, 1935; Badir, 1946). If this hypothesis were correct, it could apply to our patient, as the patient had swam in rivers where bilharzia is believed to inhabit.

A second theory is that cercariae reaching blood vessels other than the mesenteric artery may develop normally outside the portal-caval system (Faust and Meleney, 1924; Faust et al., 1934; Koppisch, 1937).

A third hypothesis is that eggs deposited by the parent worm in the usual sites are filtered through the capillary barriers in the liver and lungs and get into the general circulation to be filtered out from terminal blood vessels (Shimamura and Tsunoda, 1905).

A fourth theory is that adult worms migrate out of their normal habitats and the female worms oviposit in venules (Black, 1945). The discovery of a paired male and female *S. haematobium* in a branch of the superior ophthalmic vein provides the evidence in support of this theory.

A fifth theory for the explanation for ectopic lesions in schistosomiasis predicates the use of vertebral venous system which, by virtue of its anatomical relation to the veins of the portal caval system, would easily make the transportation of schistosoma eggs to distant places like the brain, spinal cord, conjunctiva or skin very easy. Likewise, adult worms may be swept out of abdominal venous circulation and come to rest in other systems (Baston, 1940; Gama and Marques de Sa, 1945).

All the five theories mentioned above could be applied to our patient. However, we would like to make an additional theory. This is the possibility of a sexually transmitted schistosomiasis lesion (ectopic) on the penis.

Bilharziasis of the female urethra and cervix has been reported. It is also known that the ova are excreted through the urethra. It is not impossible that in our patient the point of entry of the ova of *S. haematobium* could have been through the membrane from the cervix or indirectly from the urethra of the female. He noticed an abrasion after the sexual intercourse, bullae formed around this area about one week later and he noticed an induration around that area, that finally broke into an ulcer months after. The missing link is the proof that the female partner also had schistosomiasis and was discharging eggs at the period of intercourse. However, the circumstances point towards this possibility. Otherwise, the first theory mentioned could also have applied, as the patient swam in a schistosome infested river, and he also had *S. haematobium* ova in the dermis of the penis.

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