Clinical and Pathological Aspects of the Sequelae of Urogenital Schistosomiasis: Findings Regarding 43 Cases

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Abstract: Purpose: To describe the clinical-pathological profile of sequelae of urogenital schistosomiasis. Patients and methods: We performed a retrospective study of the sequelae for cases of urogenital schistosomiasis treated between January 2011 and December 2016. These cases were from Senegal and neighboring countries. Results: We included 43 cases. The mean age of the patients was 43.2 ± 16.6 years (14 - 75 years). The sex ratio was 3.3. The sites of the sequelae lesions were the bladder and pelvic ureters in 83.7%, the bladder only in 13.9%, and the ureters only in 2.3% of the patients. Bladder wall calcification was the most common lesion (74.4%), followed by bladder masses (48.8%). Stenosis of the orifice was the most common ureteral lesion (30.2%). The bladder masses were a squamous cell carcinoma in ten cases, a urothelial carcinoma in one case, and a schistosomiasis granuloma in five cases. In one patient, the histological type was not specified. Of the ten cases of squamous cell carcinoma, eight had died of cancer. Of the twelve cases of ureteral orifice stenosis treated by ureterocystoneostomy, the outcome was good in nine cases and poor in three patients. Conclusion: In this study, sequelae of urogenital schistosomiasis were most often observed in young adult males. The most common sequelae were vesical and ureteral calcifications, bladder cancers, and pelvic ureteral strictures.

Keywords: Urogenital Schistosomiasis, Calcifications, Bladder Cancer, Urinary Obstruction

1. Introduction

Described for the first time by Théodore Bilharz in 1851, Schistosoma hematobium (Sh) is a trematode hematophage responsible for urogenital schistosomiasis (UGS) [1]. It is a parasite of the urinary and genital perivesical venous plexuses. The laying of eggs by female worms in the submucosa of the targeted organs triggers an inflammatory reaction (granuloma) that develops into fibrosis and/or calcification. These irreversible tissue lesions are observed particularly in case of severe and repeated infections. They can result in stenoses of the excretory tract, progressive alteration of renal function, and bladder cancer [2, 3].

In Senegal, UGB is an endemic and pronounced infection, particularly in children [4]. Despite its endemic nature, there have been few studies on the sequelae lesions and their consequences [5].

The objective of this study was to report the profile of the sequelae from UGB in our area and to evaluate the outcomes of their treatment.

2. Patients and Methods

We carried out a retrospective study of the sequelae for cases of urogenital schistosomiasis treated between January 2011 and December 2016. These cases came from Senegal and adjoining countries. The diagnosis of sequelae of urogenital schistosomiasis was often made based on a set of
very indicative indirect evidence in patients who had a
history of spending time in schistosomiasis endemic areas,
swimming in freshwater holes, and terminal hematuria. This
evidence was based either on medical imaging, cystoscopy,
or also by a histological examination (revealing eggs of SH
in biopsies or surgical specimens) [2, 3, 6]. The studied
parameters were the following:
1. Epidemiological: age, gender, geographical origin;
2. Clinical: circumstances surrounding the discovery, the
type and the location of the sequelar lesions;
3. Therapeutic: the type of treatment carried out, the
outcome of the treatment, and changes after the
treatment. After ureterocystoneostomy, the outcome
(after removal of the ureteral or JJ stent) was considered
to be good in the absence of a recurrence of the ureteral
obstruction with regression or disappearance of the
upstream ureterohydrouphrosis.

The data were analyzed with Microsoft Excel 2013
software.

3. Results
We included 43 patients. The average age of the patients
was 43.2 ± 16.6 years (14 - 75 years). The sex ratio (M/F)
was 3.3.

The sequelar lesions were located both in the bladder and
the pelvic ureter in 83.7% of the cases, only the bladder in
13.9% of the cases, and only the ureter in 2.3% of the cases
(Table 1). In terms of the bladder, wall calcification (Figure 1)
was the most common lesion (74.4%), followed by a mass in
the bladder (48.8%) (Figure 2). Stenosis of one or two
ureteral orifices was the most common ureteral lesion (Table
2) followed by ureteral stricture (Figure 3). A single genital
lesion was noted in our patients. It involved calcification of
the seminal vesicles in a patient who already had stenosis of
the pelvic ureter.

The most commonly carried out treatments were
transurethral resection of the bladder in 17 cases (43.59%)
and ureterocystoneostomy in 12 cases (27.9%) (Table 3).
After transurethral resection of the bladder, histology of the
resection shavings revealed a squamous cell carcinoma in 10
cases, a urothelial carcinoma in one case, and a schistosomal
granuloma in five cases. For one patient, the histological
result had not been reported. No radical cystectomies were
carried out due to the advanced stage of the cancers at the
time of the diagnosis. Of the ten cases of squamous cell
carcinoma, eight had died as a result of their cancer. After
ureterocystoneostomy, there was a favorable outcome in nine
cases and a poor outcome in three patients with a recurrence
of the ureteral obstruction at the level of the intramural ureter
and persistence of the ureterohydrouphrosis. In the patient
who was initially treated by Boari-Küss flap, the stricture of
the pelvic ureter recurred and the intervention was resumed
with placement of a JJ stent that was changed regularly.
4. Discussion

Although urogenital schistosomiasis (UGS) can affect the entire urinary tract, it primarily affects specific sites such as the bladder, the pelvic ureter, and the seminal vesicles in men [7]. In our study, the sequelae lesions were simultaneously vesical and ureteral in 83.7% of the cases. The initial lesion is a reaction of the host against the eggs deposited in the tissues. In the beginning, it manifests as an inflammatory granuloma. Its healing or its progression toward fibrotic and calcification depends on the number of eggs retained in the tissues [2]. An intermittent infection that is treated early on and in an appropriate manner typically heals without sequelae. A severe infection, frequent reinfections, delayed and/or inadequate treatment, or a lack of treatment leads to the appearance of retractile tissue or calcification or fibrosis due to the eggs retained in the tissues [2].

We encountered twelve cases of bladder cancer, of which ten were squamous cell carcinomas. It has long been known that there is a causal link between UGS and cancer [8, 9]. The mechanisms leading to the induction of cancer have not yet been properly elucidated. However, the main mechanism is thought to involve chronic inflammation of the bladder wall as a result of the retention of Sh eggs in the tissue [10, 11]. The clinical pathology characteristics of bladder cancers induced by UGS are a young age of the patients, male predominance, a squamous cell carcinoma nature of the carcinomas, frequent intratumoral calcifications, and aggressiveness of the cancers [10, 11]. These characteristics were noted among our patients. The similarity of the clinical signs of bladder cancer (terminal hematuria and irritative lower urinary system symptoms) with those of progressive urogenital schistosomiasis results in these cancers often being diagnosed at locally advanced or metastatic stages in our context of endemic schistosomiasis. This was the case for all of our patients and the reason why none of them were treated by radical cystectomy. Once such a cystectomy is no longer feasible, the prognosis for the patients is poor as the cancers are largely refractory to both radiotherapy and chemotherapy [12]. Of the ten cases of squamous cell carcinoma, eight had died as a result of their cancer. In their series, Abdou et al. reported 15 cases of cancer that were mostly at an advanced stage [13]. Seven of these patients underwent a cystectomy and the others received palliative treatment. At five years, seven patients had died and five were lost to follow-up. The high rate of mortality with these cancers, therefore, appears to be due mainly to the delay in diagnosis.

In terms of the bladder, wall calcification was the most frequent lesion (74.4%). Fibrosis and wall calcification of the bladder alter its dynamics. They can result in hypertension or an atonic bladder responsible for poor emptying [7]. The ensuing urinary stasis promotes the occurrence of bacterial urinary infections and the induction of bladder cancer such as squamous cell carcinomas in particular [10]. Fibrosis and bladder calcification can also lead to a high-pressure contracted bladder, which constitutes a danger for the upper urinary system. Bladder retraction can justify performing an
enterocystoplasty, which was not done for our patients [13]. The bladder neck is one of the main sites for the deposition of Sh eggs [7]. The ensuing intense inflammation results in bladder outlet obstruction [3]. Such an obstruction promotes bladder urinary stasis and bacterial urinary infections. We have treated three cases of bladder-neck stenosis by an incision that allows the neck to be enlarged [3]. The risk of recurrence is high, however.

The fibrous and stenosing sequelae are frequently located at the level of the pelvic ureters. They can alter their peristaltic movement and thereby contribute to the creation of a functional obstruction [6]. They can also lead to a stenotic ureteral obstruction. This can result in unil or bilateral ureterohydronephrosis that can progress to alteration of renal function. Strictures of the pelvic ureter are hence potentially serious. They can readily be diagnosed by medical imaging, particularly by tomodensitometric urography [6, 14]. Praziquantel is effective at the early stage of UGS (arrest of egg-laying, destruction of the parasite, and regression of inflammatory granulomas) but has no effect on the nonreversible stages of fibrosis and calcification [2].

Treatment of obstructive lesions of the pelvic ureter, therefore, calls for open or endoscopic surgery. This can be done by several methods such as high-pressure balloon dilation [15], ureterocystoneostomy or ureteroileoplasty [13]. After balloon dilatation of the inflammatory strictures of the ureter, the rate of long-term recurrence is high and prolonged placement of a JJ stent allows for improvement of the outcomes [15]. We have not used this therapeutic approach with our patients due to the unavailability of the required equipment. However, we did perform a ureterocystoneostomy in twelve cases. It is suitable for treatment of stenosing lesions of the pelvic ureter, and it allows such lesions to be excised and to reestablish ureterovesical continuity. Nonetheless, with UGS, the inflammatory lesions often extend beyond the stenosed ureteral portion. Moreover, the bladder wall (on which the stoma is carried out) is often fibrotic. Anastomosis of the two altered tissues by the same fibrous remnant exposes to recurrence of the stricture [3]. In our series of twelve ureterocystoneostomy, the outcome was favorable in eight cases. Placement of a JJ-stent in six cases for a duration of six weeks after direct ureterocystoneostomy without a submucosal tunnel allowed us to obtain good outcomes [2].

This study shows that sequelae of UGS are frequent and serious. Combating this parasitic disease is based on several approaches, such as raising awareness in the population of its potential seriousness, eradication of the parasite, early diagnosis and treatment particularly in populations at risk, and chemoprevention by mass administration of praziquantel. These actions, which are currently taking place in most of the countries involved need to be pursued and strengthened.

5. Conclusion

UGS is one of the neglected tropical diseases that the World Health Organization is seeking to eliminate. In our study, sequelae were most often noted in young adult men. The most common sequelae were vesical and ureteral calcifications, bladder cancer, and pelvic ureter strictures.

Conflicts of Interest Statement

All the authors do not have any possible conflicts of interest.

References
