Case Report

A unique case of Schistosoma-related ureteral stricture: Diagnosis and surgical reconstruction

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Abbreviations & Acronyms GU = genitourinary UTI = urinary tract infection

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Received 16 April 2023; accepted 26 June 2023. Online publication 6 October 2023 **Background:** Chronic infection with *Schistosoma haematobium* can lead to pathology of the upper and lower urinary tracts. While well known as a cause of squamous cell carcinoma of the bladder, relatively little research exists on ureteral involvement. Here, we present a unique case of bilateral ureteral obstruction from schistosomiasis with concomitant ureteral stone disease.

Case presentation: A 43-year-old male Somalian immigrant was diagnosed with a right proximal ureteral stone and bilateral multifocal ureteral narrowing causing obstruction with preserved renal function. He underwent a staged repair with right robotic pyelolithotomy and non-transecting ureteroureterostomy, followed by left robotic ureteroureterostomy with stricture excision. Pathology revealed *Schistosoma* ova.

Conclusion: Ureteral stricture from schistosomiasis represents a rare diagnosis for urologists in non-endemic countries. Bilateral ureteral narrowing and concomitant ureteral stone burden presented both diagnostic and reconstructive challenges, requiring a staged repair. Minimally invasive reconstruction was achieved using robotic assistance with good functional outcome.

Key words: robotic surgical procedures, Schistosomiasis, *Schistosomiasis haematobium*, ureteral calculi, ureteral obstruction.

Keynote message

Ureteral stricture resulting from chronic Schistosomiasis is rare and no consensus exists on surgical management. A unique case of bilateral ureteral stricture with concomitant ureteral stone disease was managed surgically. Staged bilateral robotic reconstruction was achieved with good functional outcome.

Introduction

Schistosomiasis is caused by parasitic nematodes in the species *Schistosoma* and is primarily endemic in Africa and the Middle East. There are an estimated 200 million infected individuals across 54 countries,^{1,2} with transmission limited to areas where the intermediate freshwater snail hosts reside. Although transmission does not occur within the United States and many other developed countries, over 400 000 infected emigrants are estimated to reside in the United States, requiring physicians to recognize the disease's clinical manifestations and associated treatments.

Most human infections are caused by *Schistosoma haematobium*, leading to a range of urogenital manifestations, including mild benign lesions, high-grade ureteral strictures, and the development of squamous cell carcinoma of the bladder. Eggs released into circulation become embedded within genitourinary tissues,³ initiating an immunologically mediated foreign body granuloma reaction. With eventual ova destruction, these lesions are replaced by dense calcifications and fibrosis.^{4,5} In the upper urinary tract, repeat infection can cause ureteral wall fibrosis, leading to degenerated mucosa, obliterated ureteritis, and stricture formation.⁴

Management of ureteral strictures from chronic schistosomiasis depends on the extent and location. While medical therapy may reverse obstruction in some patients, persistent or high-

grade obstruction requires surgical correction.⁶ Historically, this largely mirrors that of more "traditional" stricture etiologies, ranging from endoscopic dilation or incision to more complex reconstruction. The literature is limited primarily to case reports and small studies due to its rarity. Here, we report our experience with bilateral ureteral strictures and concomitant nephrolithiasis.

Case report

A 43-year-old male presented with right flank pain and hematuria. He immigrated to the United States from Somalia at the age of 25 and has a self-reported history of nephrolithiasis requiring multiple previous surgical interventions, including ureteroscopy, with no previous diagnosis of ureteral stricture. Computed tomography revealed bilateral hydronephrosis and bilateral calculi including a 1.2 cm obstructing right proximal ureteral stone (Fig. 1a). There were multiple areas of ureteral narrowing, most prominently in the left proximal ureter (Fig. 1b). Additionally, diffuse calcifications were present within the wall of the bladder and bilateral distal ureters (Fig. 1c). He underwent bilateral ureteral stenting with retrograde pyelography showing strictures at the right proximal ureter, and throughout the left ureter. Subsequent nuclear medicine diuretic renography showed bilateral obstruction with R:L split function of 62:38%. Given the preserved renal function, definitive reconstruction was recommended.

The right side was repaired first due to the concomitant ureteral stone. Using a robotic-assisted laparoscopic approach (DaVinci Xi[®] surgical system), we first performed pyelolithotomy to remove the proximal ureteral and renal stones using a 1.9Fr nitinol-tipped basket inserted through the assistant port. Stricture location was confirmed with retrograde ureteroscopy, approximately 1 cm in length and 5Fr in diameter. A non-transecting Heineke–Mikulicz ureteroureterostomy was performed using 4-0 polyglactin suture over a ureteral stent. The pyelotomy was also closed using the Heineke–Mikulicz technique to account for pelvic redundancy. We performed routine left stent exchanges as a bridge to formal repair, during which right retrograde pyelography demonstrated significantly improved hydroureter and diagnostic left ureteroscopy showed a distinct 1 cm proximal stricture.

Left-sided reconstruction was performed 14 months later, again using the robotic platform. Identification was aided with intraureteral indocyanine green with Firefly[®] technology and retrograde ureteroscopy (Fig. 2a). The stricture was excised and sent for pathology. After ureteral mobilization, a spatulated ureteroureterostomy was performed in an interrupted fashion, allowing for a water-tight, tension-free anastomosis over a ureteral stent (Fig. 2b). Retroperitoneal tissue and omental flaps were utilized to cover the anastomosis. The ureteral stent was maintained for 6 weeks. At 3 months, the patient had not experienced any recurrence of flank pain, UTI, or nephrolithiasis. Renal function has been preserved 1 year postoperatively with resolution of right hydronephrosis and stability of chronic left hydronephrosis.

On pathology, the excised stricture displayed periureteral fibrosis, reactive atypia, and calcifications morphologically consistent with *Schistosoma* ova (Fig. 3). The patient was referred to infectious disease and given praziquantel. On further history, the patient endorsed frequently swimming in Somalian freshwater lakes.



Fig. 1 CT images highlighting the proximal ureteral stone (a), left-sided mid-ureteral narrowing (b), and calcifications with the ureter and bladder (c).

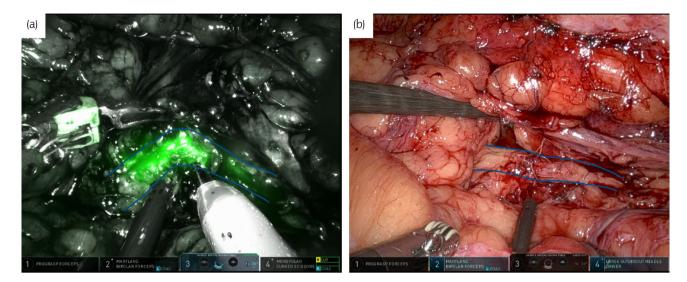


Fig. 2 Intraoperative images showing ICG usage for ureter identification (a) and completed left-sided ureteral reconstruction (b).

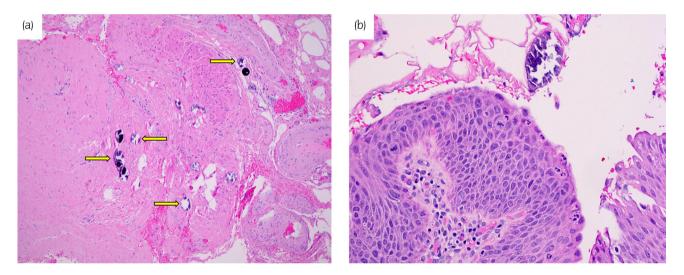


Fig. 3 Histopathological slides highlighting numerous *Schistosoma* ova (a, H&E, 100× magnification) and mucosal inflammation with reactive atypia and scattered mitoses (b, H&E, 400× magnification) Ova were identified within the ureteral lumen, smooth muscle, and periureteral soft tissue.

Discussion

Early ureteral involvement from *S. haematobium* ova deposition typically results in peristaltic dysfunction rather than obstructive pathology and rarely causes renal damage.^{7,8} Hydronephrosis develops slowly, often unilateral and asymptomatic. With chronic exposure, lesions are replaced by calcifications and fibrosis, creating the ureteral and bladder pathologies observed. As was demonstrated here, imaging often demonstrates calcifications within the bladder and/or ureteral wall (Fig. 1a), and in some men prostatic and seminal vesical calcification.⁷ Careful imaging review in patients with risk factors for GU schistosomiasis is therefore recommended.

There is no consensus on optimal surgical management of ureteral stricture disease from chronic schistosomiasis. Endoscopic management is well-described, but discouraged by the European Association of Urology⁶ due to concerns about long-term efficacy. Balloon dilation appears more durable than mechanical dilation which is prone to high recurrence rates.^{9,10} Formal reconstruction offers the highest success rate with multiple approaches described including ureteral reimplantation with Boari flap or psoas hitch, buccal graft augmentation, and ileal-ureter replacement.^{11–16} In some extreme cases, bilateral ileal replacement has been performed for complete ureteral involvement.¹⁷ Minimal data exist for robotic surgery in this setting but has been utilized for cystectomy with ileal conduit urinary diversion in a patient with end-stage bladder dysfunction from chronic schistosomiasis.¹⁸

This case highlights the diagnostic and management complexity in the setting of nephrolithiasis and bilateral strictures. His prior stone disease and intervention may have also contributed to stricture formation. Conversely, while chronic schistosomiasis does not appear to directly increase the risk of stone disease,^{19,20} poor drainage from stricture likely prevented stone passage and facilitated growth. Pathological analysis was crucial in establishing the etiology, allowing for medical treatment to eliminate any residual adult worms and prevent recurrent ova deposition.

Conclusion

We have reported our experience using a staged approach for robotic repair of bilateral ureteral strictures from chronic schistosomiasis, resulting in an acceptable functional outcome. This approach allowed for concomitant management of ureteral stones. This case also demonstrates the importance of pathological analysis of excised strictures. While GU schistosomiasis remains a rare diagnosis for most urologists, recognition and proper treatment is an important skill set to maintain.

Author contributions

Jonathan A. Seaman: Conceptualization; methodology; writing – original draft; writing – review and editing. Erika Bracamonte: Visualization; writing – review and editing. Sunchin Kim: Conceptualization; methodology; supervision; writing – review and editing.

Conflict of interest

The authors report no financial or ethical conflicts of interest associated with this research.

Approval of the research protocol by an Institutional Reviewer Board

Study determined to be exempt from IRB. Study ID STUDY00002584, University of Arizona Human Subjects Protection Program, Tucson, AZ.

Informed consent

Not Applicable.

Registry and the Registration No. of the study/trial

Not Applicable.

References

- 1 James SL, Abate D, Abate KH et al. Global, regional, and national incidence, prevalence, and years lived with disability for 354 diseases and injuries for 195 countries and territories, 1990–2017: a systematic analysis for the global burden of disease study 2017. Lancet 2018; 392: 1789–858.
- 2 Verjee MA. Schistosomiasis: still a cause of significant morbidity and mortality. *Res. Rep. Trop. Med.* 2019; **10**: 153–63.
- 3 McManus DP, Dunne DW, Sacko M, Utzinger J, Vennervald BJ, Zhou XN. Schistosomiasis. Nat. Rev. Dis. Primer 2018; 4: 13.
- 4 Khalaf I, Shokeir A, Shalaby M. Urologic complications of genitourinary schistosomiasis. World J. Urol. 2012; 30: 31–8.
- 5 Barsoum RS. Urinary schistosomiasis: review. J. Adv. Res. 2013; 4: 453-9.
- 6 Bichler KH, Savatovsky I, Naber KG et al. EAU guidelines for the management of urogenital schistosomiasis. Eur. Urol. 2006; 49: 998–1003.
- 7 Shebel HM, Elsayes KM, Abou El Atta HM, Elguindy YM, El-Diasty TA. Genitourinary schistosomiasis: life cycle and radiologic-pathologic findings. *Radiographics* 2012; **32**: 1031–46.
- 8 Umerah BC. Evaluation of the physiological function of the ureter by fluoroscopy in Bilharziasis. *Radiology* 1977; **124**: 645–7.
- 9 Wishahi MM. The role of dilatation in bilharzial ureters. Br. J. Urol. 1987; 59: 405-7.
- 10 Jacobsson B, Lindstedt E, Narasimham DL, Sundin T, Vijayan P. Balloon dilatation of bilharzial ureteric strictures. Br. J. Urol. 1987; 60: 28–32.
- 11 ElAbd SA, ElShaer AF, ElMahrouky AS, ElAshry OM, Emran MA. Longterm results of endourologic and percutaneous management of ureteral strictures in bilharzial patients. J. Endourol. 1996; 10: 35–43.
- 12 Ravi G, Motalib MA. Surgical correction of bilharzial ureteric stricture by Boari flap technique. Br. J. Urol. 1993; 71: 535–8.
- 13 Pal PO, Smith RD, Allen S et al. Schistosomiasis—a disobedient ureter, a disobedient diagnosis. J. Endourol. Case Rep. 2017; 3: 114–8.
- 14 Mee AD, Youssef AMR. The surgical management of bilharzial strictures of the ureter. Br. J. Urol. 1982; 54: 103–5.
- 15 Lorca J, Hevia V, Diez Nicolás V, González A, Sánchez Guerrero C, Burgos Revilla FJ. Minimally invasive resolution of a left ureteral stenosis after *Schistosoma haematobium* infection. Urol. Case Rep. 2019; 25: 100889.
- 16 Cornet L, Neretti J, Subreville C. Uretero-ileoplasty in bilharzian ureterohydronephrosis. J. Chir. (Paris) 1976; 111: 417–30.
- 17 Elem B. Preliminary nephrostomy and Total Ileal replacement of both ureters in advanced Bilharzial obstructive Uropathy. Br. J. Urol. 1989; 63: 453-6.
- 18 Pollock GR, Meiklejohn KM, Zeng J, Chipollini J. Robotic cystoprostatectomy with Intracorporeal Ileal conduit diversion in a patient with chronic schistosomiasis. Urology 2020; 141: e8–9.
- 19 Ibrahim A. The relationship between urinary bilharziasis and urolithiasis in the Sudan. Br. J. Urol. 1978; 50: 294–7.
- 20 Cutajar CL. The role of schistosomiasis in urolithiasis. Br. J. Urol. 1983; 55: 349–52.