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Peritoneo-Vesical Shunt: A Feasible Therapeutic Alternative for Refractory Ascites in the Low-Income Setting

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Abstract

Introduction: Schistosomiasis is a tropical parasitic disease with a very high prevalence worldwide, carrying a considerable risk for serious morbidity and mortality. Hepatic schistosomiasis may lead to chronic ascites due to portal hypertension (Increased hydrostatic pressure due to periportal fibrosis).

Presentation of Case: We describe the case of a 67-year-old African female with refractory ascites secondary to hepatic schistosomiasis, causing her respiratory distress. At least mid-term, she was successfully treated with a peritoneo-vesical shunt, allowing the controlled passage of ascitic fluid from the peritoneum into the bladder, enabling her to "urinate" the liquid, releasing the abdominal pressure and improving her respiratory status. She was able to return to her normal activities during months postoperatively.

Discussion: Schistosomasis is quite prevalent in tropical endemic areas, leading to serious complications. Patients with hepatic schistosomiasis develop secondary ascites, which is frequently refractory to standard medical treatment. Surgical procedures are required to improve the patients quality of life. Although the idea of using a shunt to evacuate peritoneal fluid into the bladder has been reported in a clinical trial, to our knowledge, no previous report of the use of a self-devised, inexpensive, controlled peritoneo-vesical shunt for the treatment of refractory ascites secondary to hepatic schistosomiasis in the low income setting has been previously described in the literature. We propose herein a novel and inexpensive surgical approach that allows the extraction of fluid from the peritoneal cavity, eliminating the liquid through normal urination, and improving the quality of life of the patient.

Conclusion: Even though a wide range of surgical approaches has been devised for the management of refractory ascites, none has so far considered the particularities of resource-limited environments. We believe this to be an innovative and feasible surgical alternative for the management of refractory ascites in the low-income setting.

Background

Schistosomiasis has afflicted mankind for millennia, as documented in historic medical recordings. Abdominal and scrotal swellings due to Schistosoma infection were recognized in the earliest dynasties, and larvae have been isolated from Egyptian and Chinese mommies. In ancient Egypt, male patients with hematuria were known as "menstruating men", and the entity was portrayed in papyri [1].

Schistosomiasis is a tropical and sub-tropical disease caused by larvae trematode of the genus Schistosoma [2]. Theodor Maximilian Bilharz identified and described the cause of "tropical hematuria" in 1851. David F. Weinand introduced the term "schistosomiasis" in 1858 [3].

Schistosomiasis is caused by infection with parasitic blood flukes known as schistosomes, which can be associated with serious morbidity and mortality. According to the World Health Organisation (WHO), 200 million people are currently infected worldwide [4]. However, another report estimates that some 200 million persons are infected in Africa alone [5].

The main species affecting humans include *Schistosoma mansoni*, *S. japonicum*, and *S. haematobium* [4]. The disease usually occurs in individuals living in endemic areas with recurrent exposure, mainly through contaminated water and snails [5].

Although most individuals infected with Schistosoma are asymptomatic, the clinical presentation varies between hosts that live in endemic areas and those who do not. Individual immunity and intensity of the infection account for such differences [6,7]. The stronger immune response after exposure to Schistosoma in non-immune individuals, such as travelers, is associated with a higher incidence of acute symptoms (i.e., Swimmer's itch and Katayama fever). On the other hand, individuals from endemic areas are exposed to a higher infection burden, making chronic complications more prevalent [4,6].

Hepatic schistosomiasis presents as an inflammatory process. Chronic hepatic schistosomiasis is one of the leading causes of non-cirrhotic portal hypertension worldwide [7]. Diffuse collagen deposits in the periportal spaces lead to Symmer's pipestem fibrosis. Such patients develop secondary ascites [4,8,9]. Hepatocellular function is usually preserved. Liver failure with hypoalbuminemia leading to chronic ascites mostly occurs in patients with coexisting liver disease [10-12].

Schistosomiasis is diagnosed by detecting schistosome eggs in stool or urine, by rectal mucosal or liver biopsies, or by various immunologic assays [4,13]. Every patient with evidence of infection should be treated with anthelmintic therapy (e.g., praziquantel) to eradicate the parasite and decrease the amount of egg laying, regardless of symptoms [14-17]. Besides treating the underlying infection, severe portal hypertension and its complications should be managed similarly to patients with non-parasitic cirrhosis, including beta-blockers, variceal sclerotherapy or ligation, and shunting surgical procedures [18-20]. When surgically dealing with parasitic liver cysts, it is important not to contaminate the abdominal cavity with the cystic fluid, since larvae will implant in the peritoneal surface and generate further lesions.

Clinically relevant ascites in patients with cirrhosis is initially managed with dietary sodium and fluid restriction, although most patients will require oral diuretic therapy [21]. Treatment options for patients with diuretic-resistant ascites include optimization of medical therapy, liver transplantation, therapeutic paracentesis, transjugular intrahepatic portosystemic shunt (TIPS) and other invasive treatments, such as Leveen peritoneovenous shunts or surgical portosystemic shunts [21-25], Venzin et al. [25] reported the use of a peritoneo-vesical automated shunt system with a pump implanted in a dog with refractory ascites. The system was effective for 10 weeks, but a skin perforation developed over the pump Bellot et al. [26] reported in 2013 the first non-randomized clinical trial using an automated pump system for cirrhosis-related refractory ascites in 40 patients with a 6-month follow-up. Several surgical complications occurred; however, the system removed 90% of the ascites and significantly reduced the median number of large volume paracentesis per month. Safety was moderate.

In Sub-Saharan Africa though, patients are seldomly recruited for clinical trials and automated pumps are not available. Thus, the technique described herein might be the best alternative in such circumstances.

To our knowledge, no previous case of a simple and inexpensive peritoneo-vesical shunting for the management of schistosomiasis-related refractory ascites has been previously described in the literature.

Presentation of Case

We describe herein the case of a 67-year-old African female farmer, previously diagnosed with hepatic Schistosomiasis by *S. mansoni*, unsuccessfully treated with praziquantel and diuretics. She presented to the surgery ward at Kilimanjaro Christian Medical Center with a four-month history of abdominal swelling, diffuse tenderness and orthopnea.

On physical exam she was pale but well hydrated. She was afebrile, mildly hypertensive (150/90 mmHg), tachypneic (33 breaths per minute), and tachycardic (128 beats per minute). She presented severe diffuse abdominal distension, mild hepatomegaly and abundant ascites. Lower extremity edema was also apparent. The rest of the examination was within normal limits. Laboratory studies were mostly normal except for anemia (hemoglobin of 9.0 g/dL) and discrete hypoalbuminemia (albumin of 2.1 g/dL). Protein content in the ascites fluid was 0.9 g/dL, for an SAAG (serum-ascites albumin gradient) of 1.2 g/dL, considered evidence of a high albumin gradient ascites (transudate).

During the first month of management as an outpatient, she underwent three therapeutic paracentesis in order to improve her respiratory status. An average of 750 ml of fluid was extracted per puncture. However, she returned to the clinic one to two weeks after each drainage, with recurrent ascites and respiratory symptoms, affecting her quality of life. Intravenous albumin replacement did not affect the development of ascites.

There were no Leveen shunts available at the institution, and leaving an external permanent drain was considered high-risk for infection and peritonitis due to her usual field labor. She needed a drainage procedure that would enable her to release the fluid from the peritoneal cavity, but as a closed system to avoid the risk of infection, and with a lock that would permit the system to be opened and closed at will.

We decided to construct a peritoneo-vesical shunt (Figure 1). A Tenckhoff-like catheter was constructed using an 18 Fr silastic tube. Side fenestrations were made on the peritoneal end of the catheter in order to avoid clogging and increase the fluid drainage capacity. Two felt cuffs were glued to it for subcutaneous fixation. It was then sterilized for surgical use.

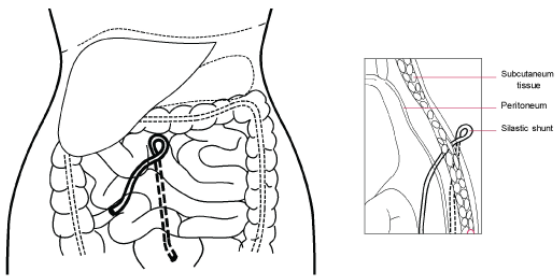

Figure 1: A) Diagram depicting the proximal end of the shunt into the peritoneal cavity, in the right iliac fossa, with an external loop for the valve that allows opening and closure of the system, and the distal tip of the catheter inserted into the bladder with a purse-string suture. B) Lateral schematic view of the external loop of the catheter and the subcutaneous tunnel for the shunt.

Under general anesthesia, a small laparotomy incision was performed, combined with a transverse suprapubic extraperitoneal bladder approach. The proximal end of the shunt was placed intraperitoneally, then a loop of the tube was left external at the abdominal skin, and the distal end of it was tunneled subcutaneously and inserted into the bladder with a double purse-string technique. Cuffs were placed subcutaneously for fixation (Figure 2). A pen cap was secured to the skin near the tubing loop with a thick polyproilene suture, for it to be used as means to close the system. The skin was closed in layers and the patient recovered from surgery uneventfully. She was discharged home on her second postoperative day.

Figure 2: Surgical image depicting the external loop of the silastic shunt, and the subcutaneous tunnel created to implant it into the bladder through a double purse-string insertion.

This way, she was able return to her normal activities, relieving the fluid from the peritoneal cavity and improving her respiratory status. Overnight, the fluid would accumulate in the peritoneum, unable to pass into the bladder due to the cap collapsing the tube at the external loop, while in the morning she could remove the cap from the tubing, opening the system and allowing the fluid to drain into the bladder by abdominal contraction, rendering her able to "urinate" the ascites, after which she could close the system again to avoid continuous micturition. She would have to keep the system locked during most of the day, particularly during normal micturition in order to avoid back flow of urine into the peritoneum.

She was evaluated weekly after discharge. Serum albumin levels were measured monthly, which evidenced a slow and mild increase over time. On the first couple follow-up visits, she was asked to skip the morning release of peritoneal fluid to do so at the hospital. An abdominal ultrasound was performed and then she was asked to release the peritoneal fluid, and the abdominal ultrasound repeated afterwards. Decrease in 80% of intraperitoneal fluid volume was documented in both occasions. The patient referred to be quite satisfied with the surgical results and had a substantial improvement in her quality of life. She did not complain about abdominal pain or fever during the time we were able to see her. However, she was lost to follow-up after her last visit to the hospital five months after the surgery. On that last visit, the system was functional and effective.

Discussion

Schistosoma parasitic infections have long been recognized to affect humans. The disease is currently quite prevalent in tropical endemic areas. Severe chronic exposure leads to several serious complications. Pathophysiology of peritoneal fluid in patients with hepatic schistosomiasis is frequently due to portal hypertension (hydrostatic pressure) rather than hypoalbuminemia (decreased oncotic pressure). In this specific patient, low albumin was considered to be secondary to malnutrition. Patients with hepatic schistosomiasis develop fibrosis ensuing portal hypertension with its usual complications. Ascites is a common secondary malady and as the disease becomes chronic, it becomes refractory to standard medical treatment. In such patients, invasive surgical procedures are required to improve their quality of life. These include repeated paracentesis, Leveen peritoneo-venous shunts, and porto-systemic vascular surgical shunts. To our knowledge, no report of the use of a simple, inexpensive and self-made peritoneo-vesical shunt has been previously described in the literature.

In order to decide on such a procedure, the species of Schistosoma causing liver disease was taken into consideration. As mentioned above, the main species of Schistosoma causing hepatic schistosomiasis is different from the one causing urinary infections, thus, minimizing the chance to develop bladder disease, which is extremely uncomfortable since it generates frequent urination, pain and tenesmus.

Although a proper clinical trial with more patients and longer follow-up would be ideal to evaluate this technique and better define the short-comings of the procedure and study of complications, we consider this approach to be a feasible and viable surgical alternative for alleviating non-cirrhotic chronic and refractory ascites. It allows for the extraction of the fluid from the peritoneal cavity, without causing extraperitoneal disease, with a closed system to decrease the risk of infection, and a flow control lock that allows the patient to open and close the system when necessary, preventing the continuous flow of fluid into the bladder. Pressure within the peritoneal cavity equals that of the bladder. However, abdominal muscle contraction increases the peritoneal pressure allowing passage of fluid into the bladder, eliminating the fluid through normal urination.

Surgery is an essential component for the treatment of many medical conditions, and in such field, the lack of resources stimulates creativity. Newer creative and problem-solving initiatives must be considered as essential conditions when dealing with complex disease entities in low- and middle-income countries. Some of these surgical challenges may be solved with innovative techniques despite the scarce available literature; therefore, this aspect deserves to be revisited. Peritoneo-vesical shunts are an example of a new prompting solution for the treatment of refractory ascites in the low-income setting. While this is useful as an initial treatment, further research and longer follow-up is required.

Conclusion

Even though the past decade has devised a wide range of new approaches for the management of refractory ascites, none has so far considered the particularities of resource-limited environments.

We believe peritoneo-vesical shunting to be a viable and useful alternative for temporary treatment of refractory ascites in low-income settings. Further research is advised.

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