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Pain Management Techniques for Loin Pain Hematuria Syndrome Case Report

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Background: Loin pain hematuria syndrome (LPHS) is a rare syndrome presenting with chronic unilateral or bilateral flank pain and gross or microscopic hematuria.

Case Report: We present a case of LPHS in a male with past medical history of atrial fibrillation, Barrett's esophagus, and loin pain hematuria syndrome who initially presented to our medical facility with gross hematuria, intractable left flank pain, and non-bloody emesis. Our case exemplifies the challenge in managing patients with LPHS.

Conclusion: Without a definitive mechanism for LPHS, current treatment focuses on symptom management. Analgesics (non-opioid and opioid) may be used orally or intravenous depending on the presence of concomitant nausea and vomiting. For patients at risk of opioid dependence, targeting afferent pain fibers originating from the kidneys and ureters has proven a successful analgesic strategy. Surgical options for renal denervation include: endovascular radiofrequency ablation, laparoscopic renal denervation, and renal auto transplant. Treatment options for this disease process include intra-ureteric bupivacaine, renal denervation, and possible spinal cord stimulation.

Key words: Flank pain, loin pain hematuria syndrome, renal denervation, spinal cord stimulation

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BACKGROUND

Loin pain hematuria syndrome (LPHS) is a rare syndrome presenting with chronic unilateral or bilateral flank pain and gross or microscopic hematuria. It affects primarily women with variation in duration and intensity of pain. Pain is localized to the costovertebral angles and may radiate to the abdomen, inguinal area, and thigh (1). The most common cause of hospitalization in patients with LPHS is due to nausea and vomiting associated with severe pain (2). LPHS has not been shown to worsen kidney function or increase mortality, and cases of spontaneous resolution have been documented (1). LPHS is not well characterized, and without consensus on diagnostic criteria it remains a diagnosis of exclusion.

The pathophysiology of primary LPHS remains to be elucidated. Postulated mechanisms include renal obstruction due to intratubular deposition of calcium or uric acid microcrystals (2), renal vasospasm (3), coagulopathy (3), hypersensitivity (4), complement activation (4), and somatoform pain disorder (5). Renal biopsies of patients with primary LPHS revealed red blood cells in renal tubules as well as irregularities in glomerular basement membrane thickness in more than 50% of biopsies (2). In particular, thin glomerular basement membrane (TGBM) disease has been documented in patients with LPHS as the sole anatomic abnormality on renal biopsy (6). Up to 50% of patients with LPHS had a history or radiographic finding of nephrolithiasis consistent with reports of increased risk of nephrolithiasis in TGBM disease (2,7). These findings implicate microscopic renal obstruction leading to hematuria and pain from distension of renal fascia. The role of elevated C-reactive protein and D-dimer levels in patients with LPHS requires further investigation (3).

CASE PRESENTATION

We present a case of LPHS in a man with a past medical history of atrial fibrillation, Barrett's esophagus, and LPHS who initially presented to our medical facility with gross hematuria, intractable left flank pain, and nonbloody emesis. His pain was uncontrolled with hydromorphone extended release 8 mg oral, methadone 5 mg oral, and duloxetine 50 mg oral that he had been taking as maintenance therapy. By the time of initial presentation, he had been diagnosed with LPHS for several years, which had been complicated by Barrett's esophagus due to recurrent vomiting. He had undergone a renal autotransplant and denervation procedure of his left kidney. He reported numerous hospitalizations for intractable flank pain associated with nausea and vomiting but the hospitalizations decreased after the renal autotransplantation/denervation procedure.

Computed tomography (CT) imaging of the abdomen and pelvis at admission revealed mild perinephric fatty infiltration and scarring consistent with surgical history as well as a calcification along the path of the mid ureter likely representing a small ureteral stone. Lab work was remarkable for mild anemia and leukocytes, nitrites, and red blood cells noted on urinalysis. The patient's history was confirmed with his primary nephrologist and online prescription-monitoring program. He was admitted for treatment with intravenous hydromorphone, intravenous diphenhydramine, and intravenous ondansetron for a total of 4 days. Although urine culture was negative, in the setting of LPHS exacerbation and positive urinalysis he was treated with ceftriaxone for possible urinary tract infection. Subsequent renal ultrasound and CT imaging did not reveal evidence of nephrolithiasis, and urine toxicology was positive for opioids and methadone consistent with his treatment regimen. The patient is currently under evaluation for left ureteral denervation and left nephrectomy.

DISCUSSION

Our case exemplifies the challenge in managing patients with LPHS. Without a definitive mechanism for LPHS, current treatment focuses on symptom management. Analgesics (nonopioid and opioid) may be used orally or intravenously depending on the presence of concomitant nausea and vomiting. For patients at risk of opioid dependence, targeting afferent pain fibers originating from the kidneys and ureters has proven a successful analgesic strategy. While intraureteric capsaicin may provide short-term relief (average 17 weeks), it has been associated with worsening symptoms in more than 50% of patients (8). Intraureteric bupivacaine and CT-guided renal hilar blockade have been utilized to identify patients who may benefit from renal denervation for LPHS (9,10). Treatment options for renal denervation include: percutaneous pulsed radiofrequency ablation of the splanchnic nerves (11), endovascular radiofrequency ablation (12), laparoscopic renal denervation, and renal autotransplant (13). Nephrectomy is considered a last resort and is usually reserved for patients with poor renal function.

More recently, neuromodulation via dorsal root ganglion and spinal cord stimulation has been shown to successfully decrease pain due to LPHS (14,15). Despite renal autotransplant and denervation of his left kidney, our patient continued to present with left

flank pain, nausea, and vomiting, which may implicate a psychopathology either secondary to substance use disorder from prior opioid treatment or as the primary etiology of LPHS. Our patient did not otherwise show signs of drug-seeking behavior and was compliant with the treatment regimen as verified by urine toxicology screening and online prescription monitoring. Further research into the pathophysiology and treatment of LPHS is warranted.

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