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Case Report

Urinothorax: A rare complication of percutaneous nephrostomy

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ABSTRACT

We present a case of a urinothorax resulting from treatment of genitourinary pathology. The presentation, diagnosis, and management of a 46-year-old female with an urinothorax are discussed. Urinothorax is a rare cause of a pleural effusion, most commonly arising from a traumatic etiology. Imaging can be crucial in the diagnosis, particularly computerized tomography (CT), which can help characterize any associated causative genitourinary abnormalities such as anatomical defects or a urinoma. A urinothorax is often posttraumatic in etiology, associated with the treatment of genitourinary pathology, as in this case. Treatment of the source of the urine leak is required to properly manage an urinothorax and often requires a multi-disciplinary approach.

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Introduction

Urinothorax is defined as urine in the pleural cavity, a rare cause of a pleural effusion. It was first reported by Corriere et al. in 1968 during experiments involving ureteral obstruction in mongrel dogs. The study demonstrated a direct relationship between hydronephrosis and pleural effusion [1]. Between 1968 and 2006, 58 cases of urinothorax have been reported, increasing the awareness of this diagnosis [2]. This recognition, along with more sophisticated imaging, a better understanding of this entity, and further studies involving biochemical analysis of pleural fluid have all resulted in earlier diagnosis [2,3]. The presentation, diagnosis, and management

of a 46-year-old female with an urinothorax resulting from treatment of genitourinary pathology is discussed.

Case report

A 46-year-old female presented to the emergency department (ED) with a 2-day history of progressively worsening right-sided flank pain, nausea and vomiting, gross hematuria, and decreased urinary output. A computed tomography (CT) renal stone study showed a 1 cm right proximal ureteral stone with moderate obstructive uropathy as well as several nonobstructing left lower pole renal stones, the largest being 1.2 cm. The patient was admitted and the obstructing stone burden

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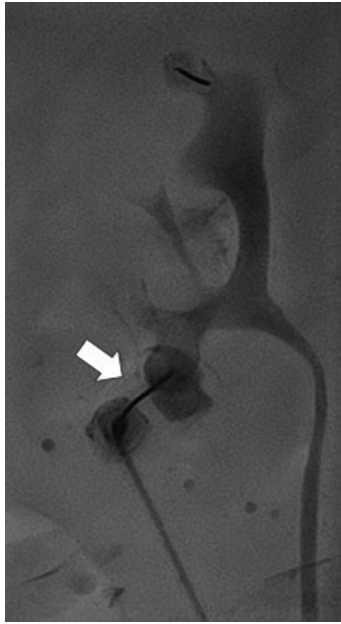


Fig. 1 – Spot fluoroscopy image from a percutaneous nephrolithotomy access procedure demonstrating a lower pole puncture directly into calculi (white arrow), used to opacify an upper pole calyx for a second puncture.

was cleared by a retrograde urologic procedure. The patient was discharged the following day with plans to follow-up in urology clinic.

At this 1 month follow-up visit, she was doing well and elected to proceed with clearing her remaining nonobstructing left-sided stone burden. She was scheduled for a left-sided percutaneous nephrolithotomy, including access by interventional radiology, 1 month following this appointment. The pre-screening ultrasound on the day of the procedure demonstrated no hydronephrosis. Therefore, a lower pole puncture (directly into the stone) was used to opacify an upper pole calyx for a second puncture (Fig. 1). A wire was advanced down the ureter from the upper pole access, which was subsequently used for percutaneous shockpulse nephrolithotomy with residual stone burden at the conclusion of the procedure due to inability to obtain lower pole access; otherwise, there were no immediately apparent complications. A Foley catheter, double-J (JJ) ureteral stent, and 22 French upper pole percutaneous nephrostomy tube were left in place, and the patient was observed overnight. The Foley catheter and nephrostomy tube were removed on postprocedural day 1 with minimal bleeding. She had no issues voiding; however, shortly thereafter, the patient developed shortness of breath. A stat chest radiograph was obtained showing mild blunting of the left costophrenic angle suggesting a small pleural effusion with no evidence of pneumothorax (Fig. 2). She was kept in the hospital for a second night but was discharged the next day given her improvement in symptoms and stable vital signs.

Ten days following discharge, she presented to the ED for a second time with progressively worsening shortness of breath, left-sided chest tightness, and new left-sided flank pain. Chest radiograph at this time revealed a large left-

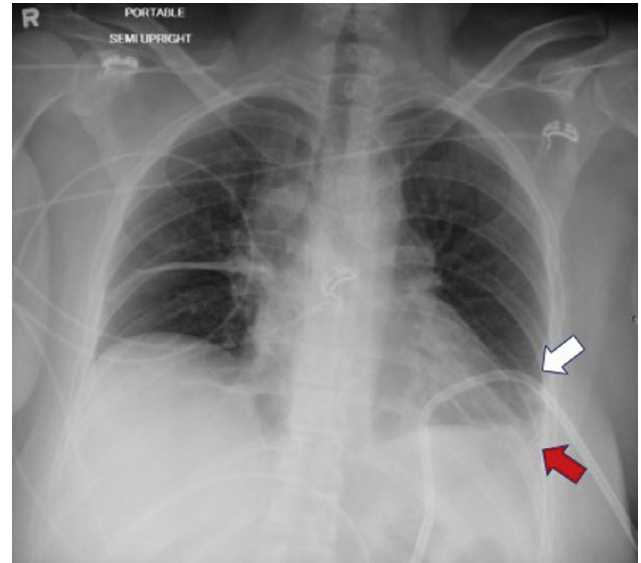


Fig. 2 – Chest radiograph on postoperative day 1 status post percutaneous nephrolithotomy demonstrating a 22 French left upper pole percutaneous nephrostomy coursing above diaphragm (white arrow). A small left sided pleural effusion is also noted (red arrow).

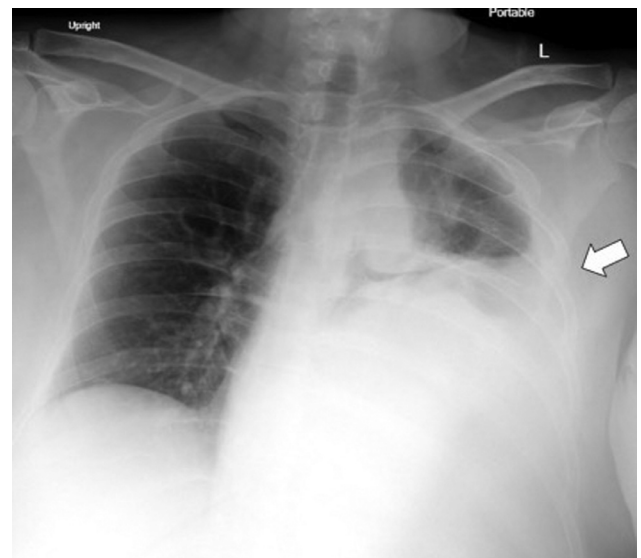


Fig. 3 – Chest radiograph on postoperative day 10 status post percutaneous nephrolithotomy demonstrating interval development of a large left pleural effusion (white arrow) 9 days following nephrostomy removal.

sided predominantly mid and lower lung zone opacity occupying greater than two-third of the left hemithorax and causing obscuration of the left heart border and hemidiaphragm. There was no significant mediastinal shift and no evidence of pneumothorax (Fig. 3). Dedicated chest CT with intravenous contrast was obtained for further evaluation and confirmed the presence of the large left pleural effusion. The effusion was again noted to occupy greater than two-third of the left



Fig. 4 – Sagittal reformatted computed tomography image demonstrating a large left sided pleural effusion (white arrow) as well as a tract (red arrow) extending from the upper pole of the left kidney to the hemidiaphragm.

hemithorax and showed multiple loculations. A linear hypodense tract, compatible with a fistula, was seen extending from the posterior aspect of the superior pole of the left kidney superiorly to the left hemidiaphragm (Fig. 4). An ultrasound guided diagnostic thoracentesis was performed, and pleural fluid demonstrated laboratory values suggestive of an exudative effusion. Pleural white blood cell count and red blood cell count were elevated at 4343 per uL and 36,182 per uL, respectively, of which there are no reference range (ref range) values for and must be interpreted within clinical context. Pleural lactate dehydrogenase was elevated at 392 U/L (ref range <201 U/L) compared to plasma concentration of 102 U/L (ref range <201 U/L) for a pleural lactate dehydrogenase/plasma lactate dehydrogenase ratio of 3.8. Pleural glucose concentration was low at less than 10 mg/dL (ref range 65–99 mg/dL) as was the pleural pH at 6.0 (ref range unestablished and must be interpreted within clinical context). Pleural creatinine concentration was elevated at 54.08 mg/dL (ref range unestablished and must be interpreted within clinical context) compared to her plasma concentration of 0.43 mg/dL (ref range 0.5–1.10 mg/dL). Her pleural fluid/serum creatinine ratio was 125 (ref range <1). All of the imaging and laboratory findings were compatible with urinorhorrax, particularly the extremely elevated pleural-to-serum creatinine ratio of 125 (ref range <1). The urinorhorrax was confirmed to be the result of a fistula, best depicted at CT (Fig. 4), between the genitourinary tract and the pleural space that had resulted as a consequence of the percutaneous nephrolithotomy. No further diagnostic imaging workup was felt to be necessary in light of this finding.

The JJ ureteral stent had remained in place since the procedure and multiple attempts at percutaneous drainage of the pleural effusion were made, including a total of 4 pigtail

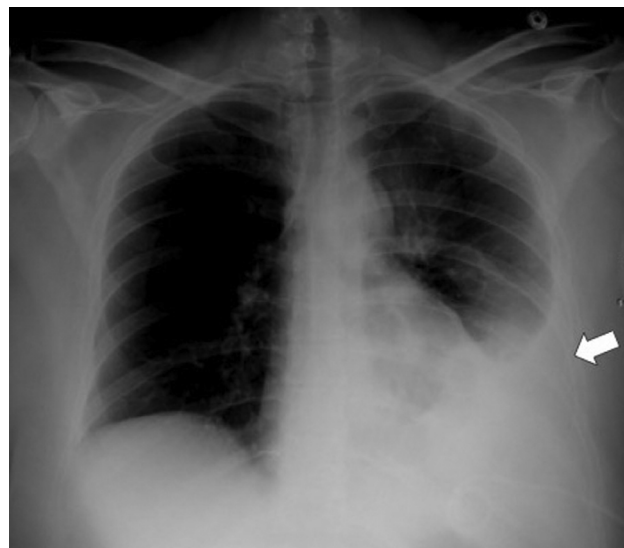


Fig. 5 – Chest radiograph following a left pigtail chest tube insertion demonstrating a residual left-sided pleural effusion (white arrow).

catheter insertion (Fig. 5). Complex fluid accumulation continued to occur rapidly following multiple drainages over the course of this second hospital stay, approximately 2 weeks following the Percutaneous nephrolithotomy (PCNL) procedure. Definitive treatment was decided upon in a multidisciplinary conference at this time. A thoracotomy and decortication of the left lung with mechanical pleurodesis and wedge resection was then performed (3 weeks following the PCNL) secondary to pulmonary injury sustained during lysis of fibrous inflammatory adhesions. On postoperation day 4, she was found to be medically stable for discharge. At 4 weeks following the operation, the patient was recovering well, and her JJ ureteral stent was removed. The patient continued to do well with no evidence of recurrent pleural effusion at 3.5 months post percutaneous nephrolithotomy and 3 months post video-assisted thoracoscopic surgery (Fig. 6).

Discussion

Etiology

The most common etiology of an urinorhorrax result following urine extravasation from the genitourinary tract leading to the formation of an urinoma. Urinomas classically arise following obstructive uropathy, trauma, or iatrogenic injury to the urogenital system. Some have reported that urinomas may form following vesicoureteral reflux secondary to high-pressure retention of urine such as in cases of bladder outlet obstruction [4]. Other cases of urinorhorraxes have been reported in the setting of genitourinary malignancies, metastatic disease, renal cysts, renal biopsies, renal transplantation, retroperitoneal inflammatory processes, and pregnancy [5–7]. The mechanism of urine transit across the diaphragm and into the pleural space is often debated, but 2 the-



Fig. 6 – Chest radiograph following thoracotomy, decortication, pleurodesis and left lung wedge resection demonstrating near complete resolution of the left-sided pleural effusion. The image was acquired approximately 3.5 months following percutaneous nephrolithotomy and 3 months post video-assisted thoracoscopic surgery.

ories predominate. The first is that diaphragmatic lymphatic drainage of urine results from an increased pressure in the retroperitoneum, typically secondary to a urinoma. The second theory is that the passage of urine into the pleural space occurs via anatomical defects in the diaphragm, often having developed following trauma. Given the low position of the diaphragm in conjunction with the very posterior location of the costophrenic sulcus and pleural space, it is conceivable how one could readily traverse the pleural space when obtaining access to the urinary tract [1,2,8,9]. Most cases of urinothoraces are unilateral and occupy greater than two-thirds of the hemithorax, although some cases of bilateral pleural involvement have been reported [5].

Presentation

An urinothorax can occur at any age, with the median age being at 45 years, and more commonly present in males than in females (male/female ratio 1.6/1). Onset of presentation is typically acute, and patients often present with dyspnea, chest pain, abdominal pain, and reduced diuresis [5]. In the setting of obstructive uropathy, patients are often febrile and exhibit flank pain. Patients may also present with tachycardia and tachypnea. Cough and ascites may be present, but more often than not are absent [5]. Physical exam typically reveals decreased respiratory movements, unilateral dullness to percussion, and absent breath sounds within the affected thorax. One should have a high degree of clinical suspicion for an urinothorax when patients present with any of these symptoms with urinary tract obstruction accompanied by a retroperitoneal urinoma [9].

Imaging

Imaging is useful in aiding with the diagnosis, and there are multiple modalities available. A chest radiograph may show an ipsilateral, or very rarely, a contralateral, pleural effusion, sometimes associated with lung atelectasis and/or mediastinal shift [8]. An intravenous pyelogram is another useful modality, sometimes revealing extravasation of contrast from the genitourinary tract into the pleural space. CT and ultrasound are both useful in determining any underlying genitourinary tract or abdominal pathology which may be driving the urinothorax formation [3]. Intravenous contrast enhanced CT may be useful in revealing a reno-pleural fistula, and even in some reported cases, noncontrast CT has utility in demonstrating a fistula [10]. There have been multiple reported cases of urinothoraces in patients in whom an urinoma was also present, suggesting the possibility of a retroperitoneal urinoma being a predisposing factor for the development of an urinothorax. On ultrasound, an urinoma may show a well-defined anechoic or hypoechoic fluid collection, partially contouring any portion of the genitourinary system. On CT, urinomas show water attenuation, and on the excretory phase of contrast-enhanced studies, urine leakage may be visualized from contrast extravasation from the genitourinary tract [11]. Renal scintigraphy has also been reported to serve a role in detecting an urinothorax [12,13]. Both Technetium-99m-diethylene-triamine-pentaacetate and Technetium-99m-mercaptoacetyltryglycine are excreted entirely by the kidney, allowing a technetium-99m renal scan to confirm the diagnosis by revealing the presence of technetium-99m labeled albumin from the genitourinary tract in the pleural space [6,12,13].

Diagnosis

Diagnosis can also be made by biochemical analysis of the pleural fluid. The pleural fluid typically has the same color and odor of urine. An elevated pleural fluid/serum creatinine ratio >1 is considered the hallmark of an urinothorax, although it is not entirely specific for diagnosis [14,15]. Other pleural fluid characteristics of an urinothorax are clear-yellow in color, paucicellular, transudative by Light's criteria, low glucose concentration, and a total pleural fluid protein <1 mg/dL [6,14]. The combination of both a pleural fluid analysis, supportive radiologic evidence, and the right clinical context is best in confirming the diagnosis of an urinothorax. In this case, CT evidence and pleural fluid analysis in the clinical setting of setting traumatic renopleural fistula confirmed the diagnosis.

Management

Prompt recognition of an urinothorax is very important in management. Management of an urinothorax typically involves the treatment of the underlying uropathy. This requires a multidisciplinary approach involving multiple specialists. Thoracentesis is performed for both diagnostic and therapeutic purposes, however persistence of an urinothorax occurs until underlying genitourinary tract pathology has been corrected [2,5,6].

In this case, urinorhox persisted despite thoracostomy, decreased stone burden and stenting of the genitourinary tract. Insufficient decompression of the genitourinary tract, including stent occlusion, residual stones or distal migration of stone fragments, may have ultimately necessitated the addition of surgical pleurodesis.

Conclusion

In conclusion, one must have a high degree of clinical suspicion for an urinorhox when patients present with dyspnea, chest pain, and urologic symptoms, especially in the setting of obstructive uropathy. Many imaging modalities are available to aid in diagnosis, including but not limited to, chest radiograph, intravenous pyelogram, ultrasound, and CT. Imaging can support the clinical diagnosis in the appropriate clinical context or even clinch the diagnosis, for example in the elucidation of a reno-pleural fistula at CT. Patients require an early thoracentesis, not only for therapeutic purposes, but also for a biochemical pleural fluid analysis. On pleural fluid analysis, an elevated pleural fluid/serum creatinine ratio >1 is indicative of an urinorhox, although it is not entirely specific for the diagnosis. Radiologic evidence, pleural fluid analysis, and proper clinical setting are best for confirming diagnosis. In many instances, such as in this case, a urinorhox is the consequence of trauma to the urinary tract associated with treatment of genitourinary pathology. Management is therefore directed at the source of the urine leak [15,16]. As such, clinical suspicion should include the pertinent history of a recent procedure in conjunction with the appropriate symptomatology. It is critical that clinicians recognize the signs and symptoms of urinorhoxes early in the clinical course, as favorable outcomes are often achieved.

Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.radcr.2019.03.022](https://doi.org/10.1016/j.radcr.2019.03.022).

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