

Urinothorax: Case report and systematic review of the literature

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Abstract

Urinothorax, the presence of urine in the pleural space, is a rare cause of pleural effusion, usually associated with obstructive uropathy, or urinary trauma. We present the case of a 3 year-old boy and a systematic review of the literature of the 44 cases encountered. After resection of a Wilm's tumour in the right kidney our patient presented acute respiratory distress associated with radiographically confirmed pleural effusion. With the initial diagnosis of pneumonia or malignant pleural effusion, a closed thoracotomy was performed. The liquid obtained suggested urine, which was confirmed by the laboratory. Cystoscopy with retrograde pyelography detected a fistula on the posterior wall of the right kidney. The report of cases worldwide is low, probably due to its low incidence but also to underdiagnosis. Respiratory symptoms are not always present and urological symptoms usually predominate. Diagnosis requires a high degree of clinical suspicion and is confirmed by the main biochemical marker: The ratio > 1.0 pleural fluid creatinine and creatinine serum.

Key Words: Pleural effusion, urinothorax, urological surgical procedures, Wilms tumor

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INTRODUCTION

Urinothorax is a rare cause of pleural effusion in patients with obstructive or traumatic uropathy.^[1-5] It has been reported in patients with blunt trauma or after abdominal surgical procedures.^[6] Disruption of the urinary tract leads to accumulation of urine in the retroperitoneal space that leaks into the pleural space.^[5,7] It is usually ipsilateral to the lesion side.^[8,9] Diagnosis can be done with renal scintigraphy with a radioactive tracer,^[10,11] or measuring creatinine levels in pleural fluids.^[12] Frequently reported in adults; we present

the case of a 3-year-old boy and a systematic review of the literature.

CASE REPORT

A 3-year-old boy was admitted for surgical treatment of Wilm's tumor in the right kidney. The patient had a history of chronic kidney disease, secondary hypertension and had undergone a left nephrectomy 2 years before due to the same condition. The surgical procedure, partial nephrectomy with total resection of the tumor and suture of the renal pelvis, was uneventful. Central venous and peritoneal catheters were placed. On postoperative day 3 the patient developed acute respiratory distress, and a chest X-ray revealed right pleural effusion, which affected two-thirds of the right hemithorax [Figure 1]. Diagnoses of pneumonia and parapneumonic effusion were considered. A thoracostomy was performed, and 1,050 ml of yellowish color liquid were drained during the following 20 h, during which he also

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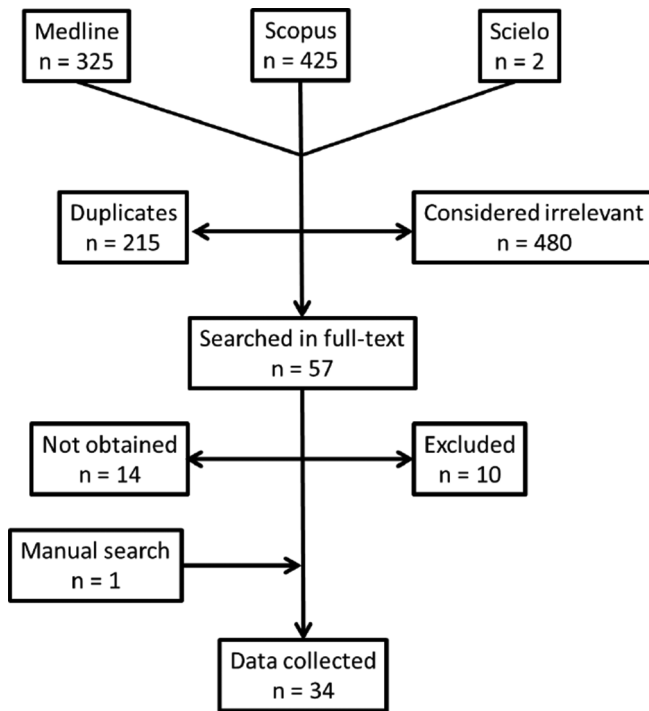


Figure 1: Systematic review flowchart

developed oliguria and fever. He received treatment with antibiotics (meropenem and vancomycin). The aspect and particularly, the smell of pleural liquid oriented the diagnosis of urinothorax. The ratio of creatinine levels in pleural fluid and blood was 4.8, which was highly suggestive of urine. Excretory urography was not performed due to the high risk of inducing renal failure.

Cystoscopy with retrograde pyelography showed a low flow fistula on the posterior wall of the right renal calyx. A ureteral catheter was placed during the surgical closure of the fistula, and nutritional support was initiated, 3 days after thoracotomy pleural effusions disappeared. Pathology confirmed contralateral Wilm's tumor (grade 5). He remained with the ureteral catheter 6 months until achieving complete closure of the fistula and was asymptomatic on last clinical control, 3 months later.

METHODS

We performed a systematic review of the literature, following a standard methodology^[13] in three databases (PubMed, Scopus and SciELO, a Latin American database), using "pleural effusion" and "urine" as free terms, with no language or time limits. A thorough review process ended up with a total of 752 references [Figure 2], 23 of which were published before 1998 (of which only 9 were located and retrieved full-text).^[14-22] Of the 57 articles in full-text, ten were excluded because they were not cases of urinothorax.

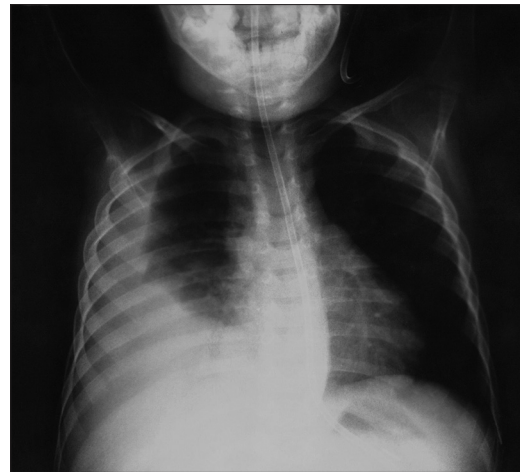


Figure 2: Chest radiography of a 3-year-old boy with right pleural effusion

RESULTS

The 34 case reports we analyzed described a total of 44 patients [Table 1], 24 of which were male. Age ranged from to 3 weeks of age to 82 years old (mean 44, standard deviation ± 25 , median 49). Only 10 were children (<18 years). Among the most common underlying conditions were: Neoplasms ($n = 14$), nephrolithiasis ($n = 9$), gynecological diseases ($n = 2$), car accident ($n = 2$), renal failure ($n = 4$), and with one each, vesicoureteral reflux and renal dysplasia (VURD) syndrome (posterior urethral valves, unilateral VURD), benign prostatic hypertrophy, hydronephrosis, urinary tract infection, Infundibular stenosis, donor renal transplantation, renal colic, pelvic pain and not available ($n = 5$).

Urinothorax was associated with surgical procedures in most patients: Nephrostomy ($n = 15$), urethral obstruction ($n = 10$), transplantation ($n = 4$), lithotripsy ($n = 4$), nephrectomy ($n = 2$), prostatectomy, ureteroscopy, combined hysterectomy and salpingo-oophorectomy, trauma (one each), and other surgery ($n = 5$).

DISCUSSION

Urinothorax should be included in the differential diagnosis of pleural effusions in patients of any age subjected to gynecological, urological surgical procedures as well as resections of intraperitoneal tumors.^[19,23,31] Most of the case reports reviewed emphasize urinothorax as an unexpected surprise. Despite being a rare condition, most large adult or pediatric urological departments should expect to see one of these cases.

Respiratory symptoms are evident in some patients but are not always present, and may be wrongly attributed to atelectasis or

Table 1: Clinical characteristics of patients with urinothorax described in the literature (n=44)

Article	Year	Gender	Age (years)	Etiology	Underlying condition
Firedland <i>et al.</i> ^[14]	1971	M	3 weeks old	Obstructions of urinary tract	Hydro ureteronephrosis
Lahiry <i>et al.</i> ^[15]	1978	F	19	Right nephrectomy	Car accident
Leung <i>et al.</i> ^[16]	1981	M	65	Obstruction of the distal portion of the left ureter	Prostatic adenocarcinoma
Baron <i>et al.</i> ^[17]	1981	M	81	Prostatectomy	Benign prostatic hypertrophy
		F	53	Obstructing urinary tract	Left ureteral calculus
		M	NA	After surgical tear of a ureter	NA
		M	NA	Percutaneous nephrostomy	NA
		M	NA	Spontaneous perforation of ureter	NA
Stark <i>et al.</i> ^[18]	1982	M	68	Cystectomy	Bladder carcinoma
		M	71	Percutaneous nephrostomy	Renal failure
		M	81	Obstructive uropathy	Bladder carcinoma
Carcillo <i>et al.</i> ^[19]	1985	F	12	Renaltransplantation	Renal failure
Ralston <i>et al.</i> ^[20]	1986	F	63	Extravasation into the left anterior pararenal space	Colon carcinoma
Salcedo <i>et al.</i> ^[21]	1986	F	11	Percutaneous kidney biopsy	Undifferentiated neurectodermal sarcoma
		F	13	Percutaneous nephrostomy	Pelvic mass compressing the bladder and ureter
		F	12	Renal transplant	Renal failure
		F	17	Renal transplant	Renal disease secondary to lupus
Miller <i>et al.</i> ^[22]	1988	M	66	Bilateral nephrostomy	Colon cancer
Ansari and Idell ^[3]	1998	F	15	Ureteral perforation	Donor renal transplantation
Hase <i>et al.</i> ^[23]	1999	M	38	Cystic lesion in the left medial pararenal space	Motorcycle accident
Oğuzülgen <i>et al.</i> ^[24]	2002	F	50	Lithotripsy	Nephrolithiasis
Hendriks <i>et al.</i> ^[25]	2002	M	68	Percutaneous nephrostomy	Nephrolithiasis
Ray <i>et al.</i> ^[26]	2003	F	53	Percutaneous nephrostomy	Bladder cancer
Garcia-Pachon and Padilla-Navas ^[27]	2004	M	64	Lithotripsy	Renal stone
Buyukcelik <i>et al.</i> ^[28]	2005	M	8	Obstructions of urinary tract	Urinary tract infection
Lee <i>et al.</i> ^[29]	2005	M	3 weeks old	Obstructive uropathy	Síndrome vurd (posterior urethral valves, unilateral vesicoureteral reflux and renal dysplasia)
Tortora <i>et al.</i> ^[30]	2005	F	38	Obstructive uropathy	Left kidney was markedly hydronephrotic and a large calculus
Karkoulis <i>et al.</i> ^[31]	2007	M	47	Dilated right ureteropelvic junction	Non-hodgkin lymphoma
Bhattacharya <i>et al.</i> ^[32]	2007	M	35	Percutaneous nephrolithotomy	NA
Deel <i>et al.</i> ^[33]	2007	M	64	Percutaneous nephrostomy	Bladder tumor
Handa <i>et al.</i> ^[34]	2007	M	35	Percutaneous nephrolithotomy	Renal calculi
Agranovich <i>et al.</i> ^[11]	2008	M	73	Percutaneous nephrolithotomy	Nephrolithiasis
Jelic and Sampogna ^[10]	2009	M	50	Percutaneous nephrolithotomy	Infundibular stenosis
Amro <i>et al.</i> ^[35]	2009	F	NA	Total abdominal hysterectomy	Premenopausal and pelvic pain
Wei <i>et al.</i> ^[36]	2009	M	50	Percutaneous nephrolithotomy	Renal calculi
Salinas <i>et al.</i> ^[37]	2009	M	78	Percutaneous nephrolithotomy	Bladder cancer
Mora <i>et al.</i> ^[12]	2010	F	82	Laparoscopic radical nephroureterectomy	Urothelial carcinoma
Brown <i>et al.</i> ^[11]	2011	F	52	Percutaneous nephrostomy	Nephrolithiasis
Antoon <i>et al.</i> ^[7]	2012	F	11	Percutaneous lithotripsy	NA
Laskaridis <i>et al.</i> ^[19]	2012	F	45	Right ureteroscopy	Renal colic
		F	32	Ureteroscopic lithotripsy	Calculus
Ameer <i>et al.</i> ^[2]	2013	F	30	Laparoscopy	Endometriosis
Dimitriadis <i>et al.</i> ^[8]	2013	F	42	Left ureteral injury	Ovarian tumour resection
Batura <i>et al.</i> ^[38]	2014	M	64	Percutaneous nephrolithotomy	Metastatic prostate adenocarcinoma

pneumonia.^[8] Biochemical testing is useful, and the ratio of creatinine and biochemical analysis of pleural fluid confirms the diagnosis.^[5,19] Diagnostic imaging, renal scan, X-ray, contrast-enhanced computed tomography or pyelogram are essential to establish the anatomical location of the defect causing urine leakage.^[24,36]

In our case, contrast means were not indicated, given that the patient had renal failure, but cystoscopy plus pyelogram in the operating room showed the atomic location of the fistula.

Initial management of urinothorax is with toracocentesis or thoracostomy^[37] but detecting the origin of the leakage and performing surgical repair is essential. As a conclusion, urinothorax is a rare complication, but should be held in mind, since an early diagnosis will improve the patient's outcome.

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