

PEDIATRIC URINOTHORAX PRESENTING AS REFRACTORY PNEUMONIA AND EMPYEMA

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Abstract. *Urinothorax, a rare cause of pleural effusion, results from urine accumulation in the pleural cavity due to abnormal communication between the urinary tract and pleural space. It is often associated with urinary tract obstruction, trauma, surgery, or rupture of the renal collecting system. Early diagnosis is challenging because of nonspecific clinical features. We report the case of a 3-year-old male with polymalformative syndrome, right-sided pneumonia, and a history of congenital hydronephrosis, who presented with respiratory distress and decreased breath sounds on the right side of the lungs. Chest X-ray and ultrasound revealed a large right-sided pleural effusion. Thoracentesis yielded turbid fluid, with biochemical analysis showing a high concentration of polymorphonuclear leukocytes and elevated lactate dehydrogenase, consistent with inflammation. Despite this, the large volume of drained fluid exhibited laboratory findings consistent with a transudate. Further imaging demonstrated worsening hydronephrosis with a dilated renal pelvis and right megaureter. A nephrostomy tube was placed, resulting in rapid clinical improvement and resolution of the pleural effusion. This case underscores the importance of considering urinothorax in the differential diagnosis of pleural effusion in children with urinary tract anomalies. Prompt pleural fluid analysis and timely urologic intervention can lead to favorable outcomes.*

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INTRODUCTION

Urinothorax is an uncommon clinical entity characterized by the presence of urine in the pleural space, leading to pleural effusion. It usually arises secondary to urinary tract obstruction, trauma, or iatrogenic injury, causing abnormal communication between the urinary and pleural cavities. First described in 1968, urinothorax remains a largely under-recognized clinical entity, with fewer than 100

cases reported in the literature to date [1]. The pathophysiology involves the leakage of urine from the retroperitoneal or peritoneal space into the pleural cavity through diaphragmatic defects or lymphatic channels [2]. Congenital anomalies of the urinary tract are an unusual but plausible cause of this condition in children. The clinical presentation is often non-specific, including respiratory distress, chest discomfort, or signs of pleural effusion, which may delay diagnosis if not considered in the differential [2, 3].

Herein, we present the case of a child with underlying polymalformative syndrome – anorectal agenesis (imperforate anus) with rectourethral fistula and congenital hydronephrosis. The child is presenting with a right-sided pleural effusion due to pneumonia, complicated by urinothorax, highlighting the diagnostic challenges, management, and the importance of considering this rare entity in children presenting with unexplained persistent pleural effusion.

CLINICAL CASE DESCRIPTION

A 3-year-old boy with a complex medical history, including congenital anorectal malformation and right-sided congenital hydronephrosis, was admitted with acute respiratory distress, fever, and right-sided pleural effusion. The child was born from the ninth pregnancy via cesarean section, with a birth weight of 3000 g.

One week prior to admission, he developed fever and cough, treated symptomatically at home without improvement. On the third day, worsening respiratory distress led to hospital admission with imaging showing right-sided pneumonia and pleural effusion. He was initially managed conservatively with broad-spectrum antibiotics and supportive care. Despite therapy, the child's condition deteriorated with increasing pleural effusion, prompting transfer to a specialized pediatric surgery center. A diagnostic thoracentesis revealed a sterile, turbid pleural fluid. Biochemical analysis of the pleural fluid shows high concentration of polymorphonuclear leukocytes and elevated LDH, consistent with inflammation.

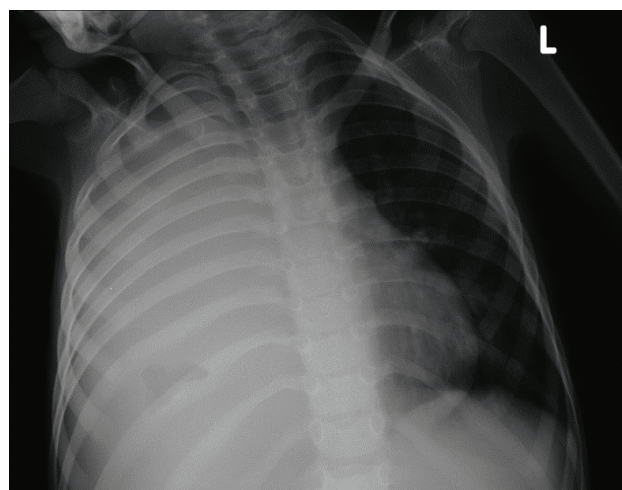


Fig. 1. Initial pulmonary assessment upon admission

During clinical observation, a notable finding was the large volume of fluid drained from the thoracic drain, exceeding 1000 mL per day, with laboratory characteristics consistent with a transudate. Biochemical analysis showed elevated creatinine levels (416 $\mu\text{mol/L}$) with a pleural-to-serum creatinine ratio > 5 , consistent with urinothorax. A renal ultrasound scan confirmed severe right-sided hydronephrosis with parenchymal thinning. An additional CT urography was performed and it revealed an extravasation of contrast material into the retroperitoneal space around the right kidney. Pulmonary CT confirmed an encapsulated pleural effusion in the right pleural cavity as well as extravasation of contrast material into the retroperitoneal space around the right kidney.

A nephrostomy was placed, resulting in immediate clinical improvement and gradual resolution of the pleural

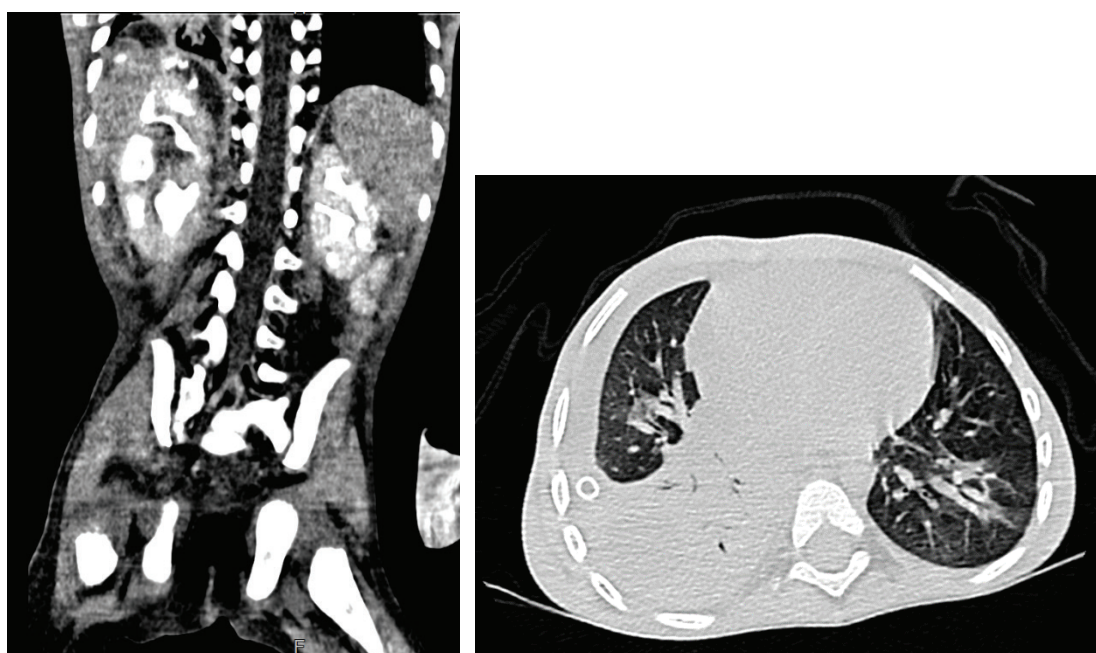


Fig. 2. CT urography and pulmonary CT on the sixth postoperative day

effusion. A video-assisted thoracoscopy was performed for decortication and liberation of the right lung due to the encapsulated pleural effusion. The child's clinical condition improved significantly following the placement of the percutaneous nephrostomy, with rapid resolution of respiratory symptoms and gradual decrease of the pleural effusion. Serial chest radiographs confirmed complete resolution of the effusion within two weeks.

The patient remained under close multidisciplinary follow-up involving pediatric urology, nephrology, pediatric surgery, and genetics teams. Plans were made for definitive surgical correction of the ureteropelvic junction obstruction.

Genetic counseling was offered to the family. The child continued to be monitored for potential immunodeficiency-related complications and hemolytic crises. Further evaluation, including molecular genetic testing, revealed a homozygous variant in the DOCK8 gene of uncertain clinical significance and a hemizygous likely pathogenic variant in the G6PD gene, indicating susceptibility to hemolytic anemia and immune dysfunction. These findings helped explain the patient's recurrent infections and episodes of anemia.

After stabilization, the child received a cutaneous ureterostomy as definitive urinary diversion. At the latest follow-up visit, the patient was in good clinical condition, with stable renal function and no recurrence of pleural effusion. Further reconstructive surgery for the anorectal malformation was scheduled.

DISCUSSION

Urinothorax is an exceedingly rare cause of pleural effusion, most commonly associated with obstructive uropathy, trauma, or post-surgical complications. Since its first description in 1968 by Corriere et al., fewer than 100 cases have been reported in the literature, with pediatric presentations being exceptionally rare [1, 2].

The case presented here highlights the diagnostic difficulty in identifying urinothorax, particularly when the initial presentation mimics more common conditions such as pneumonia with parapneumonic effusion. The child's admission with fever, cough, and pleural effusion naturally led to a working diagnosis of infectious pneumonia. Despite adequate antibiotic therapy and supportive care, the child's condition did not improve, leading to reconsideration of the diagnosis. The turning point came with the observation of massive pleural drainage of clear fluid and the biochemical finding of an elevated pleural-to-serum creatinine ratio, which raised suspicion of urinothorax. Imaging with CT urography revealed a spontaneous rupture of the upper calyx of the hydronephrotic right kidney with extravasation of urine into the retroperito-

neum and pleural cavity – a highly unusual scenario in a child with congenital uropathy [3, 8, 9]. A nephrostomy was placed, resulting in clinical improvement and resolution of the pleural effusion [4-6].

CONCLUSIONS

This case underscores how pre-existing urinary tract malformations can complicate the clinical picture and delay the diagnosis of urinothorax, especially when initial signs are dominated by respiratory symptoms. In children, where pneumonia is far more common than urinothorax, such diagnostic misdirection is understandable. The management of this patient required not only thoracic drainage but also nephrostomy to divert urine and allow for the resolution of the effusion. Definitive management was achieved through cutaneous ureterostomy and urological follow-up. Clinicians should remain alert to the possibility of urinothorax, especially in patients with congenital urinary tract anomalies, when faced with non-resolving or atypical pleural effusions. Early recognition and targeted management are essential to prevent unnecessary delays and to improve clinical outcomes.

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Ethical statement: *This study has been performed in accordance with the ethical standards as laid down in the Declaration of Helsinki.*

Consent for publication: *Consent form for publication was signed by the parent/guardian and collected.*

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