

Nephrolithiasis as a cause of hemoptysis

Bohdan Baralo, MD^a , Tetiana Voznyuk, MD^b , Bohdan Mitiuk, MD^b , Yuriy Poteha, MD^c , Raisa Baralo, MD^d , Volodymyr Kryvetskyi, MD^b , Michael Korman, MD^a , and Ihor Baralo, MD^b

^aDepartment of Internal Medicine, Mercy Catholic Medical Center, Darby, Pennsylvania; ^bDepartment of Surgery and Urology, National Pirogov Memorial Medical University, Vinnytsia, Ukraine; ^cDepartment of Urology, Vinnytsia Regional Clinical Hospital n.a. Pirogov, Vinnytsia, Ukraine, ^dDepartment of Clinical Pharmacy and Pharmacology, National Pirogov Memorial Medical University, Vinnytsia, Vinnytsia, Ukraine

ABSTRACT

We report a case of nephrobronchial fistula complicated by the development of a broncholith within the lung, which led to hemoptysis and blood loss anemia. A 71-year-old man with a medical history of untreated urinary stones was admitted for flank pain, hemoptysis, blood loss anemia, and exacerbation of chronic pyelonephritis. Computed tomography showed staghorn calculi, terminal hydronephrosis, xanthogranulomatous pyelonephritis of the left kidney, nephrobronchial fistula, and large intraparenchymal pulmonary calcification. Surgical treatment was performed in two steps: nephrectomy and then left lower lobectomy. Pathological findings were suggestive of chronic inflammatory changes.

KEYWORDS Blood loss anemia; broncholithiasis; bronchorenal fistula; hemoptysis; xanthogranulomatous pyelonephritis

he first case of nephrobronchial fistula was reported by Bowditch in 1870, and 98 cases have been reported to date. Here we present a rare case of long-standing and untreated urinary stone disease leading to nephrobronchial fistula and broncholith stones leading to hemoptysis.

CASE DESCRIPTION

A 71-year-old man was admitted with hemoptysis and dull left flank pain. He reported a chronic productive cough and exertional dyspnea for about 2 years, which became more intense and accompanied by daily hemoptysis for 2 months (1 tablespoon a day). No wheezing, night sweats, or weight loss was observed. The patient had a history of nephrolithiasis for several decades. He had undergone left-sided pyelolithotomy 40 years earlier for a left kidney stone and had a nephrostomy tube placed approximately a year after surgery, which was eventually removed. Subsequently, he had intermittent moderate dull pain in the left abdominal and flank regions. His medical history included coronary artery disease and heart failure with a preserved ejection fraction. The patient had no history of tuberculosis, asthma, or chronic obstructive pulmonary disease. He was a nonsmoker

and did not take any prescription or over-the-counter medications.

On physical examination, his temperature was 37.2°C; blood pressure, 100/60 mm Hg; pulse, 80 mm Hg; and respiration unlabored. He had a well-healed scar in the left lumbar region without any signs of drainage or visible fistulous tract. Tenderness was present on the left side. Abdominal examination revealed an approximately 15 cm tender mass in the left upper quadrant. The lungs were clear on auscultation with decreased breath sounds over the lower lung fields on the left.

Laboratory data revealed a white blood cell count of 13×10^9 /L, with 87% neutrophils, hemoglobin 7.1 g/dL, and a platelet count of 170,000; blood urea nitrogen level of 28 mg/dL; and creatinine level of 1.1 mg/dL. Urinalysis showed 10 to 12 white blood cells/mm³ and 8 to 10 red blood cells/mm³. Computed tomography (CT) of the chest, abdomen, and pelvis revealed fragmented left-sided staghorn calculi that filled the pelvis and all calices with signs of chronic pyelonephritis and perinephritis as well as rare air inclusions (*Figure 1a*); bilateral pleural effusion (more on the left), patchy infiltrates in the left lower lobe with small areas of loculated fluid and a 22 mm calcified density (*Figure 1b*); and a suspected bronchorenal fistula (*Figure 1c*). The patient

Corresponding author: Bohdan Baralo, MD, Department of Internal Medicine, Mercy Catholic Medical Center, 1500 Lansdowne Ave., Darby, PA 19023 (e-mail: bohdanbaralo@gmail.com)

The authors report no funding or conflicts of interest. The patient provided consent for the case to be published. Received October 2, 2022; Revised October 26, 2022; Accepted November 8, 2022.

228 Volume 36, Number 2

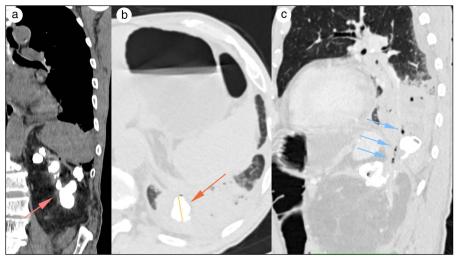


Figure 1. Noncontrast CT of the chest, abdomen, and pelvis showing (a) fragmented left-sided staghorn calculi; (b) calcification of 22 mm within the left lower lobe; and (c) bronchorenal fistula.



Figure 2. Postoperative section of the left lower lobe after resection. The blade of the scalpel points to the stone (22 mm).

was admitted with a diagnosis of staghorn calculi, acute on chronic pyelonephritis, and suspicion of nephrobronchial fistula complicated by a broncholith. He was administered ceftriaxone and tranexamic acid and received multiple transfusions of packed red blood cells for iron-deficiency anemia. He underwent a left open nephrectomy with pathology showing calculous hydronephrosis and chronic pyelonephritis.

The patient was discharged home after nephrectomy but continued to have hemoptysis and was readmitted 4 weeks later for left lower lobectomy. During the surgery, a fistula passing through the lumbocostal triangle was noted. The broncholith corresponding to that discovered on the CT scan was found within the lung parenchyma and resembled urinary stones (Figure 2). The pathology report showed chronic nonspecific purulent inflammation with foreign body granulomas. The postoperative course was complicated

by local bleeding, which eventually ceased without the need for surgical intervention. The patient was later discharged and his further recovery was uncomplicated.

DISCUSSION

Broncholithiasis is defined as the presence of calcified or ossified material within the tracheobronchial tree. Among the possible causes of bronchiolitis are calcified lymph nodes protruding in the tracheobronchial tree (usually caused by an infection such as tuberculosis, histoplasmosis, cryptococcosis, coccidioidomycosis, aspergillosis, nocardiosis, or actinomycosis), calcified endobronchial tumors, ciliary dyskinesia, and migration of intrapleural and intraperitoneal calcifications, such as nephrolithiasis caused by nephrobronchial fistula, erosion caused by bronchial cartilage, and aspiration of foreign bodies.^{3,4} The presence of the fistula observed on CT scan and intraoperatively makes it highly likely that these broncholiths were secondary to nephrolithiasis.

Nephrobronchial fistulas develop secondary to a chronic infectious process in the kidney such as xanthogranulomatous pyelonephritis, complicated pyonephrosis, or rarely tuberculosis.² The incidence of nephrobronchial fistulas has decreased in recent years due to the widespread use of antibiotics and advancements in urinary stone diagnostics and management. The proposed mechanism of fistula formation is the spread of the infection through the fusion of the fascial planes and later through the lumbocostal trigone, which is a weak place in the diaphragm.^{5,6} The symptoms experienced by patients are related to the underlying infectious process, the presence of complicating factors such as urinary stones, and symptoms related to fistula formation. The most common symptoms are fever, unilateral flank pain, night sweats, weight loss, chest pain, cough, foul-smelling sputum, and uroptisis.^{2,7} Definitive surgical treatment of nephrobronchial fistula involves nephrectomy and appropriate drainage of the perinephric abscess if present. Additional therapy (lobectomy

with excision of the fistulous tract) may be required if the infectious process above the diaphragm (e.g., lung abscess) fails to clear after nephrectomy and adequate antibiotic treatment.^{2,8,9}

In conclusion, nephrobronchial fistula is a rare entity with <100 cases described in the literature. In our patient, it was complicated by the formation of a stone within the lung parenchyma, which is an extremely rare but known complication of nephrobronchial fistulas. The presence of stones may lead to mechanical damage and significant hemoptysis, causing blood loss, anemia, and transfusion requirements. A multidisciplinary treatment approach is required for optimal surgical and medical management of this rare cause of hemoptysis.

ORCID

Bohdan Baralo (b) http://orcid.org/0000-0002-6185-3598
Tetiana Voznyuk (b) http://orcid.org/0000-0002-2562-0180
Bohdan Mitiuk (b) http://orcid.org/0000-0002-6115-1932
Yuriy Poteha (b) http://orcid.org/0000-0002-9061-1694
Raisa Baralo (b) http://orcid.org/0000-0002-7342-7860
Volodymyr Kryvetskyi (b) http://orcid.org/0000-0003-1284-6540
Michael Korman (c) http://orcid.org/0000-0003-4526-9639
Ihor Baralo (d) http://orcid.org/0000-0003-3875-904X

- Abeshouse BS. Renal and ureteral fistula of the visceral and cutaneous types; a report of four cases. *Urol Cutaneous Rev.* 1949;53(11):641–674.
- Tamburrini S, Fiorini V, Lugarà M, et al. Nephrobronchial fistula a case report and review of the literature. *Radiol Case Rep.* 2021;16(11): 3470–3477. doi:10.1016/j.radcr.2021.08.025.
- Alshabani K, Ghosh S, Arrossi AV, Mehta AC. Broncholithiasis: a review. Chest. 2019;156(3):445–455. doi:10.1016/j.chest.2019.05.012.
- Seo JB, Song KS, Lee JS, et al. Broncholithiasis: review of the causes with radiologic-pathologic correlation. *Radiographics*. 2002;22(suppl_1): S199–S213. doi:10.1148/radiographics.22.suppl_1.g02oc07s199.
- Qazi HA, Manikandan R, Holmes ME, Philip J, Parsons KF. Nephrobronchial fistula—a case report. *Int Urol Nephrol.* 2007;39(1): 31–32. doi:10.1007/s11255-006-6660-9.
- Evans JA, Meyers MA, Bosniak MA. Acute renal and perirenal infections. Semin Roentgenol. 1971;6(3):276–291. doi:10.1016/0037-198x(71)90039-3.
- Kadaria D, Sarva ST, Deaton PR. Coughing up urine: urinoptysis?
 Am J Respir Crit Care Med. 2013;187(9):1031–1032. doi:10.1164/rccm.201301-0067LE.
- Borum ML. An unusual case of nephrobronchial and nephrocolonic fistula complicating xanthogranulomatous pyelonephritis. *Urology*. 1997;50(3):443. doi:10.1016/S0090-4295(97)00296-3
- Dubey IB, Singh AK, Prasad D, Jain BK. Nephrobronchial fistula complicating neglected nephrolithiasis and xanthogranulomatous pyelonephritis. Saudi J Kidney Dis Transpl. 2011;22(3):549–551.