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Case Report

Management of spontaneous subscapular renal hemorrhage: A multidisciplinary and hybrid approach to Wunderlich syndrome ☆,☆☆

Hamza El Abidi[a,*], *Ahmed Ibrahimi*[a], *Adam El Aboudi*[a], *Mohamed Ali Mikou*[a], *Imad Boualaoui*[a], *Zineb Labbi*[b], *Omar El Aoufir*[b], *Meryem Fikri*[b], *Hashem El Sayegh*[a], *Yassine Nouini*[a]

[a] *Urology A Department, University Hospital IBN SINA, Rabat, Morocco*
[b] *Radiology Department, University Hospital IBN SINA, Rabat, Morocco*

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ABSTRACT

Wunderlich syndrome (WS), or spontaneous renal hemorrhage, is a rare and serious condition that demands prompt diagnosis and management. This report describes a 26-year-old female patient who experienced severe right-sided flank pain and hypovolemic symptoms during hemodialysis. The patient had several comorbidities, including poorly managed diabetes mellitus, diabetic nephropathy, and arterial hypertension. An active bleeding leading to a subcapsular renal hematoma was discovered by imaging. Using interventional radiology, immediate renal arterial embolization was part of the initial therapy. Recurrent bleeding required an emergency hemostatic nephrectomy despite temporary stabilization. This example emphasizes the importance of a hybrid management strategy that combines interventional radiology and surgical competence. It emphasizes how crucial a multidisciplinary team is to customizing interventions that strike a balance between the patient's underlying chronic comorbidities and the urgent requirements of a life-threatening illness. This all-encompassing strategy produced a favorable result.

Introduction

Wunderlich syndrome (WS), characterized by spontaneous renal bleeding without trauma, is a rare and possibly fatal illness that necessitates immediate diagnosis and treatment. It was first described over 3 centuries ago and continues to be a diagnostic challenge due to its wide range of appearances.

The most common underlying causes include renal tumors, particularly angiomyolipomas and renal cell carcinoma,

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* Corresponding author.
E-mail address: hamza.elabidi@yahoo.com (H. El Abidi).
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as well as vascular diseases like polyarteritis nodosa or arteriovenous malformations. This report examines the rare case of chronic hemodialysis as a probable cause of spontaneous subcapsular kidney hemorrhage in a 26-year-old patient with poorly managed diabetes. The case demonstrates the difficult balance of dealing with acute emergencies and chronic comorbidities through timely imaging, interventional radiology, and surgical therapy. The multidisciplinary approach emphasizes the need to combine new diagnostic technologies and therapy options to improve outcomes for this rare but crucial ailment.

Case report

A 26-year-old female patient with a medical history of diabetes mellitus treated with insulin was diagnosed during childhood following the onset of asthenia, weight loss, thirst, and constant polyuria. The patient has complications such as diabetic nephropathy and diabetic retinopathy, mainly due to poor treatment adherence. The patient's condition progressed to end-stage renal disease, for which she needed hemodialysis 3 times per week. Additionally, she has been diagnosed with hypothyroidism treated with levothyroxine, and arterial hypertension.

The patient has no surgical history, and she has a mother treated for type 1 diabetes. During a hemodialysis session, the patient suddenly developed acute right-sided flank pain. The pain was severe and unresponsive to analgesics, necessitating an immediate referral to the emergency department.

She received a rapid initial clinical examination. The patient was afebrile and conscious, with a pale mucocutaneous complexion. Stable hemodynamically with a heart rate of 120 bpm, blood pressure at 90/60 mmHg, no peripheral hypoperfusion signs, and preserved diuresis. She was eupneic with an SpO_2 of 98% on ambient air and no respiratory distress. Right flank tenderness was noted without palpable masses.

The laboratory workup showed hemoglobin at 6.5 g/dL, sodium at 132 mmol/L, potassium at 4.5 mmol/L, urea at 1.46 g/L, and creatinine at 79.5 mg/L. The coagulopathy profile showed a platelet count of 325,000/μL, a prothrombin ratio of 74%, and a partial thromboplastin time of 1.11. As for arterial blood gas: pH: 7.38, $PaCO_2$: 32 mmHg, PaO_2: 103 mmHg, bicarbonate: 18.9 mmol/L, base excess (BE): −2 mmol/L. The urine cytobacteriological analysis was negative.

Following an early abdominal ultrasound that revealed a subcapsular right renal hyperechogenic mass, the patient underwent an emergency contrast-enhanced CT urogram, which revealed a nonruptured, nontumoral, hypodense right renal subcapsular collection that did not enhance after injection, measuring 100 × 30 mm, with no signs of tissue necrosis suggesting a subcapsular renal hematoma. An infiltration of the right psoas muscle was also observed (Fig. 1).

The management plan includes the immediate installation of 2 peripheral venous lines, fluid resuscitation, serial arterial blood gas analysis, insulin delivery based on capillary glucose levels, and blood transfusions.

Following the diagnosis of the subcapsular renal hematoma with active bleeding, and given the relatively hemodynamic stability, a multidisciplinary decision was made for urgent management by the interventional radiology team (Fig. 2).

After informed and written consent by the patient and explaining the procedure and potential risks. Using the Seldinger technique and under local anesthesia at the access site, a guide wire is inserted into the femoral artery under fluoroscopic guidance, and then a vascular sheath (5F) is placed in the common femoral artery to facilitate catheter insertion.

A curved angiographic catheter is advanced, and selective renal angiography is then performed to identify the target vessel; extravasation of the contrast agent was spotted in the lower sector. A coil was used to achieve successful embolization.

Postembolization angiography ensures complete occlusion of the targeted vessels and identifies any accessory arteries that may require additional embolization (Fig. 2).

The puncture site is compressed with a sterile dressing, and the patient is placed on bed rest for 24 h. Ongoing care included monitoring renal function, electrolytes, and hemoglobin levels during hospitalization.

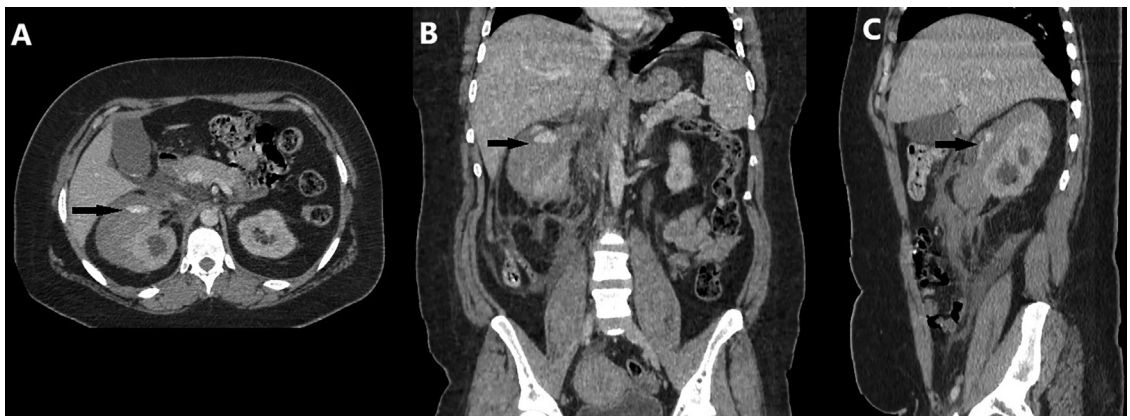

Fig. 1 – Axial (A), coronal (B), and sagittal (C) contrast-enhanced CT scan images showing subcapsular hematoma of the right kidney with active extravasation of contrast agent. The black arrow demonstrates a focal high-density spot within the hematoma, indicating active bleeding.

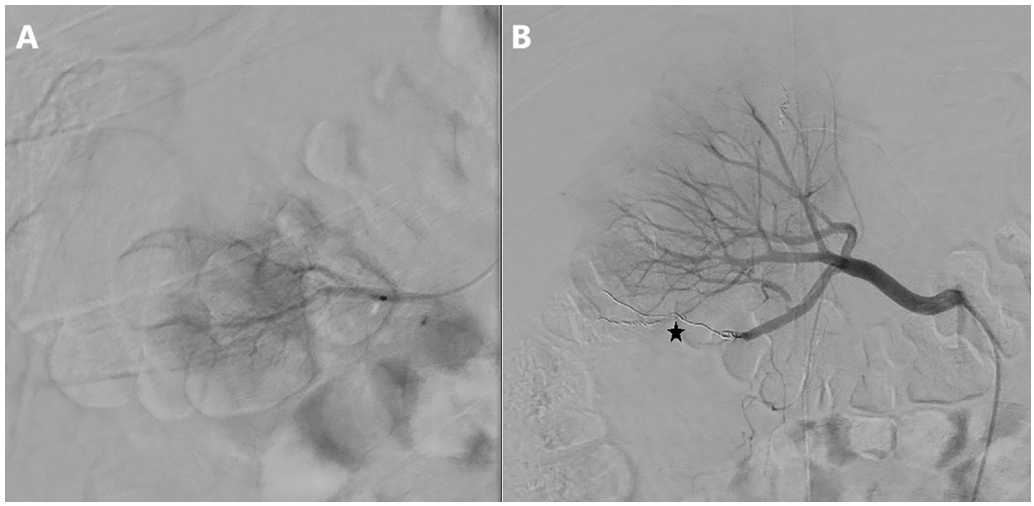

Fig. 2 – Selective right renal artery angiography showing contrast extravasation on the inferior territories (panel A black arrow). Panel (B) shows the postembolization angiogram with the coil (black asterisk).

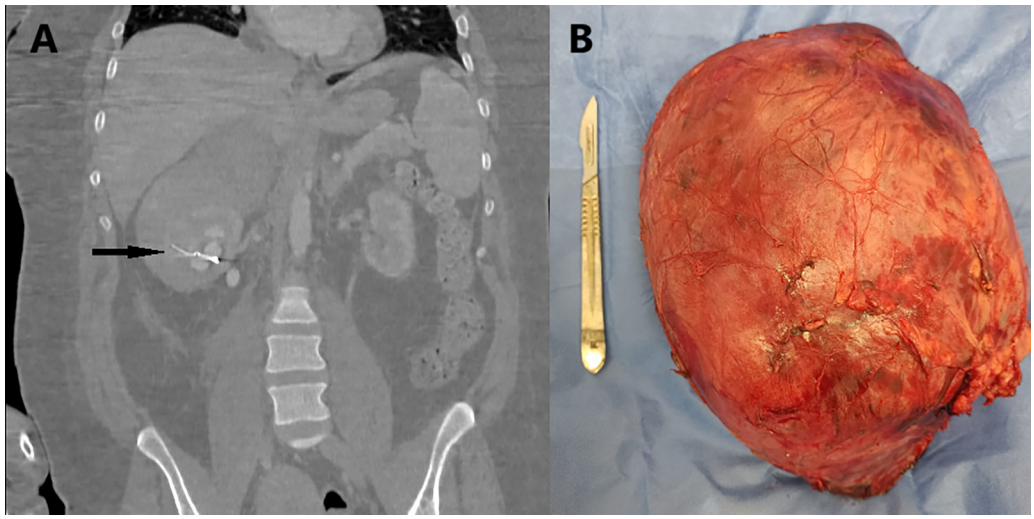

Fig. 3 – (A) Postembolization axial contrast-enhanced CT scan images showing the coil and persistent extravasation of the contrast agent (black arrow). (B) Surgical specimen from a hemostatic radical nephrectomy.

The patient had tachycardia and slight hypotension on the second day following the embolization. In light of the clinical situation, an urgent Uro-CT scan was done, which indicated a persistent hematoma and aggressive bleeding in the arterial phase.

The patient was already on chronic hemodialysis; thus, the vital prognosis took precedence over the functional prognosis, and 1 kidney would suffice for effective diuresis. An emergency hemostatic nephrectomy was then performed. The patient was brought to the surgery room after being quickly resuscitated and hemodynamically stable. A subcostal incision was used to improve visibility of the renal pedicle.

After moving the colonic angle and opening the retroperitoneum, a significant renal gap was seen. The retroperitoneal hemorrhage was modest, most likely due to blood being retained under the renal capsule and as a result of the prior embolization.

A renal pedicle ligation was performed, followed by nephrectomy and placement of a drainage tube (Redon). The postoperative course was characterized by good improvement, hemodynamic stability, cessation of deglobulization, preserved diuresis, and a drainage tube that only returned a few traces of sero-hematologic fluid. The patient was discharged on the fifth day postoperatively after the drainage tube was removed on day 2 and given an oral antibiotic prescription based on her renal function (Fig. 3).

On the histopathological examination, the renal tissue shows chronic interstitial inflammation, characterized by a dense infiltration of lymphocytes and plasma cells, along with fibrosis and tubular atrophy. Additionally, acute findings include patchy suppurative inflammation with neutrophilic infiltration and areas of tubular necrosis, reflecting the acute phase of pyelonephritis.

Discussion

Wunderlich syndrome (WS) is an acute onset of spontaneous renal hemorrhage into the subcapsular, perirenal, and pararenal spaces without prior trauma. Bonet originally reported the condition in 1700, and Karl August Wunderlich defined it clinically as 'spontaneous renal capsule apoplexy' in 1856. Coenen originally coined the term Wunderlich syndrome in 1910 [1].

A review of the MEDLINE database from 1971 until 2008 showed that WS accounts for 1%-3% of cases in long-term dialysis patients [2].

WS can have various underlying risk factors, most notably neoplasms, which account for 60%-65% of cases. The most common are angiomyolipomas, which majorly increase the risk of rupture considerably when the tumor exceeds 40 mm in diameter, as well as renal cell carcinoma, which contributes significantly to spontaneous renal bleeding. Vascular conditions may contribute to the majoration of rupture and hemorrhage risk. Polyarteritis nodosa is the most prevalent vascular cause of WS. It is an idiopathic systemic vasculitis that causes necrotizing infarction of the small and medium arteries, as well as microaneurysms, occlusions, and strictures. Renal artery aneurysms, arteriovenous malformations, and fistulas, as well as renal vein thrombosis, might cause increased pressure inside the kidney and eventual hemorrhage.

Acute pyelonephritis, emphysematous pyelonephritis, and renal abscesses constitute about 5%-10% of all cases of WS. Such infrequent causes should be investigated when appropriate. Diabetes patients, such as the one in the case study, with renal infections are more likely to develop WS due to infection-associated renal parenchymal necrosis, intravascular thrombosis, and inflammatory erosion of renal vessels, which can lead to renal parenchymal hemorrhage and rupture [3]. Infectious diseases, notably pyelonephritis, are acknowledged as a less common source of WS. Infection-induced kidney inflammation can result in the formation of an abscess and bleeding.

Overall, while neoplasms, followed by coagulation disorders, are the primary risk factors for Wunderlich syndrome, infectious processes like pyelonephritis also contribute to its development via mechanisms like infection-induced inflammation and abscess formation.

More specifically, WS in chronic hemodialysis patients can be triggered by a variety of reasons. Acquired cystic kidney disease (ACKD) is a prevalent cause, which often develops in individuals on long-term dialysis. In ACKD, the development of multiple fragile renal cysts exposes them to rupture, resulting in bleeding. Chronic hemodialysis increases bleeding risks by inducing uremic coagulopathy, which causes platelet dysfunction as a result of uremia-induced metabolic alterations. Finally, chronic inflammation and oxidative stress associated with end-stage renal disease (ESRD) weaken renal tissues and vasculature, making the kidney more susceptible to spontaneous bleeding. These characteristics together provide a high-risk setting for subcapsular hematoma development in this patient population.

Wunderlich syndrome is generally diagnosed based on clinical presentation and advanced imaging techniques. Patients often arrive with acute flank pain, hematuria, and indications of shock, or, less frequently, with the LENK's triad, which includes acute flank pain, flank mass, and hypovolemia. Computed Tomography (CT) is the gold standard for diagnosis, detecting retroperitoneal hemorrhage with about 100% sensitivity and providing insights into the underlying causes. Despite its usefulness, CT has a modest sensitivity for detecting specific etiologies (57%), highlighting the need for further diagnostic techniques in certain circumstances. Active extravasation is defined as contrast material leaking from a vessel into the surrounding tissues. During the arterial phase, CT attenuation values typically range between 175.5 ± 20.7 Hounsfield Units (HU) and 157.8 ± 21.6 HU, indicating a contrast enhancement pattern within hematomas. A jet-like extravasation pattern could indicate fast bleeding from bigger arteries. Focal high-density spots within hematomas indicate active bleeding, which becomes more diffuse with delayed scanning. While dynamic alterations between imaging phases suggest bleeding too.

Ultrasound is used as an initial screening tool; however, it has a substantially lower sensitivity (56% for detecting bleeding and underlying problems). When CT is not accessible, magnetic resonance imaging might be used. MRI is particularly valuable when CT findings are inconclusive or when detailed tissue characterization is required. MRI offers greater soft tissue contrast than CT and can distinguish between subacute and chronic hematomas based on signal features (e.g., hyperintensity on T1-weighted images for subacute blood due to methemoglobin). MRI angiography can detect vascular abnormalities such as aneurysms and arteriovenous malformations. MRI is extremely sensitive for detecting kidney cancers, particularly those that are difficult to visualize on CT. It can identify fat components in angiomyolipomas and vascularized tumors in RCC.

Angiography can be useful, both as a diagnostic and therapeutic tool, in assessing vascular causes of hemorrhage as well as for unresolved cases when cross-sectional imaging fails to identify a definitive cause [3,4].

Conservative management is indicated for hemodynamically stable patients without imaging evidence of active extravasation or large hematomas. This includes monitoring and supportive care, bed rest, and immobilization to prevent further stress and reduce the risk of re-bleeding. Fluid resuscitation with crystalloids is administered to maintain hemodynamic stability. Pain management and hemoglobin monitoring to assess for ongoing deglobulization. Antibiotic prophylaxis is indicated if there is a suspicion of infection [5,6].

Following these conservative measures, hematomas often resolve spontaneously over weeks to months. Serial imaging is recommended to track hematoma regression.

Renal arterial embolization is usually the first-line treatment. It is the preferred strategy for active extravasation, using super-selective catheterization and embolic agents such as coils or polyvinyl alcohol particles to induce rapid hemostasis while maintaining renal function; it has a high success rate and is considered a nephron-sparing approach with short hospital stays. Nephrectomy is necessary when embolization is ineffective or when there is significant hemorrhage. Preoperative embolization can lower morbidity and the likelihood of

intraoperative hemorrhage, reducing then the procedure's difficulty [5–7].

Conclusion

This Wunderlich syndrome example highlights how a planned and interdisciplinary approach to controlling spontaneous renal bleeding. The seamless integration of modern imaging, interventional radiology, and surgical competence resulted in a successful outcome, even in a patient with many comorbidities. This case underlines the need to treat WS as a diagnostic emergency and the need for tailored care regimens to deal with its various manifestations and sequelae.

Patient consent

A written informed consent for the publication of this case was obtained from the patient.

Ethical approval

Ethical approval for this study was provided by an ethical committee.

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