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CASE REPORT

Spontaneous Suburothelial Hemorrhage: The Crucial Role of Radiology in Preventing Unnecessary Interventions

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Abstract: Spontaneous suburothelial hemorrhage (SSH), also known as Antopol Goldman lesion, is a rare condition characterized by spontaneous bleeding into the renal sinus and proximal ureter wall. This case report describes the clinical presentation, imaging findings, and management of SSH in a 20-year-old female initially suspected to have urothelial malignancy. Imaging features of SSH include pre-contrast hyperdensity and non-enhancing thickening of the pelviureteric wall, which can mimic transitional cell carcinoma (TCC) and lead to unnecessary interventions. Radiologists should maintain a high level of suspicion for SSH and be aware of its imaging characteristics to avoid misdiagnosis. Additionally, clinical data, such as bleeding dyscrasia, can aid in the imaging diagnosis. This report provides insights into the diagnosis and management of SSH while offering a comprehensive literature review on its clinical presentation and imaging features. Increased awareness of SSH will facilitate accurate diagnosis and appropriate management, avoiding unnecessary interventions for patients with this benign condition.

Keywords: Antopol-Goldman lesion, hematuria, INR, non-contrast enhanced CT, SSH, spontaneous suburothelial hemorrhage

Introduction

Spontaneous suburothelial hemorrhage (SSH), also referred to as Antopol-Goldman lesion (AGL), is a rare benign condition that can mimic transitional cell carcinoma (TCC), leading to potential unnecessary interventions. Despite its clinical significance, SSH has been sparingly documented in the English literature, with only approximately 50 reported cases, primarily in the form of isolated case reports and small series. This case report presents the first documented case of SSH in Ethiopia, emphasizing the importance of recognizing and understanding this condition within diverse clinical settings.

The misdiagnosis of SSH as TCC can have hazardous implications, including unnecessary invasive procedures and treatments. Therefore, increasing awareness of SSH and its characteristic features is crucial for accurate diagnosis and appropriate patient management. Radiological imaging, complemented by proper clinical data, plays a central role in achieving an accurate diagnosis of SSH. Radiologists bear the responsibility of recognizing and differentiating SSH from malignant entities, enabling optimal patient care.

Case Presentation

We present the case of a 20-year-old female patient who was admitted two weeks after undergoing cesarean section delivery for an indication of macrosomia. She was admitted due to lower extremity deep vein thrombosis (DVT) following a 21-day history of leg swelling. The patient's complete blood count (CBC) profile was within normal limits, and her international

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normalized ratio (INR) was between 1.0 and 2.0. She was started on prophylactic warfarin therapy and subsequently discharged.

Three weeks after her discharge, the patient returned to the hospital with symptoms of vomiting, frank hematuria, and abdominal distension. Physical examination revealed skin ecchymoses, bruises, and a prolonged bleeding time, as evidenced by continuous bleeding from finger tips pricked for a blood sample and intravenous access sites. Warfarin was temporarily discontinued, and her INR was determined to be only mildly elevated, with laboratory determinations ranging between 2.0 and 3.0.

During her hospital stay, an abdominopelvic ultrasound examination was performed, revealing ascites with echodebrinous sediments and floaters, as well as bilateral pelviureteric mass-like thickening. A computed tomography (CT) scan was performed, which showed hemoperitoneum (Figure 1), thickening of the bilateral renal pelvis and proximal ureters (Figure 2), giving the appearance of enhancing urothelial masses on post-contrast images (Figures 3 and 4). It was noted

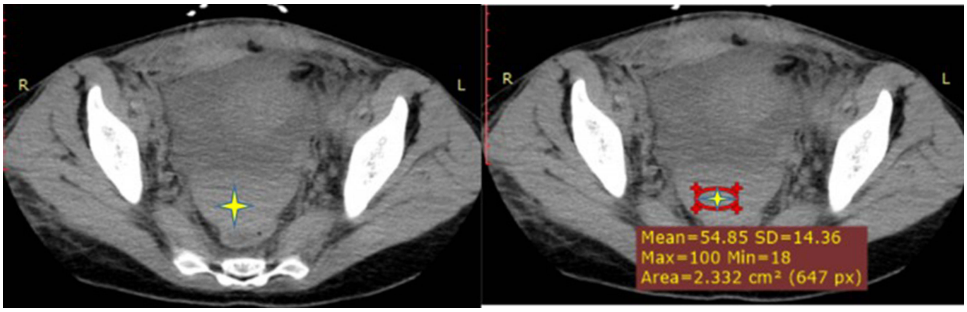

Figure 1 NCECT (left) and CECT (right) showing pelvic peritoneal hematoma (note the density of the hematoma: in the range of 50s and above here indicated is 54.85 HU).

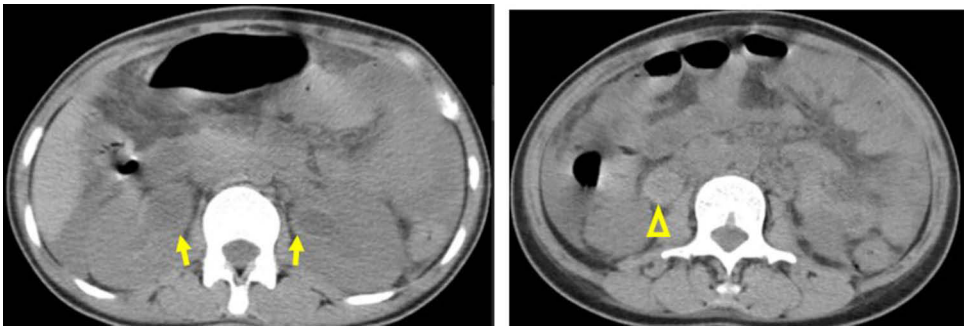

Figure 2 Axial NCECT at the level of the renal pelvis (left) and proximal ureters (right), showing symmetrical smooth, hyperdense thickening of the walls of the bilateral renal pelvis (arrows) and ureters (arrow head) with mild bilateral hydronephrosis.

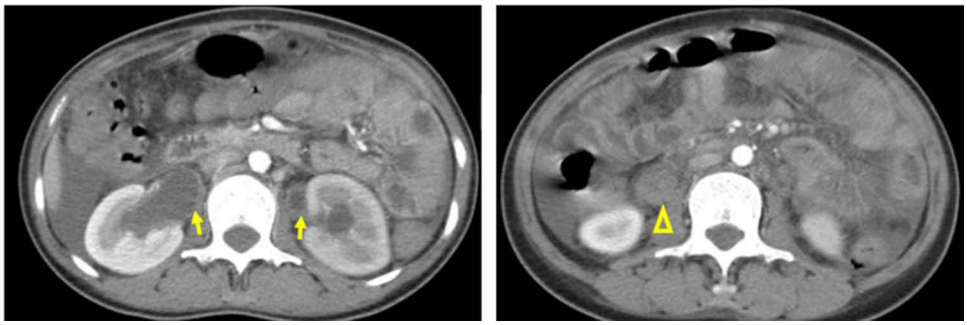

Figure 3 Axial CECT at respective levels (of Figure 1) (left and right); note how a hematoma could be easily missed as an "enhancing" mass lesion (arrows and arrow head) had the CECT only been seen.

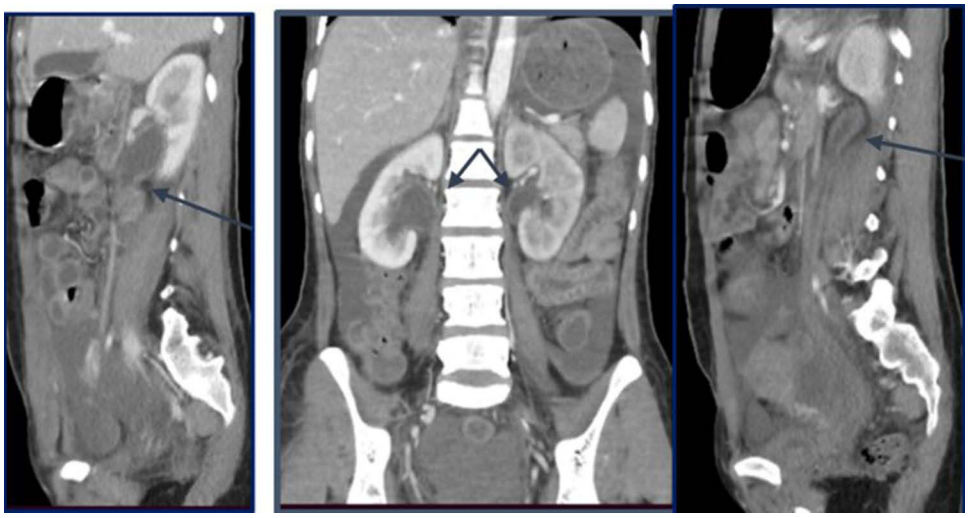

Figure 4 CECT; Sagittal (right and left) and coronal (middle) recon images showing the bilateral renal sinus, PUJ and proximal ureteric wall thickening (long arrows, angled short arrows).

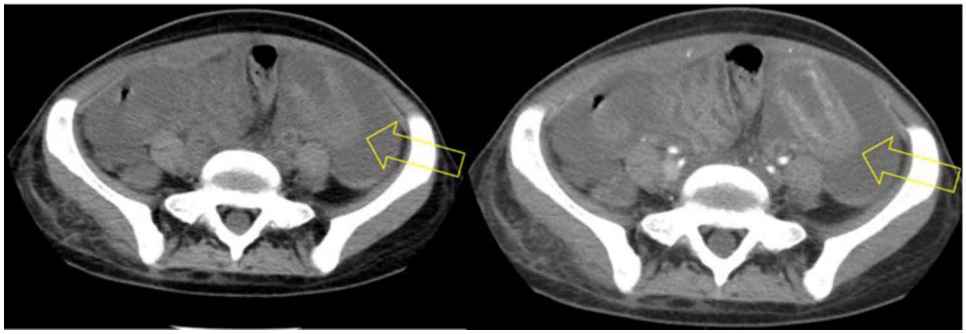

Figure 5 NCECT (left) and CECT (right) at the mid-abdomen region showing a segment of jejunal loop wall thickening with pre-contrast hyperdensity and post-contrast non-enhancement.

that little attention had been given to the non-contrast enhanced study, which revealed areas of spontaneous hyperdensity in the lesions, consistent with blood, along with hyperdense thickening (hemorrhage) of the jejunal wall (Figure 5).

At this point, spontaneous suburothelial hemorrhage was suspected, and conservative management was initiated. Subsequently, the patient was transferred to another referral hospital to receive ongoing intensive care support.

Discussion and Literature Review

Spontaneous suburothelial hemorrhage (SSH), also known as Antopol-Goldman lesion, was first described by Antopol and Goldman in 1948 based on their case series of seven patients.[1] It has been referred to using various terminologies, including suburothelial, sub-epithelial, parietal, or mural hemorrhage/hematoma, with the prefix "spontaneous" used to differentiate it from post-traumatic and iatrogenic cases.[2,3] While the exact etiology remains unclear, the majority of reported cases, including ours, have been attributed to bleeding diathesis caused by excessive anticoagulant use or other coagulation abnormalities.[2,4,5] Oral anticoagulants, especially warfarin, are the most commonly implicated agents, although other anticoagulants can also lead to similar complications.[6,7] A common trend observed in SSH cases is a supra-therapeutic international normalized ratio (INR), with an exponentially increased risk of bleeding when the INR exceeds 5. However, subtherapeutic or normal INR levels can also be associated with SSH.[7]

In general, anticoagulant-related hemorrhages occur in 4% to 24% of patients receiving prophylactic or therapeutic anticoagulation.[8–10] These hemorrhages most commonly involve the abdominal wall and gastrointestinal tract, while renal hematomas are rare. Suburothelial and renal sinus hemorrhages, which are considered the most common types of

renal hematomas associated with anticoagulant therapy, are exceptionally rare. Other forms of renal system hematomas include parenchymal, subcapsular, perinephric, and pararenal hematomas.[7]

The clinical presentation of SSH typically involves macroscopic painful or painless hematuria, with or without flank pain.[11] Although unilateral renal involvement with no left or right predilection is commonly reported, bilateral symmetrical disease, as observed in our case, suggests a systemic disease rather than a malignancy.[4] Although the available case reports are more frequently linked to the elderly population, likely due to increased anticoagulant use for various indications in this age group, SSH can occur at any age, from neonates to young adults, regardless of sex.[12] Spontaneous hemorrhages in other organs, mucosal sites, and petechial bleedings can aid in the diagnosis but are rarely reported. Additionally, associated intestinal mural hematoma, which was noted in our case, is a rare finding.[13] We have not yet found any records of associated hemoperitoneum; however, our case showed hematoma in the bilateral paracolic gutters, inter-loop spaces, and pelvis. The intraperitoneal hemorrhagic effusion seen in our case could be related to altered intramural osmotic gradients resulting from the intramural hematoma.[14] It remains unclear whether this mechanism is responsible for the expansion of subepithelial hematomas in cases of SSH. Rare cases of retroperitoneal hematoma associated with AGL have been reported.[8]

The majority of SSH cases completely resolve within 1–4 weeks of conservative treatment.[8] In cases where conservative management fails to resolve hematuria, selective arterial embolization has been applied.[15] Due to a lack of awareness, underreporting, and limited experience with this condition, many cases have been misdiagnosed as urothelial malignancies, leading to unintended radical nephrectomies. The reported misdiagnosis rate post-nephrectomy is greater than 30%.[16–18]

On CT, suburothelial hematomas present as mural thickening of the renal pelvis and upper ureter. Their pre-contrast hyperdensity, post-contrast non-enhancement, and subsequent spontaneous resolution are key discriminators of this entity from other mimickers, such as urothelial malignancies. Focal pelviureteral wall irregularity on intravenous pyelography (IVP) and mass-like thickening of the renal pelvis on ultrasound have been reported as imaging findings. Non-contrast-enhanced CT (NCECT) followed by contrast-enhanced CT (CECT) is the preferred imaging modality, revealing a hyperdense, non-enhancing mural thickening.[19] The differential diagnosis on IVP includes transitional cell carcinoma, pyeloureteritis cystica, or submucosal edema, while on CT, transitional cell carcinoma or renal cell carcinoma remains the primary consideration.[20] Other mimickers include ureteral wall thickening related to infectious and/or inflammatory ureteritis and reactive thickening from impacted ureteral stone.

Reported complications of SSH include amyloidosis, compressive urinary tract obstruction, rupture into the collecting system with resultant clot formation, and forniceal rupture leading to retroperitoneal hematoma.[19] Although associations with hydronephrosis and urinary obstruction are uncommon, mild hydronephrosis was seen in our case. Recommendations for screening CT and the necessity of follow-up CT have produced mixed conclusions. Some studies suggest a confirmatory follow-up CT within two to four weeks,[4,7,21] while others propose that increased awareness and confident diagnosis could eliminate the need for follow-up imaging.[22,23] Magnetic resonance imaging (MRI) can also show thickening of the urothelial wall, similar to CT, with variable T1 and T2 signal intensity changes reflecting the age of the hematoma, acute and subacute bleeds, and hemosiderin deposition.[24]

Conclusion

Spontaneous suburothelial hemorrhage is a commonly misdiagnosed entity that follows a benign and self-limiting course. Given that the majority of reported cases, including ours, are secondary to the underlying coagulation abnormalities, the term "spontaneous" can be misleading. We recommend adopting a more descriptive terminology, such as "coagulopathy-related suburothelial hemorrhage", to better reflect the underlying etiology.

This report not only underscores the importance of differentiating SSH from TCC to halt inessential interventions but also emphasizes the pivotal role of radiology in diagnosing SSH. Furthermore, it highlights the significance of proper clinical data integration to aid in accurate diagnosis. By reporting the first case of SSH in Ethiopia and providing a comprehensive discussion on this condition, we hope to raise awareness, encourage further research, and enhance clinical practice in the recognition and management of this rare entity. Given the paucity of literature on MRI of SSH and the versatility of MRI in modern clinical practice, we notably advice further study into the characteristics of SSH on MRI. And since prevention is the most crucial strategy in possibly preventable cases like this one, we expect that future research on this entity would concentrate on looking for potential preventative measures.

Institutional Ethical Clearance

Institutional approval is not required at both Addis Ababa University/Tikur Anbessa specialized hospital and Arsi University/Asela Referral and teaching hospital for a case report if proper patient consent is guaranteed.

Declaration of Patient Consent

The patient has been well informed that we are publishing the case for an academic purpose and the patient has given consent that her CT images and important clinical data will be included in the case report and literature review. The patient clearly understands that anonymization of her initials and name is guaranteed and due efforts will be made to conceal her identity.

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Disclosure

The authors report no conflicts of interest in this work.

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