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# Adrenal Hemorrhage as a Presenting Symptom in a Patient with Undiagnosed Cushing syndrome: A Case Report

**Mutlaq Almalki[1], Assel shams[2], Mahmoud Shaikh[3], Moayad alrefaei[3], Musab Alnadwi[3], Renad Abo Alshamat[3]**

1. Consultant breast and endocrine surgery, Alnoor specialize hospital
2. General surgery resident in Alnoor specialize hospital
3. General surgery Senior registrar in Alnoor specialize hospital.

## ABSTRACT

Introduction: Spontaneous adrenal hemorrhage is a rare occurrence, often associated with trauma or underlying adrenal pathology. It poses a diagnostic challenge due to its nonspecific presentation and rarity. Adrenal hemorrhage as an initial presentation of Cushing syndrome is particularly uncommon. [1]

Case Presentation: A 23-year-old Saudi male with no significant medical history, except for prior gastric sleeve surgery, presented with sudden-onset left flank pain. Imaging revealed a left adrenal hemorrhage and further workup identified ACTH-independent Cushing syndrome secondary to adrenal cortical hyperplasia. The patient underwent a successful laparoscopic adrenalectomy, with histopathology confirming adrenal cortical hyperplasia with hemorrhage.

Discussion: Adrenal hemorrhage is rarely the initial manifestation of Cushing syndrome, with most cases typically associated with trauma or coagulopathy. [2] In this case, the patient presented with spontaneous hemorrhage and was later diagnosed with Cushing syndrome, which contributed to the adrenal pathology. Persistent obesity post-gastric sleeve surgery added complexity to the case, highlighting the need for thorough endocrine evaluation.

Conclusion: This case illustrates the diagnostic challenges of spontaneous adrenal hemorrhage and its association with underlying Cushing syndrome. A multidisciplinary approach, including conservative management and elective surgery, ensured successful patient outcomes. Early diagnosis and treatment are critical in preventing life-threatening complications in similar cases.

**KEYWORDS:** Hemorrhage, Cushing syndrome, diagnosis.

## 1. Introduction

Adrenal hemorrhage is a rare but potentially life-threatening condition, often associated with trauma or underlying adrenal pathology. The spontaneous occurrence of adrenal hemorrhage in otherwise healthy individuals is uncommon and usually

poses a diagnostic challenge due to its nonspecific presentation and the variety of underlying causes. [1]

In this report, we present the case of a 23-year-old Saudi male with no prior medical history, except for a past gastric sleeve surgery, who presented to the emergency department with left flank pain. The patient was subsequently diagnosed with a spontaneous adrenal hemorrhage.

Upon further assessment, an underlying ACTH-independent Cushing syndrome due to adrenal cortical hyperplasia was diagnosed. This case illustrates the diagnostic challenge and the importance of a multidisciplinary team in managing his condition.

In this case report, we aim to highlight the unique aspects of this case, including the initial presentation with hemorrhage, the incidental diagnosis of Cushing syndrome, which necessitated thorough evaluations, and the surgical management and outcome.

## 2. Case presentation

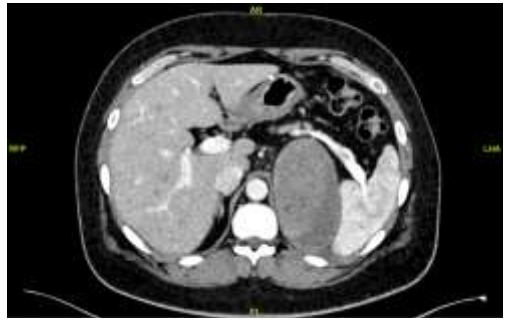
A 23-year-old Saudi male with no known prior medical history and a past surgical history of gastric sleeve surgery, presented to the emergency department at Alnoor Specialist Hospital. He complained of sudden-onset left flank pain, which persisted for one day and was accompanied by nausea and vomiting. He denied any symptoms of hematuria, dysuria, or changes in urinary frequency. Upon physical examination, the patient's abdomen was soft and non-distended, with tenderness noted upon deep palpation in the right flank.

Initial laboratory investigations revealed: hemoglobin 14.8 g/dL, white blood cell count of 11 x 10^9/L, platelets 37 x 10^9/L, glucose 144 mg/dL, INR 0.95, and partial thromboplastin time 28.8 seconds. His electrolyte levels were as follows: potassium 3.6 mEq/L, sodium 142 mEq/L, creatinine 77 µmol/L, and blood urea nitrogen 5.2 mmol/L. Other lab results were within normal limits.

A contrast-enhanced CT scan of the abdomen (fig. 1) revealed a poorly defined heterogeneous mass in the left upper retroperitoneal region with mixed densities and adjacent fat stranding, suggestive of left adrenal hemorrhage. The patient was admitted under the care of the general surgery team for further evaluation and management of left adrenal hemorrhage.

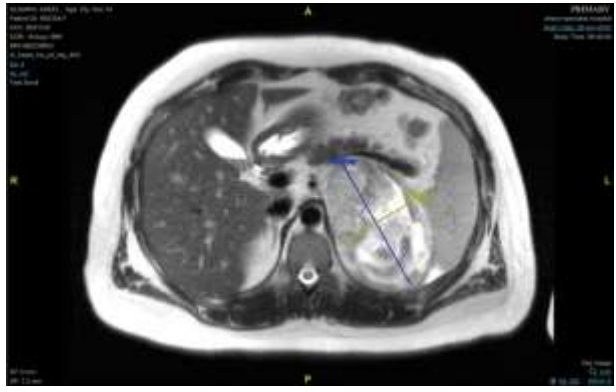
During the hospital stay, the patient was managed conservatively with regular monitoring of vital signs, serial abdominal exams, imaging studies, and consultations with hematology and endocrinology. Further imaging, including a contrast-enhanced CT (adrenal protocol) scan of the abdomen, confirmed a heterogeneous lesion in the left adrenal gland with free fluid, consistent with adrenal hemorrhage but without active bleeding. (fig. 2)

An MRI revealed a large retroperitoneal hematoma with a solid enhancing mass in the left adrenal region, suggestive of a possible pheochromocytoma. (fig. 3) A benign-appearing lesion in the spleen was also noted, possibly representing a hemangioma or hamartoma.

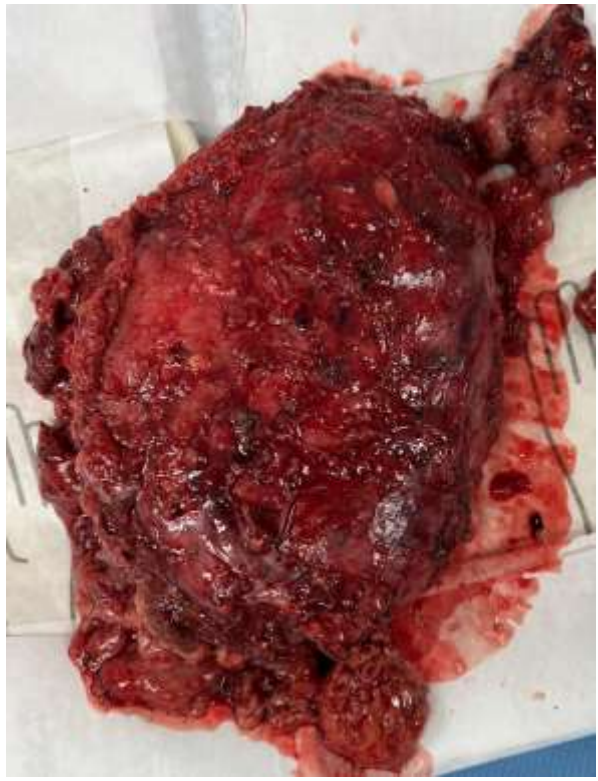


(fig. 1)

(fig. 2)



(fig. 3)



(fig. 4)

Further laboratory tests showed a morning cortisol level of 427 nmol/L, low factor VIII (61.6%), normal factor VII (106.2%), and von Willebrand factor results were pending. After initial evaluations, the patient's condition stabilized, and his pain was managed effectively without any further bleeding or hemoglobin decline. He was discharged for outpatient follow-up.

At a follow-up in the endocrinology clinic, the patient appeared stable, weighing 90 kg with a height of 168 cm which categorizes the patient as obese despite prior gastric sleeve surgery. He had a buffalo hump and recent-onset acne but no striae. Testing for pheochromocytoma yielded plasma metanephrine levels below 25 pg/mL, normetanephrine at 440 pg/mL, and 24-hour urinary metanephrine levels of 80 mcg, with urinary normetanephrine at 174 mcg. A dexamethasone suppression test indicated that non-suppressed cortisol and plasma ACTH were low at 2.62 pg/mL, with urinary ACTH at 688.8 pg/mL. HbA1c was 5.9%.

The patient was diagnosed with ACTH-independent Cushing’s syndrome, along with hypertension, pre-diabetes, and gallbladder stones. After completing the workup, he was electively admitted for laparoscopic adrenalectomy. The preoperative assessment showed the patient to be symptom-free, with stable vitals and a soft, non-distended abdomen.

Intraoperatively, the left adrenal gland was markedly enlarged (10x7x14 cm), with adhesions to the descending colon, abdominal wall, and left kidney. Histopathological analysis confirmed left adrenal cortical hyperplasia with extensive hemorrhage and infarction, predominantly involving the zona reticularis. (fig. 4) The patient's postoperative course was uneventful, and he was discharged for outpatient follow-up.

## 3. Discussion

There is no specific clinical presentation associated with adrenal hemorrhage, but a delay in diagnosis and management can be life-threatening. Abdominal pain due to adrenal hemorrhage is an exceedingly rare initial manifestation of Cushing syndrome, with most cases arising from trauma or underlying medical conditions. [1] However, in this case, the bleeding occurred spontaneously. Additionally, the patient had a history of persistent obesity despite undergoing sleeve gastrectomy. The persistence of the condition post-bariatric surgery reflects the complexity of weight management in such patients. The patient was later diagnosed with ACTH-independent Cushing syndrome. While Cushing syndrome is generally caused by adrenal adenomas or hyperplasia, it is unusual for adrenal hemorrhage to be the presenting feature. [1,2]

Computed tomography scanning has played a vital role in guiding the diagnosis of such cases. Previous cases of adrenal hemorrhage were only diagnosed during autopsy. [1,2] Initially, imaging suggested the presence of a pheochromocytoma, a diagnosis often considered when adrenal bleeding is detected. [3] Standard laboratory investigations that are often ordered as initial workups for abdominal pain do not often help in establishing a diagnosis in such cases. [2,4] However, further diagnostic workup ruled out pheochromocytoma.

This case emphasizes the diagnostic challenges in such scenarios. Computed tomography (CT) scanning has made the management of adrenal hemorrhage less invasive. Depending on the stability of the patient, cases can be managed conservatively or via angioembolization, yielding better outcomes for elective interval adrenalectomy. [4]

In our case, we were able to treat the patient after acute presentations as an outpatient for elective surgery. The size of the adrenal gland and the pathological findings suggest a significant adrenal abnormality that contributed to both the hemorrhage and Cushing syndrome. In our patient's case, the initial presentations of adrenal hemorrhage secondary to Cushing's, the data is limited.

## 4. Conclusion

This case highlights the rare presentation of spontaneous adrenal hemorrhage as the initial manifestation of ACTH-independent Cushing syndrome due to adrenal cortical hyperplasia. The absence of trauma or underlying coagulopathy, coupled with the patient's relatively healthy history, posed a significant diagnostic challenge. The incidental finding of Cushing syndrome following the adrenal hemorrhage

underscores the complexity of such presentations and the importance of thorough investigation in atypical cases.

The successful management of this patient through a multidisciplinary approach, including conservative treatment, accurate diagnosis, and elective laparoscopic adrenalectomy, demonstrates the efficacy of a coordinated effort in addressing this rare condition. Early identification and intervention are critical to achieving favorable outcomes, as delays in diagnosis can lead to life-threatening complications. This case contributes to the limited data on the relationship between spontaneous adrenal hemorrhage and Cushing syndrome, emphasizing the need for awareness and careful evaluation in similar presentations.

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Authors' Contributions:

AS, RA contributed to the literature review and writing the case, and MS, MA paraphrased the case. AS and RA helped in the writing of the case report and images. All authors have reviewed and approved the final manuscript.

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Declarations:

Ethical Approval and Consent to Participate: Not applicable

Consent for Publication: written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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