

Spontaneous Superior Adrenal Artery Rupture Causing a Large Retroperitoneal Hematoma: Case Report and Literature Review

Luigi Cayre, Alberto Aiolfi*, Andrea Sozzi, Davide Bona

***Corresponding author:**

Alberto Aiolfi, MD FACS FEBS (UGI)
I.R.C.C.S Ospedale Galeazzi
Sant'Ambrogio, University of Milan
Via Cristina Belgioioso, 173, 20157
Milan, Italy
E-mail: alberto.aiolfi86@gmail.com

Department of Biomedical Science for Health, I.R.C.C.S Ospedale Galeazzi Sant'Ambrogio,
Division of General Surgery, University of Milan, Italy

ABSTRACT

Introduction: Spontaneous hemorrhage resulting from rupture of an adrenal artery is extremely rare. The exact pathophysiology of this phenomenon remains unclear in patients not assuming oral anticoagulant or antiplatelet therapies.

Case report: A 55-year-old male with no comorbidities was referred to our emergency department with worsening abdominal pain after iterate episodes of violent coughing. The abdominal CT scan revealed a large retroperitoneal hematoma resulting from the rupture of the superior adrenal artery with active arterial contrast extravasation. The bleeding was successfully managed via angioembolization. The post-interventional course was uneventful.

Discussion: Spontaneous bleeding caused by rupture of an adrenal artery is a rare condition. The available literature primarily focuses on patients assuming anticoagulant or antiplatelet therapy. The exact pathophysiology of spontaneous arterial bleeding originating from adrenal gland remains unknown. It has been hypothesized that the configuration of the vascular network inside the adrenal gland may predispose to development of turbulent flow with consequent increased risk of arterial rupture in case of increased intra-abdominal pressure. Treatment may be different (conservative, angioembolization, and surgery) depending on the severity of bleeding, hematoma extent, and patient hemodynamic stability.

Conclusions: We report the case of a spontaneous superior adrenal artery rupture with retroperitoneal hematoma after iterate episodes of violent coughing in a patient not assuming antiplatelet or anticoagulant medications. Hypertension and violent coughing likely contributed to the abrupt increase in intravascular pressure within the adrenal vessel leading to rupture. In hemodynamically stable patients with active adrenal artery bleeding, angioembolization may be considered as valuable treatment option. Further research is warranted to elucidate the underlying mechanisms and establish the best treatment strategies for this rare condition.

Key words: spontaneous adrenal hemorrhage; adrenal artery rupture; retroperitoneal hematoma; coughing; angioembolization

Received: 10.04.2023

Accepted: 14.06.2023

INTRODUCTION

Spontaneous hemorrhage originating from rupture of an adrenal artery is rare (1). There is a paucity of literature on this specific topic while only few cases have been reported. The majority are related to spontaneous venous bleeding

in patients assuming anticoagulant or antiplatelet treatments. The pathophysiology of spontaneous suprarenal gland's bleeding remains indefinite. The adrenal gland is one of the most well-perfused organs of the body and the vascular network is complex. The gap between the rich arterial inflow and the relatively limited venous drainage has been advocated to potentially cause a "vascular barrier" with development of turbulent flow thus potentially predisposing to spontaneous arterial bleeding in case of abrupt increase of intra-abdominal pressure (2). Treatment remains controversial and depends on the extent of hematoma, bleeding severity, degree of hormonal insufficiency and hemodynamic stability (3). Treatment options are conservative, angioembolization and surgery.

We report the case of retroperitoneal bleeding originating from spontaneous superior adrenal artery rupture after iterate and violent episodes of coughing. To the best of our knowledge this is the first case of spontaneous superior adrenal artery rupture after episodes of violent coughing in a patient not assuming antiplatelet or anticoagulant medications and with no evidence of adrenal artery aneurysm.

CASE PRESENTATION

A 55-year-old man was referred to our emergency department for the onset of abdominal pain after iterate episodes of forceful cough. Past medical history included hypertension. No medications were reported. On admission, physical examination was unremarkable and patient vital signs were stable with blood pressure of 150/90 mmHg, heart rate of 108 bpm and oxygen saturation of 97% on room air. Laboratory work-up showed reduced hemoglobin (8.7 g/dL) and hematocrit (34%) concentrations, normal C-reactive protein (3.3

mg/dL), white blood cells (WBC) (8020 mm^3) and I.N.R. (1.13). Abdominal computer tomography (CT) scan showed a large ($12 \times 8.4 \times 11 \text{ cm}$) retroperitoneal hematoma on the left adrenal lodge with high density center and active arterial contrast extravasation (*fig. 1*). Selective arteriography confirmed an active retroperitoneal blush arising from the superior left adrenal artery with no concomitant aneurysms. Angioembolization with coils was successfully performed. The post-interventional course was uneventful. Abdominal CT scan on postinterventional day 2 did not show residual arterial blush (*fig. 2*). The patient was discharged on post procedural day 3 in good clinical conditions with stable hemoglobin (10.8 g/dL) and hematocrit (31%) concentrations. The 12-month follow-up abdominal CT scan did not show evidence of residual collections in the left adrenal lodge with no alteration of the gland function.

DISCUSSION

Spontaneous adrenal hemorrhage from adrenal artery rupture is rare (1). There is paucity of literature reporting this topic while foremost risk factors for non-traumatic hemorrhage include: hemorrhagic diathesis, adrenal tumors, arterial aneurysm, pregnancy, antiphospholipid syndrome, coagulopathies, thrombocytosis, anticoagulant and antiplatelet medications (3). Most of previously reported cases describe the treatment of spontaneous venous bleeding in patients assuming anticoagulant or antiplatelet treatments (*table 1*) (1-3).

The pathophysiology of spontaneous arterial suprarenal gland bleeding remains unknown. While the adrenal gland is well-perfused, its vasculature is complex with a complex vascular network. Specifically,

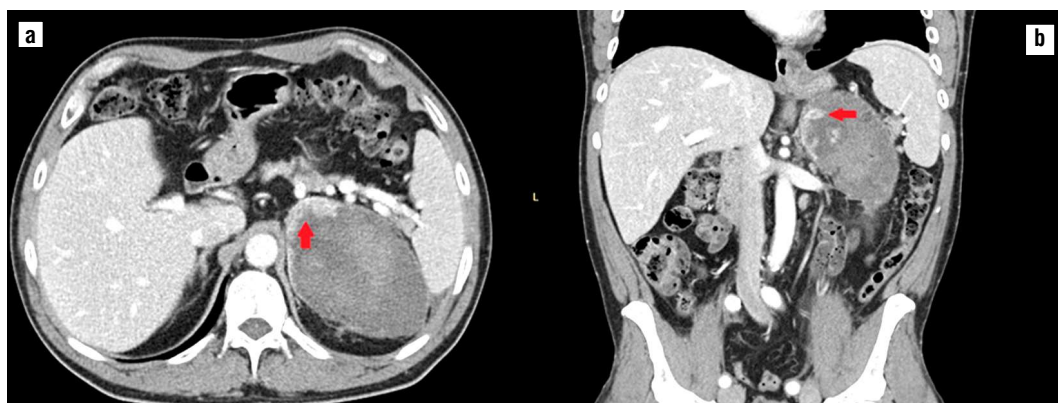


Figure 1 - Abdominal computer tomography showing the retroperitoneal hematoma on the left adrenal lodge with high density center and contrast active extravasation (red arrow). Axial (a) and coronal (b) plane.



Figure 2 - Abdominal computer tomography after angioembolization of the superior adrenal artery showing no residual contrast extravasation. Axial (A) and coronal (B) plane.

three adrenal arteries (superior, middle and inferior) originate from the inferior phrenic arteries, abdominal aorta and renal arteries, respectively. Afterward they brusquely divide into 50-60 capillary branches draining into medullary sinusoids thus generating one adrenal vein. Venous drainage is therefore limited to a single vein which drain into the inferior vena cava on the right and the renal vein on the left. This gap between the rich arterial inflow and the relatively narrow venous drainage has been advocated to be possibly cause a “vascular barrier” with possible generation of turbulent flow (3). Capillary fragility, especially of those capillaries situated within the distal corticomedullary junction, associated to this particular configuration may theoretically predispose to spontaneous nontraumatic

bleeding in case of abrupt increase of intra-abdominal pressure as may occur during a violent cough.

Treatment is controversial and depends on the extent of hematoma, bleeding severity, and hemodynamic status (4). Options are conservative treatment, angioembolization and surgery. In case of significant bleeding and hypotension or in the absence of interventional radiology facilities the surgical option may be indicated (1-3). In contrast, if the patient is hemodynamically stable and there are no signs of active extensive bleeding a watch-and-wait approach is prudent. In our patient, because evidence of active extravasation with concomitant hemodynamic stability we aimed for selective arteriography and angioembolization of the superior adrenal artery.

Table 1 - Literature review; spontaneous adrenal hemorrhage from adrenal arteries. nr not reported. Legend AG adrenal gland

Author, year, country	No. pts	Age (yrs)	Sex	Risk factors/ Comorbidities	CT-scan			Treatment	Antiplatelet medication
					Arterial aneurism	Hematoma dimension / location	Artery location		
González Valverde, 2006, Spain (1)	1	42	M	Poorly controlled hypertension	Yes	15x11x9 cm / Left AG	Inferior adrenal artery	Embolization	nr
Mazumder, 2011, USA (2)	1	60	F	Bronchial asthma and violent coughing; Hypertension; Coronary artery disease	No	Nr/Right AG	Non specified adrenal artery	Conservative	Clopidogrel Aspirin
Charalampakis, 2018; UK and Greece (3)	2	47	M	Smoke; Obesity; Uncontrolled hypertension	No	Nr/Left AG	Non specified adrenal artery	Conservative	Aspirin
		60	M	Hypertension; Myocardical infarction	No	Nr / Left AG	Middle adrenal artery	Embolization	Clopidogrel Aspirin

CONCLUSIONS

To the best of our knowledge this is the first case of spontaneous superior adrenal artery rupture with retroperitoneal hematoma after iterate episodes of violent cough in a patient not assuming antiplatelet /anticoagulant medications with no evidence of arterial aneurysm. We can theoretically assume that hypertension in conjunction with forceful cough can induce an abrupt increase of intravascular pressure in the adrenal vessels with a potential increased of full-thickness rupture risk.

Author's contributions

LC: data analysis & collection, manuscript writing. AA: project development, data analysis & collection, manuscript writing. AS: data analysis & collection. DB: project development, data analysis & collection, manuscript writing.

Conflict of interest

The authors declare no competing interests.

Funding

No funding sources.

Informed consent

Informed consent was obtained from the patient.

REFERENCES

1. González Valverde FM, Balsalobre M, Torregrosa N, Molto M, Gómez Ramos MJ, Vázquez Rojas JL. Spontaneous retroperitoneal hemorrhage from adrenal artery aneurysm. *Cardiovasc Intervent Radiol*. 2007;30(2):307-9.
2. Mazumder M, Afzal A, Bhagat R, Arya M, Misra P, Mutyala M. Retroperitoneal hematoma due to violent coughing: rare presentation of abdominal pain. *American J Gastroenterology*. 2011;106: S274.
3. Charalampakis V, Stamatou D, de Bree E, Christodoulakis M, Zoras O. Spontaneous adrenal hemorrhage. Report of two cases and review of pathogenesis, diagnosis and management. *J Surg Case Rep*. 2018;2018(6):rjy129.
4. Aiolfi A, Inaba K, Strumwasser A, Matsushima K, Grabo D, Benjamin E, et al. Splenic artery embolization versus splenectomy: Analysis for early in-hospital infectious complications and outcomes. *J Trauma Acute Care Surg*. 2017;83(3):356-360.