


Case Report

Spontaneous adrenal hemorrhage with dehydroepiandrosterone-sulfate elevation in a patient with suspected adrenal cortical carcinoma

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Abbreviations & Acronyms

ACC = adrenal cortical carcinoma
 CT = computed tomography
 DHEA-S = dehydroepiandrosterone-sulfate
 FDG = fluorodeoxyglucose
 MRI = magnetic resonance imaging
 PET = positron emission tomography

Introduction: Spontaneous adrenal hemorrhage is a relatively rare disease that is sometimes difficult to differentiate from adrenal cortical carcinoma. We herein report a case of spontaneous adrenal hemorrhage with dehydroepiandrosterone-sulfate elevation.

Case presentation: The patient was a 78-year-old man with an adrenal tumor that had increased in size to 42 mm. With the exception of an elevated dehydroepiandrosterone-sulfate level (2281 ng/mL), the results of a hormone analysis were almost normal. Laparoscopic adrenal tumor resection was performed. The pathological diagnosis was adrenal hematoma.

Conclusion: We reported a case of spontaneous adrenal hemorrhage in a patient with dehydroepiandrosterone-sulfate elevation.

Key words: adrenal cancer, adrenal cortical carcinoma, adrenal hemorrhage, adrenal tumor, DHEA-S.

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Keynote message

DHEA-S was elevated by spontaneous adrenal hemorrhage.

Introduction

Spontaneous adrenal hemorrhage is a relatively rare disease that is sometimes difficult to differentiate from ACC.¹ We herein report a case of spontaneous adrenal hemorrhage in a patient with DHEA-S elevation.

Case presentation

The patient was a 78-year-old man who was undergoing follow-up after receiving brachytherapy for prostate cancer 1 year previously. He had a 10-year history of hypertension and was controlled by amlodipine besylate 5 mg. CT revealed an adrenal tumor initially detected a year ago that had increased in size from 26 to 42 mm (Fig. 1). With the exception of DHEA-S elevation (2281 ng/mL), a hormone analysis revealed almost normal findings (adrenocorticotropic hormone 29.7 pg/mL, cortisol 8.6 µg/dL, aldosterone 69.6 pg/mL, adrenaline 34 pg/mL, noradrenaline 119 pg/mL, dopamine <5 pg/mL, urine metanephrine 0.05 mg/day, urine normetanephrine 0.08 mg/day, renin activity 0.5/mL/h). PET-CT revealed no uptake of FDG by the adrenal tumor (Fig. 2). Although there was no uptake of FDG, we decided to perform surgical resection due to increase in the size of the tumor and elevation of the patient's DHEA-S level. Laparoscopic adrenal tumor resection was performed. The tumor was not adhered to the surrounding organs and easily to be resected. The resected specimen was 50 × 22 × 13 mm in size and weighed 45.5 g. The pathological diagnosis was adrenal



Fig. 1 CT revealed left adrenal mass from (a) 26 mm to (b) 42 mm. (c) Preoperative contrast-enhanced CT.

hematoma covered by fibrosis and hyalinization (Fig. 3). No apparent malignant findings were observed. After surgery, the patient's DHEA-S level decreased to 164 ng/mL.

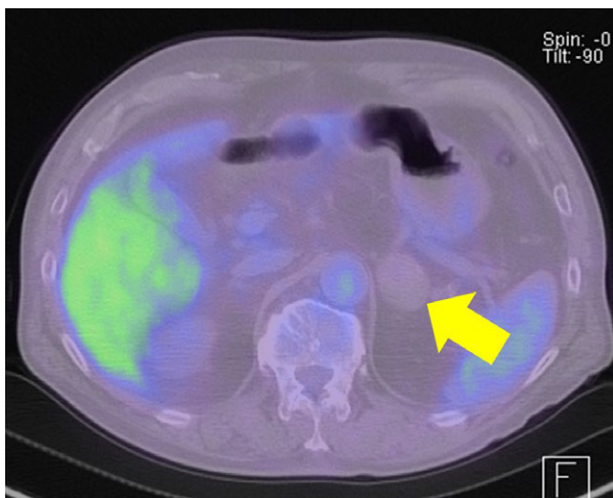


Fig. 2 PET-CT revealed no FDG uptake.

Discussion

Marti *et al.* reported 133 cases of adrenal hemorrhage in patients with various conditions, including pheochromocytoma (46%), metastatic adrenal tumor (13%), myelolipoma (13%), hemorrhage (12%), ACC (7%), adenoma (4%), and others (5%).^{2,3} Adult renal hemorrhage is a relatively rare disease that can be classified as injury-associated cases and non-injury cases. Approximately 63% of adult adrenal hemorrhage cases are considered to be injury-associated cases.⁴ The patient in the present case showed spontaneous adrenal hemorrhage, and was therefore categorized as a non-injury case. Only 20–30 cases of spontaneous adrenal hemorrhage have been reported in Japan.^{1,5,6}

The detailed mechanisms of spontaneous adrenal hemorrhage are still unknown. One possible mechanism involves the blood supply of the adrenal gland. The adrenal gland is supplied by many tiny arteries, but the outlet flow is limited and the adrenal vein is very fragile. Increasing blood pressure increases the risk of hemorrhage from the adrenal vein. Another possible mechanism is that the production of adrenocortical steroids, which is induced by stress, results in higher adrenal flow. The clinical symptoms include upper and flank abdominal pain followed by fever, hypotension, a decreased appetite, and vomiting.⁷

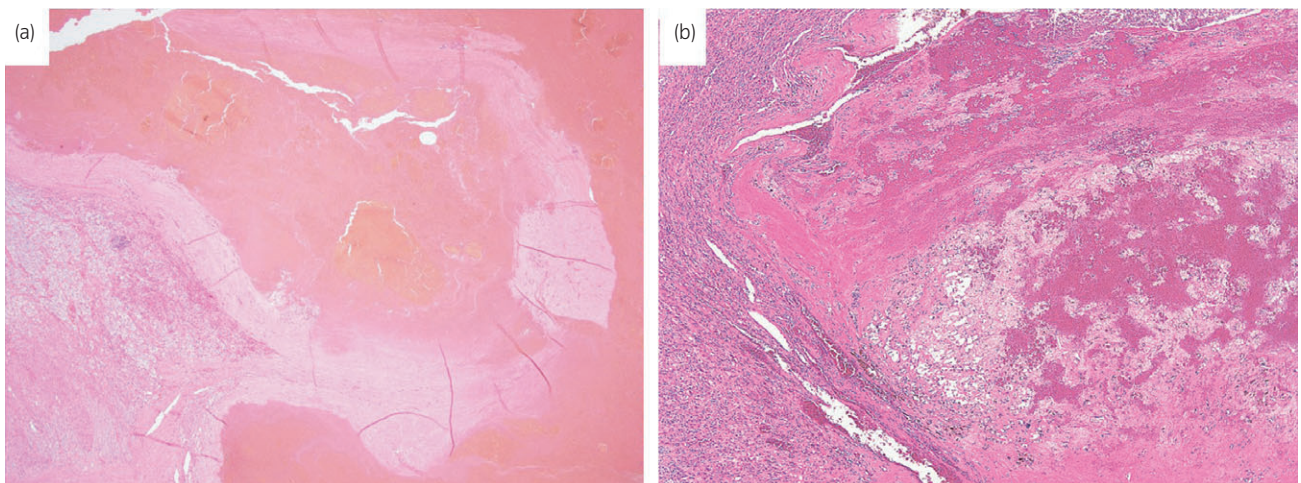


Fig. 3 HE in (a) low power field and (b) high power field.

The present patient displayed none of these characteristics. CT, ultrasonography, and MRI were difficult to determine adrenal hemorrhage or ACC. Ultrasonography revealed a low echoic lesion covered by highly echoic organized tissue. CT revealed an area that displayed high density just after hemorrhage had a low density at 2–3 weeks after hemorrhage. In the present case, the timing of hemorrhage was not clear due to the absence of clinical symptoms. MRI revealed an area of low intensity on T1WI just after hemorrhage that had a high intensity on T2WI. This gradually changed to low intensity on T2WI.^{8,9} FDG-PET CT has been reported to be useful for detecting pheochromocytoma, ACC, and metastatic adrenal tumors. On the other hand, adrenocortical hyperplasia and adrenal hemorrhage also show the uptake of FDG in some cases.¹⁰

In the present case, we initially observed an adrenal tumor that was slightly increasing in size, with DHEA-S elevation. These findings led us to perform surgical resection. DHEA-S elevation is sometimes seen in ACC. A tumor size >4 cm is a well-known imaging finding for differentiating malignant tumors, with a sensitivity of 81%.¹¹ A previous report showed that 10%, 19%, and 47% of adrenal tumors with diameters of <4 cm, <6 cm, and <8 cm were ACCs.^{12,13} ACC loses the uniformity of steroidal synthesis, which results in the upregulation of urinary 17-ketosteroid. DHEA-sulfotransferase, which transfers DHEA to DHEA-S, is often seen in ACC in comparison to other adrenal tumors, and the serum DHEA-S is elevated in ACC patients.¹⁴ However, not all cases of ACC produce DHEA-S and the correlation between DHEA-S elevation and adrenal hemorrhage has not been reported.¹⁵ Based on these findings, surgical resection was performed despite the fact that the tumor showed no uptake of FDG.

In conclusion, we reported a case of spontaneous adrenal hemorrhage with DHEA-S elevation.

Conflict of interest

The authors declare no conflict of interest.

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