

[CASE REPORT]

Giant Cavernous Hemangioma of the Adrenal Gland in an Elderly Patient

Ayana Hashimoto¹, Hiroshi Yoshino¹, Fukumi Yoshikawa¹, Naoki Kumashiro¹,
Yasuyo Ando¹, Fumito Yamabe², Yuri Akishima-Fukasawa³, Naoko Honma³,
Hiroshi Uchino¹ and Takahisa Hirose¹

Abstract:

Cavernous hemangioma is a rare, non-functional, benign adrenal tumor. Adrenal cavernous hemangioma is often diagnosed after surgery with a histologic examination. A 70-year-old man complaining of appetite loss was admitted to our hospital. An incidental large left adrenal mass was found by computed tomography (CT). There were no clinical signs of adrenogenital syndrome, Cushing's syndrome or primary aldosteronism. Total resection was carried out. The pathological diagnosis was cavernous hemangioma. The differentiation of adrenal tumor is necessary in cases of large tumors, and resection is desirable given the risks of hemorrhaging and rupture.

Key words: cavernous hemangioma, adrenal gland, elderly

(Intern Med 57: 1317-1319, 2018)

(DOI: 10.2169/internalmedicine.9711-17)

Introduction

Cavernous hemangioma is a rare, non-functional, benign adrenal tumor. Only 63 cases have been reported (1). Adrenal cavernous hemangioma is often diagnosed after surgery with a histologic examination. Patients usually have no signs or symptoms and are noticed only when they develop a large palpable mass or hypovolemic shock caused by spontaneous rupture. It is very difficult to distinguish a large adrenal hemangioma from a malignant tumor. We herein report a case of giant cavernous hemangioma of the adrenal gland in an elderly patient.

Case Report

A 70-year-old Asian man was admitted to our hospital because of loss of appetite. He had no significant medical history. A physical examination revealed a mass on the left hypochondrium without tenderness. An incidental large left adrenal mass (14.7×15.8×10.6 cm) was found by computed to-

mography (CT).

All laboratory data were almost within normal ranges (Table 1), and the findings on functional investigations concerning blood cortisol and dexamethasone suppression test were almost normal (Table 2).

On magnetic resonance imaging (MRI), the mass showed a heterogeneous area with an internal hemorrhagic component (Figure A). Although positron emission tomography (PET)-CT scan was performed, there was no hypermetabolic area in the left adrenal gland mass. There were no clinical signs of adrenogenital syndrome, Cushing's syndrome or primary aldosteronism. On the 8th day, total resection was carried out. A pathological examination revealed a 27×17×5.5 cm adrenal mass weighing 2,000 g with peripheral sectors of the adrenal cortex (Figure B). A histopathological evaluation revealed cavernous proliferation showing a single layer of endothelial cells in the peripheral area of the mass (Figure C). A large necrotic area was observed centrally. Even in the necrotic area (Figure D), an immunohistochemical examination showed CD 31 positivity, suggesting a vascular origin (Figure E). The pathological diagnosis was cav-

¹Department of Internal Medicine, Division of Diabetes, Metabolism and Endocrinology, Japan, ²Department of Urology, Toho University Graduate School of Medicine, Japan and ³Department of Pathology, Toho University Graduate School of Medicine, Japan

Received: June 24, 2017; Accepted: August 29, 2017; Advance Publication by J-STAGE: December 27, 2017

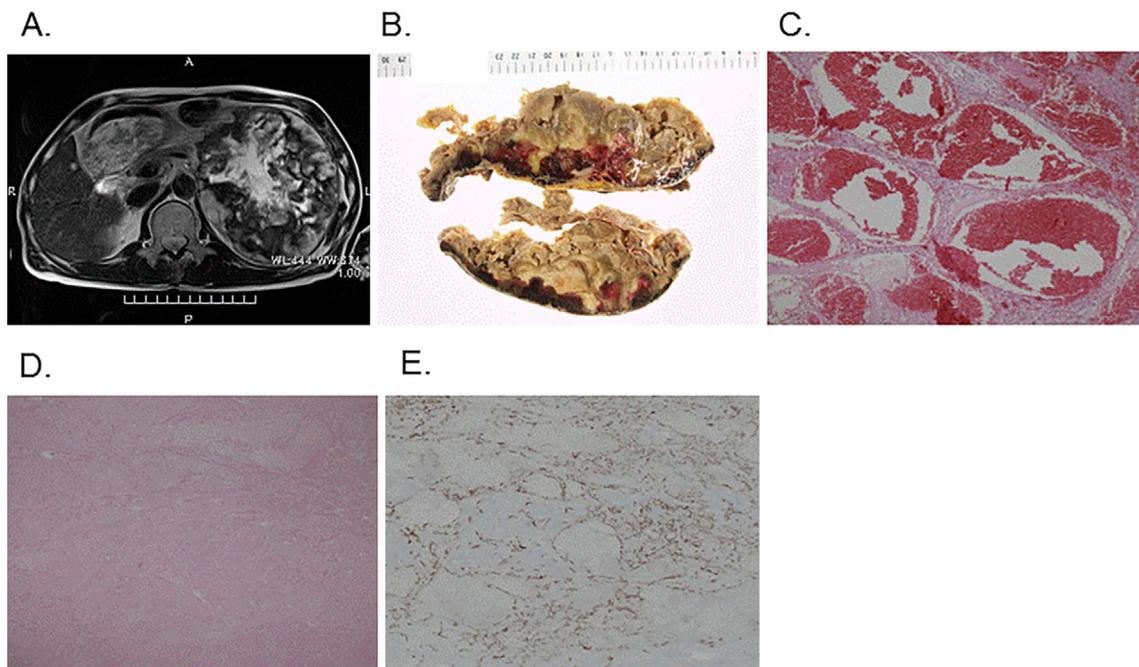
Correspondence to Dr. Hiroshi Yoshino, hiroshi.yoshino@med.toho-u.ac.jp

Table 1. Laboratory Data.

	Before Surgery	After Surgery 8 months later	Reference Range
Plasma ACTH	7.8	30.1	7.2-63.3 (pg/mL)
Serum cortisol	14.7	9.68	6.2-33.1 (µg/dL)
Plasma renin Activity	<0.2	0.7	0.2-3.9 (Ung/mL/hr)
Plasma aldosterone Concentration	50.5	32.2	29.9-159 (pg/mL)
Urine free cortisol	60.3		35-160 (µg/day)
Plasma			
Epinephrine	≤0.01		≤0.17 (ng/mL)
Norepinephrine	0.11		0.15-0.57 (ng/mL)
Dopamine	≤0.02		≤0.03 (ng/mL)
Urine			
Epinephrine	1.5		1-23 (µg/day)
Norepinephrine	36.7		29-120 (µg/day)
Dopamine	410		100-1,000 (µg/day)

Table 2. Functional Investigations Concerning Blood Cortisol and Dexamethasone Suppression Test.

Diurnal rhythm	Clock time	6:00	20:00	23:00
		ACTH (pg/mL)	7.8	3.3
	Cortisol (µg/dL)	14.7	2.7	2.2
Dexamethasone test	Dexamethasone (mg)		1mg	8 mg
	ACTH (pg/mL)		≤0.2	≤0.2
	Cortisol (µg/dL)		1.1	1.1

**Figure.** A: Magnetic resonance imaging transverse section (T2-weighted imaging) revealing a high-signal-intensity mass with a heterogeneous internal structure. B: Large adrenal hemangioma. C, D: Hematoxylin and Eosin staining (×40). E: Immunohistochemistry for CD31 (×40).

ernous hemangioma. Fourteen days later, the patient recovered without any surgery related complications and was discharged. Eight months later, his laboratory data had recovered to almost normal levels (Table 1).

Discussion

Cavernous hemangioma is a rare, non-functional, benign adrenal tumor. Due to the low frequency and the lack of specific symptoms, the majority of adrenal hemangiomas are diagnosed postoperatively. The recent increase in the detection rate of adrenal masses may be due to the widespread use of modern imaging techniques. In cavernous hemangiomas of the adrenal gland, MRI may indicate a homogeneous adrenal mass on T1-weighted imaging with a high intensity signal on T2-weighted imaging (2). However, Kieger et al., pointed out that the appearance of adrenal hemangioma on MRI is also frequently non-specific (3). According to Noh et al., the most common symptom was flank pain or discomfort (21.2%). Only 5.8% of cases were reported to show endocrinologic disturbances with subclinical Cushing's syndrome and primary hyperaldosteronism (4). They hypothesized that the hemangioma itself does not have endocrine activity, but arteriovenous malformations within it may release endocrinologically-active metabolites into the bloodstream (4). Adrenal cavernous hemangiomas are often detected at older ages simply because they usually do not manifest clinical symptoms until the size of the mass has grown significantly (4). Histologically, adrenal hemangiomas are usually cavernous and rarely capillary in type. The lesions are well-encapsulated and located in the adrenal cortex (5). Forbes reported that the indications for resection of

this rare neoplasm are to relieve mass-effect-type symptoms, to exclude malignancy, and to treat complications such as hemorrhaging (6).

In summary, the differentiation of an adrenal tumor is necessary in cases of large tumors, and resection is desirable given the risks of hemorrhaging and rupture.

The authors state that they have no Conflict of Interest (COI).

Acknowledgement

We thank Dr. Nobuyuki Shiraga, Dr. Kayoko Ikehara, and Prof. Tetsuo Mikami for their advice during the preparation of this manuscript.

References

1. Wang L, Dang Y, He R, Chen G. Rare cavernous hemangioma of adrenal gland. *Sao Paulo Med J* **132**: 249-252, 2014.
2. Silverberg D, Paramesh AS, Roayaie S, Swartz ME. Giant hemangioma of the adrenal. *Isr Med Assoc J* **6**: 705-706, 2004.
3. Kieger AJ, Nikolaidis P, Casalino DD. Adrenal gland hemangioma. *J Urol* **186**: 2415-2416, 2011.
4. Noh JJ, Choi SH, Hwang HK, Kang CM, Lee WJ. Adrenal cavernous hemangioma: a case report with review of the literature. *JOP* **15**: 254-257, 2014.
5. Tarchouli M, Boundhas A, Ratbi MB, et al. Giant adrenal hemangioma: unusual cause of huge abdominal mass. *Can Urol Assoc J* **9**: 11-12, 2015.
6. Forbes TL. Retroperitoneal hemorrhage secondary to a ruptured cavernous hemangioma. *Can J Surg* **48**: 78-79, 2005.

The Internal Medicine is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (<https://creativecommons.org/licenses/by-nc-nd/4.0/>).