Case Report

A Huge Adrenal Hematoma in an Adrenal Cavernous Hemangioma Manifesting as a Chronic Expanding Hematoma of the Retroperitoneum: A Case Report

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Abstract

This report presents the case of a huge chronic expanding hematoma in the right adrenal gland. A 66-year-old Japanese woman was referred for the evaluation of a huge mass in the middle of her abdomen. Her physical condition was unremarkable except for bilateral edema of the lower extremities. She had undergone hysterectomy and left oophorectomy due to uterine leiomyoma 18 years earlier, but her medical history had included neither anticoagulant therapy nor abdominal trauma. The laboratory data revealed anemia but showed that all tumor markers were within normal range. In the right abdomen, abdominal computed tomography (CT) demonstrated a well-demarcated, 35-cm mass compressing the surrounding organs to the left side. The peripheral region of the mass was slightly enhanced, and the central part was hypovascular, indicating central necrosis or hematoma. No right adrenal gland was identified. Magnetic resonance imaging showed that the inner aspect of the mass appeared to be unequally hyperintense on T1- and T2-weighted images. The rim of the mass was hypointense on the T2-weighted image but hyperintense on diffusion-weighted images. Abdominal CT angiography indicated that the right capsular branches of the right renal artery and right inferior phrenic artery were feeding arteries for the mass. The preop-

erative diagnosis was a chronic expanding hematoma, but the central necrosis of the tumor was not completely ruled out. Laparotomy revealed that the huge mass was encapsulated. Moreover, no tight adhesion was observed, indicating that the lesion could be easily detached. Histopathological examination of the surgical specimen showed that it was a hematoma, and the rim of the hematoma was composed of the adrenal cortex and medulla, suggesting the hematoma originated in the adrenal gland. Further, a cavernous hemangioma was observed in the periphery of the hematoma, suggesting the hemangioma was the cause of an adrenal hemorrhage and manifested as a chronic expanding hematoma. This report describes the largest adrenal hematoma reported in Japan, which presented as a chronic expanding hematoma of the retroperitoneum.

Key words: retroperitoneum, chronic expanding hematoma, adrenal cavernous hemangioma, adrenal hematoma

Introduction

A chronic expanding hematoma (CEH) was first described by Reid et al. in 1980¹⁾. CEH persists and enlarges for more than 1 month after hemorrhage in patients with no coagulation disorders and no malignancies. CEH occurs after surgery or trauma but in some cases is idiopathic. CEH has been observed in a variety of locations, such as the head, chest, abdomen, and extremities, and often simulates neoplasms. As reported in Japanese case reports, CEH occurs most frequently in the intrathoracic cavity

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Fig. 1 Abdominal computed tomography (CT) and CT angiogram. The axial (a) and coronal (b) view of the CT showed that a round, encapsulated, well-demarcated mass was located at the right abdomen and that the surrounding organs were displaced to the left side. The inner lesion was hypointense, suggesting the lesion to be an old hemorrhage or hematoma. The rim of the mass showed spotty contrast enhancement. CT angiogram showed that the capsular branches of the right renal artery (arrow) and the right inferior phrenic artery (arrowhead) were the feeding arteries for the mass (c).

and very rarely in the retroperitoneum²⁾. This report describes a rare clinical case of a huge CEH that occurred in the retroperitoneum and that was caused by a hemorrhage in an adrenal cavernous hemangioma.

Case Report

A 66-year-old Japanese woman was admitted to the hospital because of enlargement of an abdominal mass in February 2012. She first noticed the abdominal mass in September 2011. She had undergone hysterectomy and left oophorectomy due to uterine leiomyoma 18 years earlier, but her medical history included neither anticoagulant therapy nor abdominal trauma. The results of the physical examination were unremarkable other than a hard unmovable mass in the middle of her abdomen and bilateral edema of the lower extremities. Despite the huge mass, no respiratory failure, dietary distur-

bance, or constipation was present. Her laboratory data showed that anemia was present but that all tested tumor markers were within normal range. Gastrointestinal radiographic and endoscopic examinations showed the compression of the alimentary tracts but no mucosal lesions in the stomach or large intestine.

Plain chest and abdominal radiographs showed an elevation of the right diaphragm and an abdominal tumor shadow. Abdominal computed tomography (CT) revealed a well-demarcated huge mass in the right abdomen compressing the right kidney to the low-medial side, as well as the deviation of the portal vein and inferior vena cava to the left side due to the compression by the mass (**Fig. 1a, b**). These findings indicated the huge mass was a retroperitoneal tumor. The rim of the tumor was enhanced and showed a nodular or granular appearance, while the inner portion of the mass was less enhanced,

Fig. 2 Abdominal magnetic resonance imaging (MRI). MRI showed the signal of the mass to be heterogeneously high on T1 and T2-weighted images, whereas the intensity of the rim of the mass was low on T2-weighted MRI (a, b). Diffusion-weighted imaging showed the irregularly high intensity in the periphery of the mass (c).

thus suggesting hemorrhagic necrosis or a hematoma (**Fig. 1a, b**). No right adrenal gland was observed. Abdominal CT angiography showed dilatation of the right capsular branches of the right renal artery and the right inferior phrenic artery, indicating these arteries were the feeding arteries for the mass (**Fig. 1c**). The T1- and T2-weighted images revealed the inner aspect was unequally hyperintense (**Fig. 2a, b**). Moreover, the rim of the mass was unequally hypointense on T2-weighted magnetic resonance imaging (MRI; **Fig. 2b**), but hyperintense on diffusion-weighted images (**Fig. 2c**). The preoperative diagnosis was a CEH.

Laparotomy with an abdominal median incision showed that a huge mass, 35cm in diameter, occupied the abdominal cavity, and that all of the abdominal organs were shifted to the left side. The mass was well capsulated and slightly adhered to the surrounding organs. No thick feeding arteries

were observed, and the right adrenal gland was unrecognized. The tumor was safely resected. The surgery lasted for 3 hours, and the blood loss was 1,360ml. The blood loss was due to exudative bleeding from the ablated side. Blood transfusion was not performed.

A macroscopic examination revealed the specimen had a smooth surface, measured 30 × 35cm in size, and weighed 8,500g. Dark red and altered blood filled the mass. The patient was discharged without any complications 8 days after the operation, and she showed no symptoms 3 months after the surgery. Leg edema and anemia were improved immediately. A microscopic examination showed the tumor was encapsulated by thin tissue and was filled with old coagulated tissue (**Fig. 3a**). Thin-section histopathology revealed no malignant cells and showed that the wall of the hematoma was composed of hyalinized connective tissue with spotty

Fig. 3 Gross appearance and histopathology. The cut surface of the removed specimen (diameter, 35cm; weight, 8,500g) revealed a well-defined dark red and solid mass (a). The cut surface of the rim of the mass showed yellowish and thin tissues (arrow), suggestive of the right adrenal gland (b). Histopathology of the wall of hematoma revealed the adrenal cortex (c. Hematoxylin and eosin stain, 100×). Cell clusters adjacent to the adrenal cortex were positive for chromogranin A, indicating these cells to be the adrenal medulla (d. inset. 200×).

a|b
c|d

calcification. Yellowish tissue was observed in the thin wall of the mass (**Fig. 3b**), which was revealed by histopathology to be the adrenal gland (**Fig. 3c**). Moreover, immunohistochemistry showed the cells adjacent to the adrenal cortex were positive for chromogranin A, indicating these cells were the adrenal medulla (**Fig. 3c**, inset). The thin wall of the mass was composed of the adrenal gland, indicating the hemorrhage had occurred in the center of the right adrenal gland. Histopathology also revealed cavernous endothelial proliferation adjacent to the adrenal gland, suggesting the hemorrhage occurred inside an adrenal cavernous hemangioma (**Fig. 3d**). The patient's clinical history, images, and pathologic features indicated the hemorrhagic lesion had persisted for a long period as a slowly expanding space-occupying mass. Therefore, the large mass was diagnosed as CEH due to an adrenal hemorrhage from a hemangioma.

Discussion

CEH is a rare condition and gradually increases in size over the long term. CEH may be misdiagnosed as a malignant soft tissue tumor because of its large size and slow progressive enlargement. An Ichushi search (in Japanese, Japan Medical Abstracts Society) for Japanese cases of CEH revealed that 82 cases were reported from 1987 to 2011, with most of these cases occurring in the chest or soft tissue. Kaneko et al. reviewed 6 cases of CEH in the retroperitoneum and found that 4 of the 6 cases were postoperative CEHs²⁾. The present case of CEH represents the largest adrenal CEH to date.

The MR T2-weighted image was useful for diagnosing CEH, and the mosaic of heterogeneous signal intensities (the so-called "mosaic sign") is a characteristic feature of CEH³⁾. However, the preoperative diagnosis of CEH is difficult in some CEH cases. Oka et al. described 2 of 6 CEHs as difficult to dif-

ferentiate from other diseases based on conventional MRI⁴⁾. Recent advances in the imaging modalities for the diagnosis of CEH include diffusion-weighted imaging (DWI). The mean apparent diffusion coefficient value on DWI in CEHs is significantly higher than that of soft tissue tumors; therefore, DWI is useful for differentiating CEHs from malignant soft tissue tumors⁴⁾. However, an accurate diagnosis of CEH is quite difficult to make prior to surgery, since central necrosis in the tumor cannot be completely excluded. Therefore, surgical exploration is recommended.

Marti et al. reported 6 cases of spontaneous adrenal hemorrhage with an associated mass and reviewed 133 reported cases⁵⁾. The median patient age was 50 years (range:17-77years), and 55% of the patients were male. Forty-nine percent of the lesions were right-sided; 46%, left-sided; and 5%, bilateral. The median size was 9cm (range: 1.5-25cm). The most frequently reported pathological diagnosis was pheochromocytoma (48%), followed by metastatic cancer of the adrenal glands (14%), hematoma (13%), myelolipoma (10%), adrenocortical carcinoma (7%), adrenocortical adenoma (4%), pseudocyst/hematoma in pregnancy (4%), and lipoma (1%).

Koizumi et al. described 14 cases of idiopathic adrenal hematoma from 1983 to 2010 on the basis of an Ichushi search (in Japanese), although no cases of adrenal hemangioma were found among these cases⁶⁾. These reports indicate that a hemorrhagic adrenal mass caused by a cavernous hemangioma is extremely rare. Cavernous adrenal hemangiomas are usually unilateral and become apparent in the sixth to seventh decade of life. Approximately 50 cases have been reported in Japan⁷⁾. This tumor is usually discovered as an incidental radiological feature without any clinical symptoms; therefore, by the time of diagnosis, the cavernous hemangioma is manifested as a large tumor⁸⁾.

Yamada et al. reported that contrast-enhanced CT displays a characteristic peripheral patchy enhancement and highly dense peripheral rim in adrenal hemangioma⁹⁾. This pattern of peripheral spotty contrast enhancement with centripetal enhancement is crucial for diagnosing adrenal hemangioma and these features were found in the current case. However, the differential diagnosis of an adrenal heman-

gioma from other neoplasms, such as adrenocortical adenoma and malignant tumors is difficult¹⁰⁾. Therefore, a precise preoperative diagnosis is often impossible to make, even though the adrenal hemangioma shows some characteristic radiological findings.

In conclusion, this report presented the case of a huge CEH of the retroperitoneum due to a hemorrhage in an adrenal cavernous hemangioma. Although an adrenal hemangioma is extremely rare, this tumor should be classed among the differential diagnoses of a retroperitoneal hematoma. Furthermore, careful pathological examination of the wall of the hematoma is necessary for the correct diagnosis of a cavernous hemangioma of the adrenal gland in cases where a CEH in the retroperitoneum is resected.

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