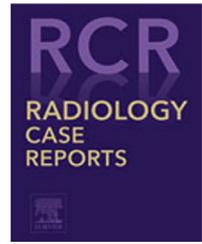


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Case Report

A collision between vascular adrenal cyst and adrenocortical adenoma ☆☆☆

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ARTICLE INFO

Article history:

Received 5 February 2021

Revised 27 February 2021

Accepted 28 February 2021

Keywords:

Adrenal vascular cyst

Adrenocortical adenoma

Collision tumor

ABSTRACT

We report a patient with sigmoid colon cancer who revealed a unique collision of hemorrhagic vascular adrenal cyst and adrenocortical adenoma with myelolipomatous changes. Two months before referral to our hospital, anticoagulant therapy was started for acute myocardial infarction. The components of the adrenocortical adenoma demonstrated a typical signal drop in opposed-phase magnetic resonance (MR) images although macroscopic fat was also depicted both on CT and MR images. The components of the vascular adrenal cyst demonstrated peripheral nodular enhancement with progressive enhancement on dynamic contrast-enhanced CT and a hemorrhagic change in the central region, which showed hyper intensity on T1-weighted images (T1WI) and hypo intensity on T2-weighted images (T2WI). Microscopically, the cyst was filled with foci of hemorrhage, fibrin, fibrosis, and hemosiderin. Furthermore, a white thrombus was found that corresponded to the central low signal intensity depicted on T2WI. Dilated vascular channels that were immunohistochemically positive for CD31 and CD34 were identified within the cyst. They were consistent with the pathological findings of hemorrhagic vascular adrenal cyst. Radiologists should be aware that the diagnosis of adrenal vascular cyst could be challenging since image findings may resemble hemangiomas, pheochromocytomas, and malignancy.

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☆ Competing interests: None.

☆☆ Patient consent: Written informed consent was obtained from the patient.

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<https://doi.org/10.1016/j.radcr.2021.02.067>

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Fig. 1 – Computed tomography images of the right adrenal tumor. (A) The pre-contrast scan shows high attenuation (60 Hounsfield units) in the right large component (arrow) and slightly lower attenuation (27 Hounsfield units) in the left small component (arrowhead). **(B)** The right component shows peripheral nodular enhancement in the arterial phase. **(C)** The right component shows progressive but incomplete enhancement in the delayed phase. In addition, homogeneous moderate enhancement is observed in the left component. **(D)** Furthermore, macroscopic fat (arrowhead) is observed in the left component.

Introduction

Adrenal collision tumor is a rare tumor defined as the coexistence of two or more adjacent but histologically distinct neoplasms in the adrenal gland without a histological admixture at the interface. Adrenal cysts are rare benign lesions, with an incidence of 0.06% in an autopsy series [1]. Most adrenal cysts are nonfunctional and usually asymptomatic. In 1966, Foster has classically divided adrenal cysts into four types: endothelial cysts (24%–45%), pseudocysts (39%–56%), epithelial cysts (6%–9%), and parasitic cysts (2%–7%) [2]. The etiology of pseudocysts remains unclear; however, immunohistochemical and electron microscopy studies have suggested that both pseudocysts and endothelial cysts are vascular in origin and variants of vascular adrenal cysts [1]. Vascular adrenal cysts account for 80%–84% of all adrenal cysts. They are mostly diagnosed in the fifth to sixth decade of life of patients and more prevalent in women than men (women: men = 3:1) [1]. Here we report a case of a collision of adrenocortical adenoma with myelolipomatous changes and hemorrhagic vascular adrenal cyst.

Case report

A 58-year-old man with a chief complaint of bloody stool was found to have a sigmoid colon cancer on colonoscopy and referred to our hospital for further evaluation and treatment of sigmoid colon cancer. The patient had a medical history of diabetes mellitus and recent myocardial infarction. Two months before referral to our hospital, anticoagulant therapy was started for acute myocardial infarction.

A preoperative contrast-enhanced computed tomographic (CT) colonography revealed sigmoid mural thickening and a large adrenal mass. Otherwise, no distant metastases and no nodal involvement were observed. The right adrenal tumor with a maximal diameter of 5.1 cm demonstrated two densities in the pre-contrast study (Fig. 1A) with different dynamic enhancement patterns. The large right component of the lesion showed high attenuation without haziness of the periadrenal fat or calcification in the pre-contrast CT. This component showed peripheral nodular enhancement in the arterial phase (Fig. 1B) and progressive but incomplete enhancement in the delayed phase (Fig. 1C). The small left component

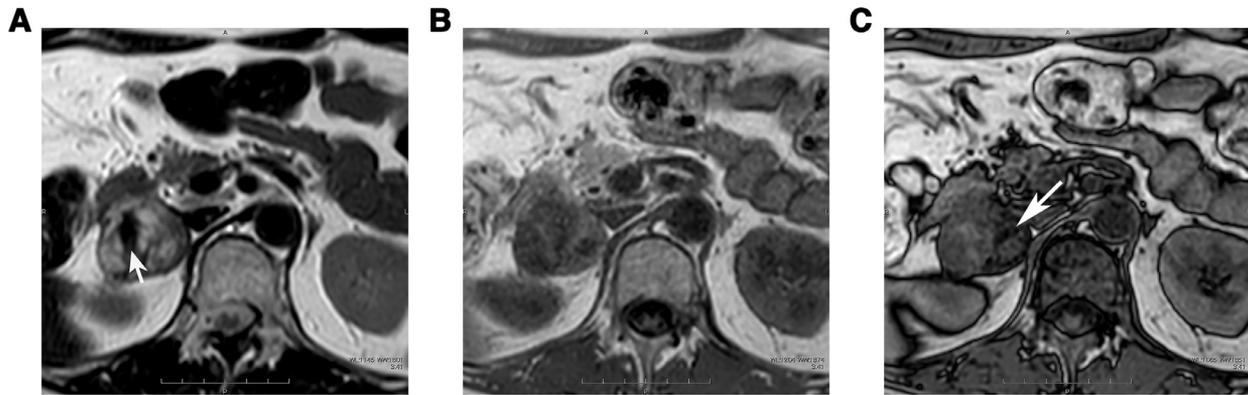


Fig. 2 – Magnetic resonance (MR) images of the right adrenal tumor. (A) Axial T2-weighted MR image shows peripheral high and central low signal intensities (arrow) in the right component of the lesion. (B) Axial T1-weighted in-phase MR image shows an intermediate signal. (C) Axial T1-weighted out-of-phase MR image shows a significant drop in signal intensity (arrow) in the left part of the lesion.

of the tumor showed slightly lower attenuation and homogeneous moderate enhancement without washout (Fig. 1C). In addition, it contained macroscopic fat (Fig. 1D). The peripheral nodular enhancement and progressive enhancement on CT suggested hemangioma; however, no delayed scan after 10–15 min was performed, and the spread of enhancement inside the whole tumor was not confirmed. Adrenal hematoma/hemorrhage, adrenal metastasis, pheochromocytoma, and adrenal cortical carcinoma, which may show central low attenuation and peripheral enhancement, were included in the differential diagnoses.

Magnetic resonance (MR) imaging was performed 23 days after the CT examination. On MR imaging (Fig. 2), the large right component of the tumor showed peripheral high and central low signal intensities on T2-weighted images (T2WI). This is a frequent imaging feature of hemangioma; thus, hemangioma was suspected. The right component partially showed a high intensity on T1-weighted images (T1WI), which suggested hemorrhage or high protein levels. The small left component of the tumor showed a homogeneous signal drop in opposed-phase images; thus, adrenocortical adenoma was suspected. Both components had no restricted diffusion. In addition, I-131 adosterol scintigraphy and I-123 MIBG scintigraphy were performed for further evaluation. I-131 adosterol scintigraphy (Fig. 3) showed apparently high uptake in the mass, which indicated adrenocortical adenoma. The tumor did not show accumulation on I-123 MIBG scintigraphy. The patient was asymptomatic, and the right adrenal tumor was hormonally inactive. Furthermore, the patient has no history of trauma. Based on the aforementioned findings, the adrenal lesion was provisionally diagnosed as a collision tumor of hemangioma and adrenocortical adenoma.

Surgeons and urologists conducted laparoscopic left hemicolectomy for the sigmoid colon cancer and right adrenalectomy for the right adrenal mass because the surgeons were concerned about malignancy in terms of tumor size.

Macroscopic examination of the resected right adrenal mass (Fig. 4A) showed a 42 × 40 × 40 mm mass that con-

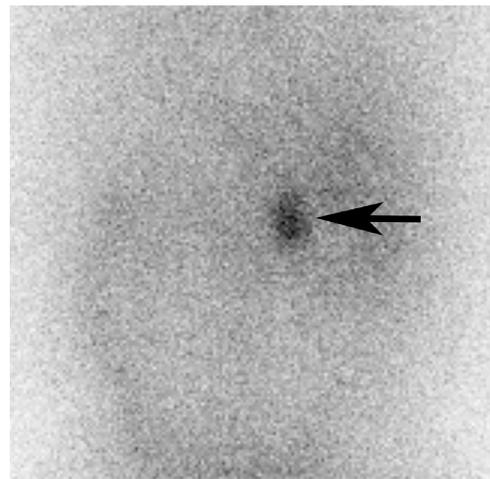


Fig. 3 – I-131 adosterol scintigraphy. The uptake of the right adrenal gland (arrow) is significantly higher than that of the left adrenal gland.

sisted of two components; a golden yellow mass corresponding to the left component of the lesion and a cystic lesion surrounded by a fibrotic wall corresponding to the right component of the lesion. Microscopically, the golden yellow mass was mainly composed of clear cells, admixed with compact cells (Fig. 5A). Furthermore, the tumor had foci of myelolipomatous changes corresponding to the macroscopic fat depicted on the CT and MR images (Fig. 5B). Microscopically, the cyst was filled with foci of hemorrhage, fibrin, fibrosis, and hemosiderin. In addition, a white thrombus was found (Fig. 4B) that corresponded to the central low signal intensity on T2WI. Particularly, dilated vascular channels that were immunohistochemically positive for CD31 and CD34 were identified within the cyst (Fig. 6). Moreover, focal papillary hyperplasia was present. There were no metastatic cells in the resected

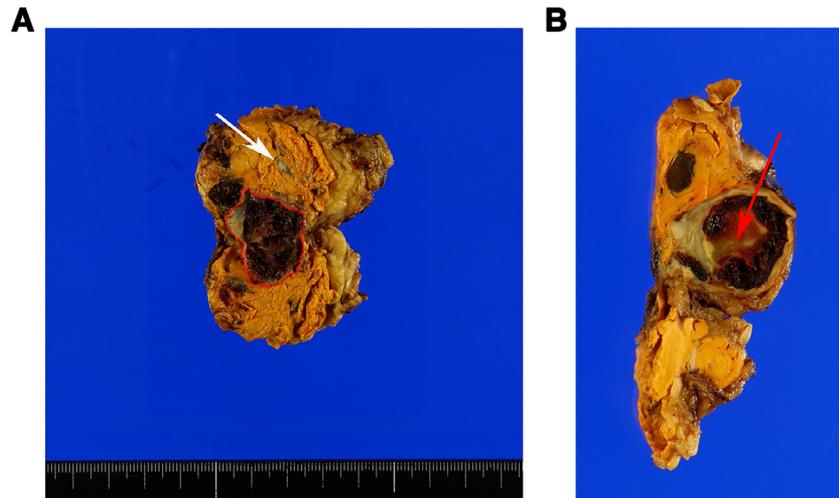


Fig. 4 – A cut surface view of the resection specimen. (A) The adrenal vascular cyst (surrounded by a red line) is observed to be adhered to the adrenocortical adenoma. Macroscopic fat inside the adrenocortical adenoma is observed (arrow). (B) A longitudinal cut surface view of the specimen shown in [Figure 4A](#). A white thrombus (arrow) inside the adrenal vascular cyst is identified, which corresponded to the central low signal intensity depicted on T2-weighted MR images ([Fig. 2A](#)).

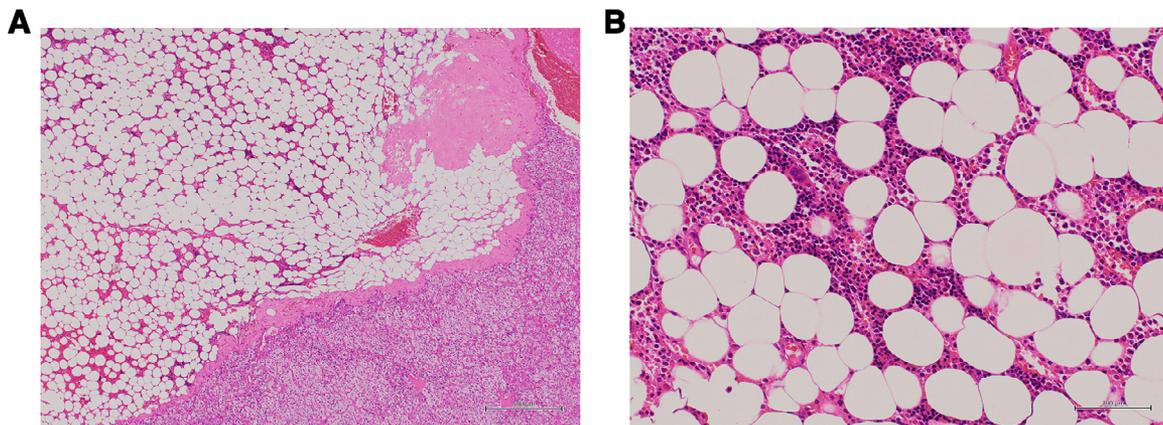


Fig. 5 – Adrenocortical adenoma. (A) Hematoxylin and eosin (H&E) stain, magnification 4x. The right inferior area shows the adrenocortical adenoma. (B) H&E stain, magnification 20x. Myelolipomatous changes are observed as well.

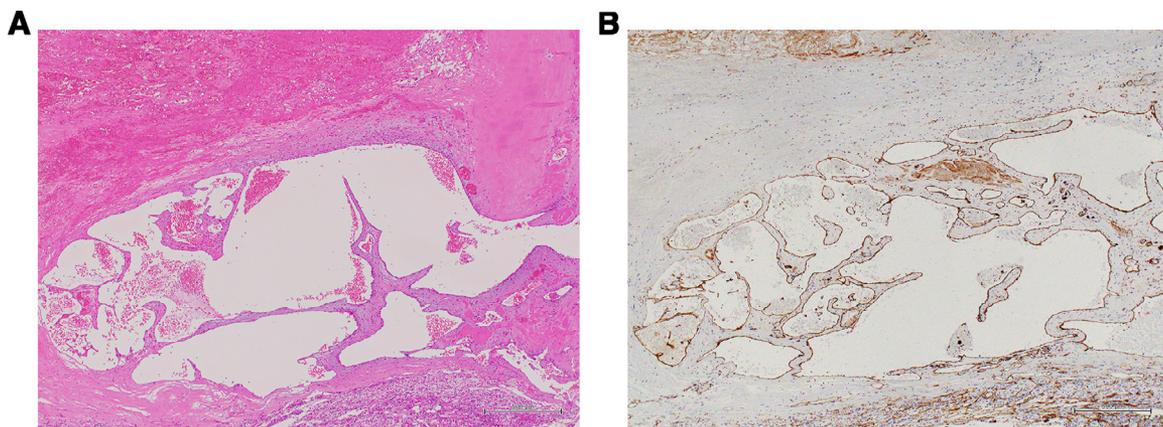


Fig. 6 – Dilated vascular channels within the vascular adrenal cyst. (A) Hematoxylin and eosin stain, magnification 4x. (B) Immunohistochemical staining (magnification 4x) showing positivity for CD31 in the vascular adrenal cyst.

right adrenal mass. Postoperative pathological diagnosis was a collision of adrenocortical adenoma with myelolipomatous changes and hemorrhagic vascular adrenal cyst.

Discussion

We demonstrated a radiologic-pathologic correlation of a collision between adrenocortical adenoma and hemorrhagic vascular adrenal cyst although the vascular cyst is exactly non-neoplastic. Moreover, it mimicked adrenal hemangioma and malignant tumors in the imaging studies. To make this diagnosis, the coincident hemorrhage in the adrenocortical adenoma should be distinguished. Vascular adrenal cysts often contain islands of cytologically normal adrenal cortical cells [3]; however, the foci of adenoma are not recognized in the cystic component like in this case. This suggests that the component of vascular cyst was distinct from adrenocortical adenoma, indicating a collision of two distinct lesions.

The component of adrenocortical adenoma showed the typical image finding: signal loss on opposed-phase T1WI. In addition, they demonstrated macroscopic fat, indicating myelolipoma; however, macroscopic fat representing myelolipomatous changes in the nonfunctioning adenoma was reported [4]. Then, the left component as adrenocortical adenoma is not controversial.

A simple vascular adrenal cyst typically shows well-defined margins and thin walls with water-density contents (less than 20 HU) and no contrast enhancement on CT scans. On MR imaging, it is generally hypointense on T1WI and hyperintense on T2WI due to pure fluid content. When hemorrhage is complicated within the cyst, it demonstrates increased internal attenuation on CT scans and hyperintensity on T1WI [5]. In this case, the hemorrhage in the vascular adrenal cyst showed a similar density and signal, which may have stemmed from the anticoagulant therapy that started two months before CT examination for acute myocardial infarction. The adrenal hemorrhage in the tumor was associated with anticoagulant therapy or stress [6].

The vascular adrenal cyst in this case demonstrated peripheral nodular enhancement with progressive enhancement and central low density/signal region. The enhancement pattern on CT scans was noted in the dilated thin-walled vascular channels and would reflect vascular channels. The central low density/signal region represented the hemorrhagic component identified using histopathology. These are typical imaging features of adrenal hemangioma that would reflect multiple vascular lakes and central modifications [7]; however, dilated thin-walled vascular channels are often recognized in hemorrhagic vascular adrenal cysts [1,3] and pathological similarity was surmised. Kyoda et al. have reported two hemorrhagic vascular cysts that showed a central low area due to the necrosis and hemorrhage with enhancement in the peripheral region [8]. The enhancement pattern may resemble our case, but they only showed the images in the parenchymal phase.

The peripheral and progressive but incomplete enhancement with internal hemorrhage may make diagnosing even benign or malignant lesions difficult on an imaging study.

In fact, the surgeons were concerned about the possibility of malignancy in our case. Peripheral enhancement with a central non-contrast-enhanced area is caused by tumor necrosis and hemorrhage, which could also occur in other diseases and, thus, is not specific for hemorrhagic vascular cyst. In addition, it has been reported in adrenal hematoma/hemorrhage, pheochromocytoma, and malignant tumors, including adrenocortical carcinomas and metastasis [8–12]. Albano et al. have presented a case of breast cancer with adrenal metastasis that demonstrated central low intensity on T2WI due to hemorrhagic components [10].

The management of vascular adrenal cysts remains controversial. As mentioned above, ruling out malignancy in some vascular adrenal cysts is difficult on CT and MR images. Therefore, surgery can be considered for a symptomatic vascular adrenal cyst or lesions larger than 4–5 cm in diameter or when malignancy cannot be ruled out [5]. Continued observation can be considered for asymptomatic small nonfunctioning adrenal vascular cysts.

In conclusion, we report a patient with sigmoid colon cancer who revealed a collision of vascular adrenal cyst and adrenocortical adenoma with myelolipomatous changes. The component of the vascular adrenal cyst demonstrated peripheral nodular enhancement and progressive enhancement, which would reflect dilated vascular channels, with a central hemorrhagic and thrombosed region. These images resembled hemangioma but cannot rule out pheochromocytomas or malignancy.

Acknowledgments

The authors would like to thank Enago (www.enago.jp) for the English language review.

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