[CASE REPORT]

Tumor Embolism as a Cause of Renal Artery Occlusion and Acute Kidney Injury Diagnosed and Treated with Endovascular Intervention in a Patient with Mediastinal Undifferentiated Sarcoma

Kazuya Tsubouchi¹, Ritsu Ibusuki¹, Kenji Makisumi², Hirofumi Okamoto³, Takeshi Iwasaki^{4,5}, Yuki Okamatsu¹, Katsuhiro Inoue¹ and Taishi Harada¹

Abstract:

A 72-year-old man presented with back pain due to a mass in the left posterior mediastinum that had surrounded and partly infiltrated the descending aorta. Mediastinal undifferentiated sarcoma was diagnosed. After the diagnosis, sudden anuria was observed. Contrast-enhanced computed tomography revealed an enhancement defect at the origins of the bilateral renal arteries. He received catheter-directed thrombolysis and was weaned off dialysis. The aspirated artery thrombus contained tumor cells, proving our diagnosis of acute kidney injury secondary to bilateral renal artery tumor embolism. In cancer patients, endovascular intervention may be a useful diagnostic and therapeutic option in cases of acute kidney injury secondary caused by peripheral thromboembolic complications.

Key words: acute renal artery occlusion, arterial tumor embolism, endovascular treatment, acute kidney injury, undifferentiated sarcoma

(Intern Med 60: 1907-1910, 2021) (DOI: 10.2169/internalmedicine.6249-20)

Introduction

Acute renal artery occlusion (RAO) due to thromboembolism is an uncommon cause of acute kidney injury (AKI), occurring in only 1% of AKI cases (1). Although a hypercoagulable state related to cancer is a well-known risk factor for venous thrombosis, acute peripheral arterial occlusion secondary to malignant tumor embolism is a rare event (2). The early diagnosis and treatment of acute RAO leads to better renal outcomes. However, the diagnosis is difficult, as the clinical presentation is often nonspecific (1).

We herein report a rare case of AKI secondary to RAO induced by tumor embolism that was diagnosed and treated successfully with endovascular techniques in a patient with mediastinal undifferentiated sarcoma.

Case Report

A 72-year-old Japanese man with no medical history presented with back pain and a poor appetite. Contrastenhanced computed tomography (CT) showed a solid mass in the left posterior mediastinum that had surrounded and partly infiltrated the descending aorta at the T8-T10 level (Fig. 1A, B). A histopathological examination of the mediastinal tumor obtained by a CT-guided percutaneous needle biopsy revealed proliferation of epithelioid tumor cells with large, vesicular, and oval-to-polygonal nuclei, prominent nucleoli, and abundant eosinophilic cytoplasm. The cells were arranged in a sheet-like pattern, and coagulation necrosis was observed. Immunohistochemically, the tumor cells were positive for vimentin. However, a morphological examina-

Received: September 5, 2020; Accepted: November 29, 2020; Advance Publication by J-STAGE: January 15, 2021

Correspondence to Dr. Kazuya Tsubouchi, tubouchi@med.kyushu-u.ac.jp

¹Department of Respiratory Medicine, Japan Community Health Care Organization Kyushu Hospital, Japan, ²Department of Radiology, Japan Community Health Care Organization Kyushu Hospital, Japan, ³Department of Nephrology, Japan Community Health Care Organization Kyushu Hospital, Japan, ⁴Department of Pathology, Japan Community Health Care Organization Kyushu Hospital, Japan and ⁵Department of Anatomic Pathology, Graduate School of Medical Sciences, Kyushu University, Japan

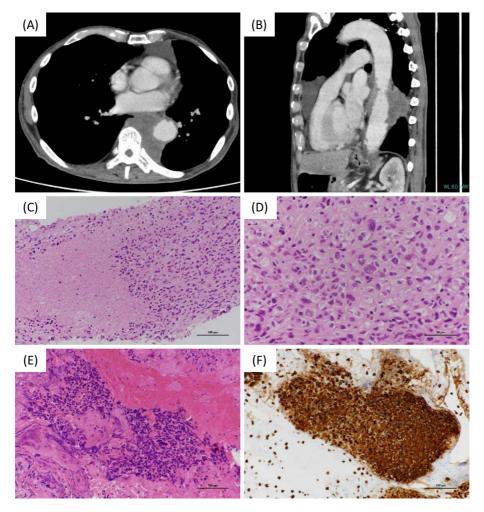


Figure 1. (A) Primary tumor on chest axial computed tomography (CT). (B) Primary tumor on chest sagittal CT showing infiltration of the aortic vessel wall. (C) A mediastinal biopsy specimen containing atypical cells arranged in a sheet-like pattern with coagulation necrosis [Hematoxylin and Eosin (H&E) staining ×20]. (D) A mediastinal biopsy specimen showing proliferating epithelioid tumor cells with marked nuclear pleomorphism and abundant eosinophilic cytoplasm (H&E staining ×40). (E) Samples obtained by endovascular catheter containing atypical cells resembling those at the primary site (H&E staining ×20). (F) Immunohistochemically, the embolic tumor cells were positive for vimentin (×20).

tion using microscopy, immunohistochemistry with vascular markers, and *in situ* hybridization did not reveal any specific differentiation. Therefore, primary posterior mediastinal undifferentiated sarcoma was diagnosed (Fig. 1C, D).

Three weeks after the diagnosis, sudden anuria was observed, accompanied by an increase in creatinine levels from 0.86 to 7.19 mg/dL. He did not present any new symptoms, only the back pain that he had complained of previously. Because his blood tests revealed AKI and hyperkalemia, hemodialysis was immediately performed. Contrastenhanced CT revealed an enhancement defect at the origins of the bilateral renal arteries and the absence of a nephrographic effect; this led to a suspicion of AKI caused by bilateral RAO (Fig. 2A).

The patient received catheter-directed thrombolysis (using urokinase 480,000 units and heparin) in the left renal artery (Fig. 2B, C). The aspirated renal artery thrombus contained

a small cluster of tumor cells. Morphologically and immunohistochemically, the tumor cells resembled those seen in the mediastinal biopsy specimen (Fig. 1E, F), proving our diagnosis of AKI secondary to bilateral renal artery tumor embolism. A left renal artery angiogram showed successful recanalization after catheter-directed thrombolysis. After the endovascular treatment, his creatinine levels decreased to 1.32 mg/dL. The renal dysfunction improved remarkably, allowing the withdrawal of hemodialysis.

However, despite the success of the AKI treatment, the tumor progressed rapidly, and the patient died four months after the diagnosis.

Discussion

Acute RAO secondary to malignant tumor embolism is a very rare event. AKI from other causes is common in pa-

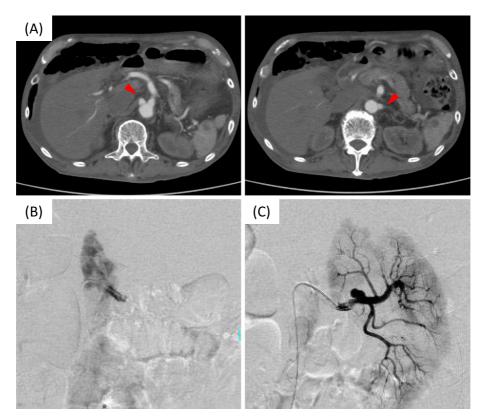


Figure 2. (A) Contrast-enhanced computed tomography revealing occlusion of the bilateral renal arteries at their origins (arrowheads). (B) An angiogram showing occlusion of the left renal artery. (C) A left renal artery angiogram showing successful recanalization after catheter-directed thrombolysis.

tients with cancer and represents a significant event that increases morbidity and mortality (3). AKI in these patients is often directly caused by primary or metastatic cancer and the nephrotoxic effects of anticancer therapies. In our case, RAO due to tumor emboli derived from posterior mediastinal undifferentiated sarcoma led to a very unusual case of AKI, and endovascular techniques were useful for diagnosing arterial tumor emboli and remarkably improved the patient's renal function.

The early diagnosis and treatment of acute RAO leads to better renal outcomes. However, the diagnosis is difficult because the clinical presentation is often nonspecific. Most patients typically experience the acute onset of constant abdominal, flank, or back pain (1). Arteriography is the diagnostic modality for identifying an arterial occlusion. Furthermore, it can distinguish between embolism and thrombosis (4). In our patient, although the back pain from infiltration of the vertebral body made the diagnosis of RAO more difficult, acute RAO was diagnosed based on contrastenhanced CT and renal arteriography findings. In addition to the diagnosis of RAO, the cytological specimens obtained using an endovascular catheter confirmed renal artery tumor thrombosis.

Tumor emboli are also difficult to confirm, except by autopsy or operative findings (5). A recent report suggested that pulmonary microvascular cytology may be useful in the diagnosis of pulmonary tumor thrombotic microangiopathy,

suggesting that cytology using samples obtained from an endovascular catheter may be helpful in the detection of tumor emboli (6). Anticoagulation, surgical thrombectomy, systemic thrombolysis, and catheter-directed thrombolysis have been performed to treat acute RAO. Although endovascular treatment remains controversial for the management of acute RAO, it is a safe modality and should be considered as not only a diagnostic procedure but also a therapeutic option (7). In addition, treatment at the primary site may be effective for preventing recurrence of tumor emboli.

Arterial embolization of tumor tissue is a rare complication. In a review of 3 large groups of patients, symptomatic arterial embolization of macroscopic tumor fragments was detected in only 2 out of 840 patients (8). Previous reports have described arterial tumor emboli from primary intracardiac tumors or cancer invasion of a pulmonary vein (4, 9). Primary malignant tumors of the aorta (PMTAs), which are extremely rare and highly aggressive, typically exhibit peripheral thromboembolic complications. PMTAs are categorized as intimal or mural (10). The intimal type forms intraluminal polyps or extends along the lumen, causing peripheral emboli or aortic obstruction, while the mural type extends extramurally to para-aortic tissues and lymph nodes (11). The primary malignancy in the present case was aggressive and caused peripheral arterial occlusion similar to that of the intimal aortic type. Although this tumor partly infiltrated the aortic vessel wall, it mainly extended extramurally to the thoracic vertebrae, similar to the primary mural aortic type. Although the lesion was immunohistochemically negative for vascular markers, the biological behavior of this tumor was similar to that of both intimal and mural aortic sarcoma.

Cancer patients presenting with AKI are often managed conservatively. The early diagnosis and treatment should be attempted in order to salvage the kidney. We believe that endovascular intervention may be a useful diagnostic and therapeutic option in cases of AKI caused by peripheral thromboembolic complications such as tumor embolism.

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

References

- Koivuviita N, Tertti R, Heiro M, Manner I, Metsärinne K. Thromboembolism as a cause of renal artery occlusion and acute kidney injury: the recovery of kidney function after two weeks. Case Reports Nephrol Dial 4: 82-87, 2014.
- Morasch MD, Shanik GD. Tumor embolus: a case report and review of the literature. Ann Vasc Surg 17: 210-213, 2003.
- Rosner MH, Perazella MA. Acute kidney injury in the patient with cancer. Kidney Res Clin Pract 38: 295-308, 2019.
- 4. Schreffler SM, Paolo WF, Kloss BT. Spontaneous showering of tu-

- mor emboli in a patient with advanced primary lung cancer: a case report. Int J Emerg Med 5: 1, 2012.
- Hottenrott G, Mentzel T, Peters A, Schröder A, Katenkamp D. Intravascular ("intimal") epithelioid angiosarcoma: clinicopathological and immunohistochemical analysis of three cases. Virchows Arch 435: 473-478, 1999.
- Ishiguro T, Takayanagi N, Baba Y, et al. Case series of pulmonary tumor embolism and intravascular lymphoma: evaluation of the usefulness of pulmonary microvascular cytology. Intern Med 55: 2679-2684, 2016.
- Silverberg D, Menes T, Rimon U, Salomon O, Halak M. Acute renal artery occlusion: presentation, treatment, and outcome. J Vasc Surg 64: 1026-1032, 2016.
- **8.** Morasch MD, Shanik GD. Tumor embolus: a case report and review of the literature. Ann Vasc Surg **17**: 210-213, 2003.
- Grazziotin MU, Turnipseed WD. Arterial tumor embolism caused by metastatic melanoma: case report and literature review. J Vasc Surg 36: 191-193, 2002.
- 10. Thalheimer A, Fein M, Geissinger E, Franke S. Intimal angiosarcoma of the aorta: report of a case and review of the literature. J Vasc Surg 40: 548-553, 2004.
- Auchoybur ML, Su C, Chen X, Huang F. Intimal angiosarcoma of the aorta: single case report and effective surgical management. J Curr Surg 7: 64-68, 2017.

The Internal Medicine is an Open Access journal 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/).

© 2021 The Japanese Society of Internal Medicine *Intern Med 60: 1907-1910, 2021*