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Case Report

A case of bilateral adrenal infarction with preserved adrenal function diagnosed by dual-energy computed tomography☆,☆☆

Misaki Shoji, MD[a], *Tomoko Hyodo, MD*[a,*], *Yosuke Nagasawa, MD*[b], *Masahiro Nishihara, MD*[b], *Hideki Nakamura, MD, PhD*[b], *Yoshihiro Nakamura, MD*[c], *Hiroki Kobayashi, MD*[c], *Masanori Abe, MD*[c], *Masahiro Okada, MD*[a]

[a] *Department of Radiology, Nihon University School of Medicine, 30-1 Ohyaguchi-kami-cho, Itabashi-ku, Tokyo 173-8610, Japan*
[b] *Division of Hematology and Rheumatology, Department of Internal Medicine, Nihon University School of Medicine, 30-1 Ohyaguchi-kami-cho, Itabashi-ku, Tokyo 173-8610, Japan*
[c] *Division of Nephrology, Hypertension and Endocrinology, Department of Internal Medicine, Nihon University School of Medicine, 30-1 Ohyaguchi-kami-cho, Itabashi-ku, Tokyo 173-8610, Japan*

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ABSTRACT

We report a case wherein adrenal function remained preserved despite bilateral adrenal infarction, as evidenced by dual-energy computed tomography (DECT) iodine density images. The patient was a 37-year-old man with a history of antiphospholipid syndrome concomitant with systemic lupus erythematosus. The patient underwent contrast-enhanced DECT, which revealed bilateral adrenal infarction. Laboratory tests revealed preserved adrenal function. On the iodine density images, the infarcted and noninfarcted areas in the adrenal glands were visually different. The volume of the non-infarcted area was 8.9 mL, which was 41% of the total adrenal volume. DECT may be a useful complementary tool for assessing the preservation of adrenal function.

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* Corresponding author.
E-mail address: neneth@m.ehime-u.ac.jp (T. Hyodo).

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Introduction

Adrenal infarction is defined as the injury or necrosis of the adrenal glands due to inadequate blood supply. It typically arises from thrombophilia or a hypercoagulable state such as antiphospholipid antibody syndrome, pregnancy, or coronavirus disease 2019 [1]. The occurrence of this condition in bilateral adrenal glands is rare, with only a few cases reported in the literature.

Dual-energy computed tomography (DECT) imaging technology has significantly advanced in recent years, leading to an expansion of its applications. DECT offers quantitative expression in addition to conventional computed tomography (CT) attenuation values. Several studies have demonstrated the utility of iodine density, fat fraction imaging, and virtual unenhanced imaging for differentiating adrenal adenomas in other tumors [2–4]. However, to the best of our knowledge, there are no reports on the application of DECT for the diagnosis of adrenal infarction.

DECT not only facilitates the identification of the infarction site but also may potentially be used to calculate the iodine content within the infarcted and non-ischemic areas separately, thereby aiding in the prediction of functional prognosis. Studies have demonstrated the utility of iodine density imaging in differentiating between healthy and ischemic or necrotic myocardium [5] as well as distinguishing between frequent blood-brain-barrier disruption and rare intracerebral hemorrhage immediately following endovascular recanalization therapy [6].

Herein, we report a case in which adrenal function was preserved despite bilateral adrenal infarctions, as evidenced by iodine density imaging on DECT.

Case report

A 37-year-old man presented to our emergency department with abdominal pain and nausea. His medical history included systemic lupus erythematosus (SLE), antiphospholipid syndrome (APS), and ulcerative colitis. He had a history of brain stroke 8 months prior and pulmonary embolism/deep vein thrombosis 4 years before visiting our hospital. On admission, his height, weight, and body mass index were 177 cm, 66 kg, and 21.1 kg/m^2, respectively.

Laboratory investigations revealed no hyponatremia (serum sodium level, 139 mEq/L) or hypoglycemia (serum blood glucose level, 149 mg/dL). Activated partial thromboplastin time was prolonged (73.7 s). Blood D-dimer level remained within normal limits (<1.0 µg/mL). The prothrombin-time/international normalized ratio was 1.49 s. Rapid adrenocorticotropic hormone stimulation test showed no abnormalities (serum cortisol levels were 21.3 µg/dL at 30 minutes and 25.5 µg/dL at 60 minutes). Based on these findings, adrenal insufficiency was ruled out.

Unenhanced CT showed hypoattenuation enlargement of the left adrenal gland and mild surrounding fat stranding, indicative of adrenal infarction. On the day after the onset of symptoms, contrast-enhanced CT of the chest and abdomen

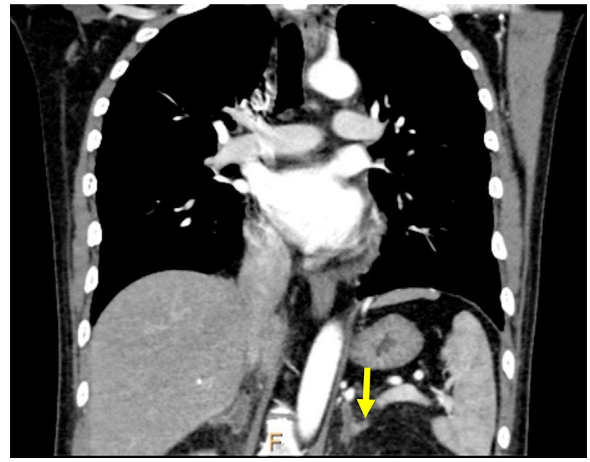

Fig. 1 – Conventional coronal computed tomography (CT) image (120 kV) in the arterial phase. Partial contrast enhancement is observed in the left adrenal gland (arrow). Contrast enhancement was not significant in other parts of the bilateral adrenal glands.

was performed using dual-layer DECT (IQon Elite Spectral CT scanner, Philips Healthcare, Best, Netherlands) to evaluate blood flow in the adrenal glands and rule out acute pulmonary embolism. The arterial phase scan was performed 30 s after injecting the 560 mgI/mL contrast injection (2.5 mL/s, 100 mL total). Postcontrast conventional 120 kV CT images in the arterial phase revealed a filling defect and contrast-enhanced areas in the bilateral adrenal glands, along with adrenal enlargement and mild surrounding fat stranding (Fig. 1). Unenhanced CT revealed no adrenal hemorrhage. The sigmoid colon and rectum exhibited mild wall thickening, but this finding remained unchanged on a CT performed 1 month earlier, and the patient was not considered to have progressive ulcerative colitis. Dual-energy analyses using a workstation (Intellispace Portal Release 11.1; Philips Healthcare) revealed that the iodine density images in the arterial phase demonstrated heterogeneous contrast enhancement in the bilateral adrenal glands, suggesting the coexistence of components with preserved blood flow, ischemia, or infarction (Fig. 2). Given the bimodal distribution of the iodine density histogram in the adrenal gland region, binarization with a threshold of 1.0 mgI/cm^3 was performed to quantify the amount and volume of iodine in each component with and without significant blood flow (hereafter referred to as noninfarcted and infarcted areas, respectively) (Fig. 3). The iodine density values in the right and left adrenal gland areas were 0.38 and 0.46 mg/cm^3 for infarcted areas, and 1.42 and 1.69 mg/cm^3 for noninfarcted areas, respectively. The volumes of the right and left adrenal glands were 12.83 and 8.70 cm^3, respectively. Consequently, the total iodine amounts in the right and left adrenal gland areas were 9.62 and 9.31 mg, respectively. The iodine content in the noninfarcted area was 13.80 mg. The volume of the non-infarcted area was 8.9 mL, representing 41% of the total adrenal gland volume. (Fig. 3, Table 1).

The patient was initially treated with 10,000 units/day of heparin for 10 days, followed by 7000 units/day for 2 days. Abdominal pain and nausea improved 5 days after onset.

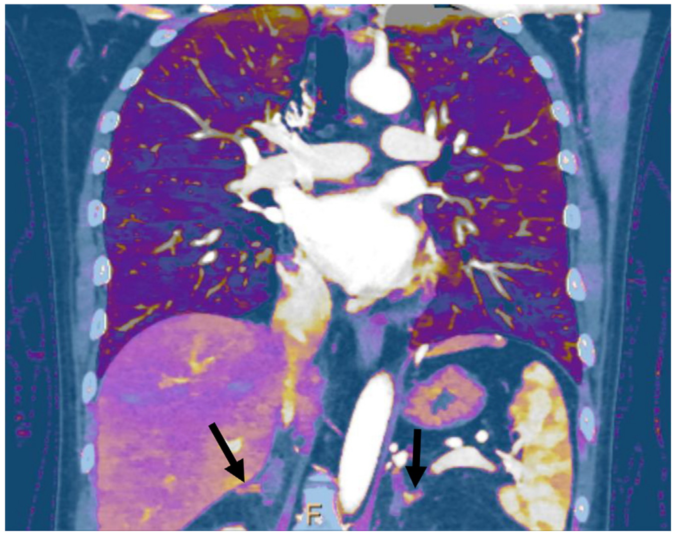

Fig. 2 – A color-coded iodine density image overlaid on a conventional CT image (120 kV) of a coronal section of the same cross-section as Figure 1. The areas with (arrows) and without contrast enhancement in the bilateral adrenal glands are delineated.

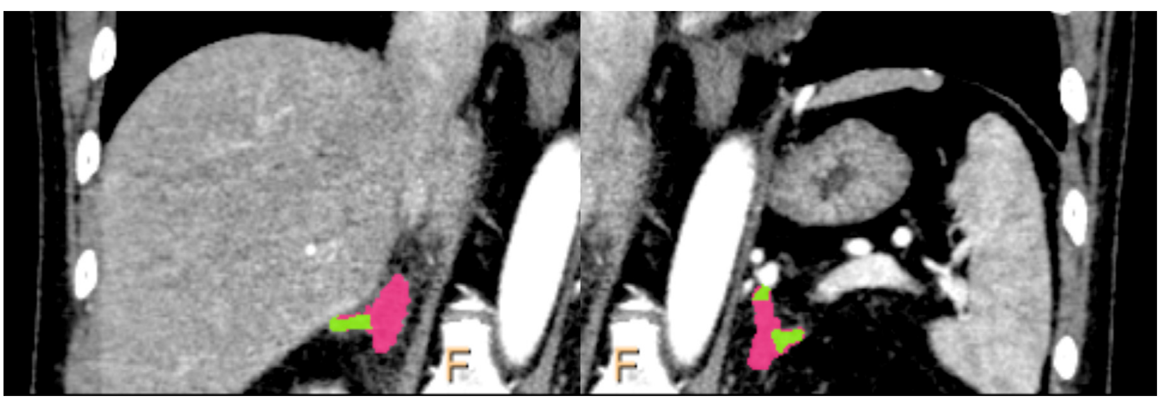

Fig. 3 – A representative coronal slice of the volume was analyzed. Quantitative analysis was performed separately for infarcted (pink) and noninfarcted (green) areas using an iodine density of 1.0 mgI/cm^3. The right and left adrenal glands were analyzed separately and combined into a composite figure.

Table 1 – Summary of volume, iodine density and computed tomography attenuation values for the bilateral adrenal glands at onset and day 17.

Adrenal gland	Site	Volume (cm^3)	% Volume (out of full tissue)	Iodine density (mg/mL)	CT attenuation at 120 kV (HU)
Onset					
Right	0.00 < Iodine density (mg/mL) < 1.00	8.22	64	0.38 ± 0.31	29.7 ± 2.53
	1.00 < Iodine density (mg/mL) < 2.49	4.61	36	1.42 ± 0.31	60.3 ± 20.7
	Full tissue	12.83	100	0.75 ± 0.59	40.7± 27.9
Left	0.00 < Iodine density (mg/mL) < 1.00	4.41	51	0.46 ± 0.28	19.0 ± 31.2
	1.00 < Iodine density (mg/mL) < 7.59	4.29	49	1.69 ± 0.45	66.9 ± 25.6
	Full tissue	8.70	100	1.07 ± 0.72	42.6 ± 37.3
Day17					
Right	0.00 < Iodine density (mg/mL) < 1.00	6.15	75	0.36 ± 0.33	5.1 ± 41.8
	1.00 < Iodine density (mg/mL) < 2.18	2.04	25	1.21 ± 0.17	58.2 ± 20.3
	Full tissue	8.19	100	0.57 ± 0.47	18.3 ± 44.1
Left	0.00 < Iodine density (mg/mL) < 1.00	2.25	51	0.51 ± 0.31	14.0 ± 31.9
	1.00 < Iodine density (mg/mL) < 2.77	2.14	49	1.45 ± 0.28	57.4±22.5
	Full tissue	4.39	100	0.97 ± 0.56	35.1 ±35.2

Note. –Unless otherwise indicated, data are mean ± standard deviation.

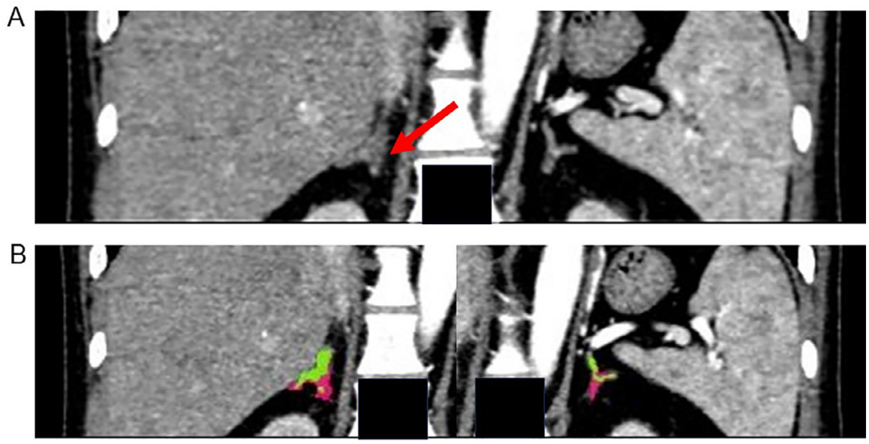

Fig. 4 – (A) Conventional coronal computed tomography (CT) image (120 kV) in arterial phase 17 days after the onset, showing no expansion of infarcted areas (arrow). (B) A representative coronal slice of the volume was analyzed. We analyzed each right and left adrenal gland using the same method as described in Figure 3, and delineated them with pink and green coloring, respectively.

However, he developed heparin-induced thrombocytopenia and required treatment with argatroban hydrate and warfarin. Owing to argatroban-induced rash, the patient was transitioned to warfarin monotherapy. His adrenal function remained stable throughout hospitalization. On day 17 after symptom onset, follow-up contrast-enhanced CT imaging revealed no expansion of the nonenhanced areas within both adrenal glands, and regions maintaining contrast enhancement were noted, indicating successful control of the adrenal infarction (Fig. 4A and B). The same analysis as that for the initial CT showed that the iodine content in the noninfarcted area of the bilateral adrenal glands was 5.57 mg. The volume of the noninfarcted area was 4.18 mL, representing 33% of the total adrenal volume (Table 1).

Discussion

APS is an autoimmune disorder characterized by recurrent venous and arterial thrombotic events associated with the persistent presence of antibodies directed against phospholipid-protein complexes. Approximately half of patients with APS have SLE. In this case, the observed hypoadrenalism was likely linked to adrenal vein thrombosis and subsequent hemorrhagic infarction, a common pathogenic mechanism in the APS [7].

The vascular anatomy of adrenal glands plays a crucial role in adrenal infarction. The unique vascular structure of the adrenals, characterized by a rich arterial supply but limited venous drainage through a single vein, creates an environment conducive to hemorrhagic infarction [8]. Patients with APS and other hypercoagulable states are prone to thrombosis, and the unique vascular anatomy of the adrenal gland further predisposes these individuals to infarction and subsequent hemorrhage [7,9].

Up to 36% of patients with primary APS present with adrenal insufficiency as the first manifestation of the disease [10]. The prevalence of adrenal infarction is not well defined.

Bilateral adrenal infarctions often complicate adrenal insufficiency; however, in the present case, the patient did not exhibit adrenal insufficiency. This suggests that some degree of blood flow was preserved within the adrenal parenchyma, potentially preventing the complications of adrenal infarction.

Adrenal infarction is diagnosed using contrast-enhanced CT to assess adrenal gland enlargement, fat tissue density, and the presence of contrast enhancement defects. Table 2 compares the clinical characteristics and CT findings of 10 cases of adrenal infarction, encompassing 9 previous reports and the case currently being reported. Among these reported cases, bilateral adrenal infarction occurred in 8 cases, and adrenal function was preserved in 3 cases. There were no other cases in which the infarcted areas were quantitatively evaluated. Additionally, Magnetic resonance imaging(MRI) was performed in the cases reported by Agarwal et al. [11] and Riddell et al. [12]. MRI is useful for evaluating adrenal infarction, especially in pregnant patients, owing to its lack of radiation exposure and the ability to detect infarction via diffusion-weighted imaging without the need for contrast agents. However, challenges persist in differentiating between hemorrhage and infarction, as in conventional CT, along with longer scan durations and potential contraindications for metallic devices.

DECT has been shown for the visualization of infarcted areas in other organs, such as the myocardium and intestine [13,14]. In this case, iodine density imaging provided a clearer depiction of the adrenal gland components with better-preserved blood flow than conventional CT imaging.

Furthermore, DECT allows the quantification of iodine per pixel value. Iodine quantification in the adrenal area may be used to predict prognosis and guide treatment decisions for complicated adrenal insufficiency, such as weaning off of corticosteroids [15]. To the best of our knowledge, there is currently no definitive information regarding the amount of blood flow required to prevent adrenal insufficiency. In the present case, the volume of the noninfarcted area was 8.9 mL, representing 41% of the total adrenal gland volume. Further studies are needed to elucidate the relationship between preserved blood flow and adrenal gland function.

Table 2 – Summary of 10 case reports.							
Age (y) sex	Source	Symptom at presentation	comorbidity	D dimer (μg/mL)	Imaging findings	Location	Adrenal function
81 male	Hoshino et al. [1]	severe bilateral backache	bladder and prostate cancer; colon cancer; MDS/MPN-U	2.8	bilateral adrenal hypertrophy, non-contrast-enhancing areas, and adjacent fatty inflammatory changes	bilateral	preserved
63 male	Presotto et al. [8]	abrupt pain to his right flank	APS	unknown	bilateral adrenal enlargement consistent with gland hemorrhage	bilateral	not preserved
21 female	Agarwal et al. [11]	right-sided abdominal pain, nausea and emesis	asthma; pregnancy	unknown	CT: hypoenhancing enlarged right adrenal gland with mild stranding MRI: abnormal right adrenal gland with increased T2 signal with surrounding oedema	right	preserved
42 male	Riddell et al. [12]	left flank pain	APS; deep vein thrombosis	unknown	CT: enlargement of the bilateral adrenal glands with mild inflammatory changes in the adjacent fat MRI: Axial T1-weighted gadolinium-enhanced image shows lack of contrast enhancement in both adrenal glands .	bilateral	not preserved
47 female	Mavridis et al. [16]	intense, penetrating pain in the epigastrium and left hemithorax	APS;deep vein thrombosis	1.8	bilateral adrenal gland enlargement with hemorrhage after the omset	bilateral	not preserved

(continued on next page)

Table 2 (*continued*)

Age (y) sex	Source	Symptom at presentation	comorbidity	D dimer (µg/mL)	Imaging findings	Location	Adrenal function
14 male	Inam et al. [17]	Abdominal pain, weight loss, vomiting and fever	APS	unknown	Bilaterally enlarged hypodense adrenals with no contrast enhancement	bilateral	not preserved
34 male	Sapkota et al. [18]	abdominal pain,nausea and emesis	hypertension; metham-phetamine abuse	unknown	thickening of the left adrenal gland with inflammatory stranding of the surrounding fat and internal hypoenhancement of the gland	left	unknown
53 female	Yonezaki et al. [19]	epigastric pain, nausea, and vomiting	pulmonary tuberculosis; TAFRO syndrome	unknown	decreased blood flow in the bilateral adrenal glands	bilateral	preserved
70 female	Kumar et al. [20]	fever, left-sided chest pain, cough and mild dyspnoea, fatigue, abdominal pain, vomiting and diarrhea	COVID-19 infection; Hypertension hypercholes-terolemia	unknown	enlarged, diffusely hypoattenuating bilateral adrenal glands demonstrating poor enhancement and surrounding fat stranding	bilateral	not preserved
37 male	present case	abdominal pain and nausea	SLE, APS, ulcerative colitis, cerebral infarction, pulmonary embolism, deep vein thrombosis	<1.0	filling defect and contrast-enhanced areas in the bilateral adrenal glands, along with adrenal enlargement and mild surrounding fat stranding	bilateral	preserved

Note. APS, Antiphospholipid syndrome; COVID-19, coronavirus disease; MDS/MPN-U, myelodysplastic/myeloproliferative neoplasm unclassifiable; SLE, systemic lupus erythematosus; TAFRO, thrombocytopenia, anasarca, fever, reticulin myelofibrosis/renal failure, organomegaly.

Conclusion

Here, we report a case of bilateral adrenal infarction with preserved adrenal function. The components of the preserved adrenal blood flow were visually and quantitatively evaluated using DECT.

Patient consent

The written informed consent was obtained from the patient for being included in the case and it is available upon request.

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