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Case Report

An anomalous left adrenal vein draining into both the left renal vein and inferior vena cava: A case report *,**

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ABSTRACT

A male patient in his forties was diagnosed with primary aldosteronism following blood tests. The patient requested surgical treatment and was transferred to our department for adrenal vein sampling. Preoperative contrast-enhanced computed tomography (CT) revealed that the left adrenal vein (LAV) did not form a common trunk with the left inferior phrenic vein, and instead drained into both the left renal vein (LRV) and inferior vena cava (IVC) after the bifurcation. Angiography from the LAV showed a branch draining into the IVC. Left-sided sampling was performed. Aldosterone levels were elevated at all 3 sites. Because the sampling results were bilaterally positive, the patient was contraindicated for surgery. There are no reports of an anomalous LAV draining into both the LRV and IVC, making this case extremely unique. Regarding the LAV sampling site, it has been reported that aldosterone levels are higher in the common trunk than in the LAV. Therefore, we increased the number of blood sampling sites. In adrenal vein sampling, we often focus on the anatomy of the right adrenal vein because of difficulties in accessing to it. However, anomalies of the LAV may also occur. Therefore, it is important to determine the anatomy of both adrenal veins using preoperative contrast-enhanced CT to plan an appropriate sampling strategy.

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Introduction

Adrenal vein sampling is a diagnostic technique used to determine whether aldosterone overproduction occurs in uni-

lateral or bilateral adrenal glands. Primary aldosteronism is treated by unilateral adrenalectomy if aldosterone overproduction is unilateral, or by drug therapy if it is bilateral. Therefore, it is clinically important to perform adrenal vein sampling if a patient requests surgical treatment [1].

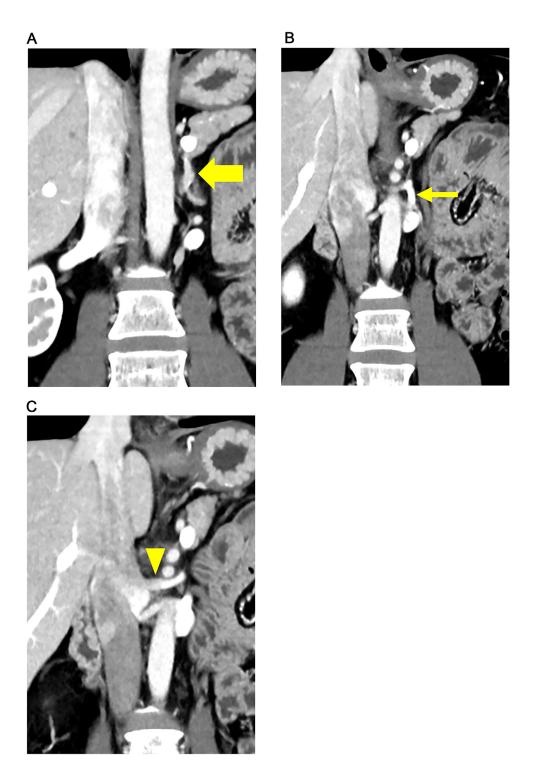


Fig. 1 – Coronal contrast-enhanced computed tomography images of the abdomen. The left adrenal vein (LAV) drains into both the left renal vein (LRV) and inferior vena cava (IVC). (A) LAV (thick arrow). (B) Branch draining into the LRV (thin arrow). (C) Branch draining into the IVC (arrowhead).

When performing adrenal venous sampling to diagnose primary aldosteronism, it is recommended to collect blood samples from at least 3 positions: the bilateral adrenal veins and the inferior vena cava (IVC) caudal to the left renal vein (LRV) [1]. Sampling of the left adrenal vein (LAV) has a success rate of >90%, compared with a success rate of 75%-95% for sampling of the right adrenal vein (RAV) [2]. This is because the LAV generally joins the left inferior phrenic vein to form a long, thick common trunk that flows into the LRV, making it easier to insert a catheter into the LAV [3]. However, some rare cases have been reported in which the LAV did not form a common trunk with the inferior phrenic vein [4] or the LAV flowed directly into the IVC without joining the LRV [5]. To our knowledge, this is the first case in which the LAV did not form a common trunk with the inferior phrenic vein and drained into both the LRV and the IVC.

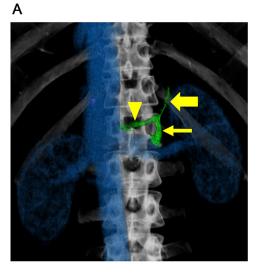
Case report

The patient was a male in his forties who was taking 4 10 mg tablets of Nifendipine (Towa Pharmaceutical Co., Ltd., Osaka, Japan) daily for hypertension and who had systolic blood pressure as high as 150 mmHg.

Abdominal computed tomography (CT) revealed a small nodule in the left adrenal gland. A diagnosis of primary aldosteronism was made following blood tests. The patient requested surgical treatment and was transferred to our department for adrenal vein sampling. Preoperative contrastenhanced CT revealed that the LAV did not form a common trunk with the left inferior phrenic vein, and instead drained into both the LRV and IVC after the bifurcation (Fig. 1).

In our department, blood sampling is usually performed at the following 4 positions: (1) the peripheral IVC, (2) the LAV, (3) the common trunk of the LAV and the left inferior phrenic vein, and (4) the RAV. However, in this case, because the left inferior phrenic vein was not observed by CT, we decided to collect blood samples at a total of 5 positions, including the following 2 sub-positions: (3A) the branch draining into the LRV and (3B) the branch draining into the IVC (Fig. 2).

A 5 Fr long sheath was inserted into the bilateral femoral veins. Sampling was performed using 5 Fr adrenal vein catheters (Medikit, Tokyo, Japan), high-flow microcatheters (2.6/2.8 Fr Masters 125 cm Asahi, Tokyo, Japan), and microguidewires (CHIKAI, Asahi, Tokyo, Japan) just before and 15 min after adrenocorticotropic hormone loading. A 5 Fr catheter was inserted into the LAV. Angiography from the LAV showed an anomalous branch draining into the IVC (Fig. 3). Left-sided sampling was performed at the distal portion of the LAV, the branch draining into the LRV, and the branch draining into the IVC using a microcatheter. On the left side, aldosterone levels were elevated at all 3 sampling positions (2, 3A, and 3B), in the descending order of positions 2 (28,596 pg/mL), 3A (17,747 pg/mL), and 3B (3,846 pg/mL). The aldosterone/cortisol ratios were higher in the descending order of positions 2 (21.96), 3B (18.49), and 3A (16.76). Because the sampling results were bilaterally positive, the patient was contraindicated for surgery.



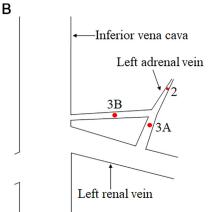


Fig. 2 – Sampling positions of the left arterial vein (LAV). (A) Preoperative 3-dimensional contrast-enhanced computed tomographic image showing the 3 sampling positions (thick arrow, thin arrow and arrowhead). An anomalous LAV (green) draining into both the left renal vein and inferior vena cava (blue). (B) Schematic of the LAV showing the 3 sampling positions (red dots).

Discussion

Generally, the RAV drains into the right posterior lateral side of the IVC, and the LAV drains into the LRV. Anomalies are common in the RAV, which often drains into the accessory hepatic vein [6]. In addition, insertion of a catheter into the RAV has a lower procedural success rate than insertion into the LAV [2]. Therefore, we often focus on the anatomy of the RAV before sampling. However, rare anomalies of the LAV have also been reported, generally accompanied by anomalies of the LRV and/or IVC. For example, the LAV drains into the anterior renal vein in patients with a duplicate LRV [7]. The LAV may also drain directly into the IVC in patients with a left IVC [8]. LAV anomalies without other anatomic abnormalities, which are extremely rare, include cases in which the LAV drains into the gonadal or lumbar vein [9] and the LAV does not join the

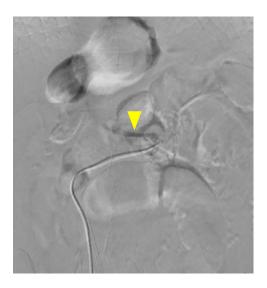


Fig. 3 – Angiography of the left adrenal vein showing an anomalous branch draining into the inferior vena cava (arrowhead).

main trunk of the LRV but drains directly into the IVC [5]. In the present case, the LAV drained into both the LRV and IVC, without anomalies of the renal veins or IVC. To our knowledge, there are no reports of an anomalous LAV draining into both the LRV and IVC.

The recommended sampling positions of the LAV vary with the guidelines. The 2014 expert consensus statement advocates blood sampling in the LAV on just the cranial side of the entrance of the left inferior phrenic vein [10]. In comparison, the 2021 Endocrine Society guidelines do not recommend a specific sampling position [10]. Regarding LAV sampling positions, it was reported that aldosterone levels are lower in the common trunk than in the LAV because of dilution by blood from the left inferior phrenic vein. However, in some cases in which the LAV and the left inferior phrenic vein do not form a common trunk but instead flow independently into the LRV, aldosterone levels are higher in the left inferior phrenic vein than in the LAV. Therefore, sampling in the common trunk is considered to be important [11]. Aldosterone levels may differ between the proximal and distal portions of the LAV due to the influence of the superficial or fine tributary veins [12]. Based on the above findings, we hypothesized that the aldosterone levels in the branches draining into the LRV and IVC were higher than those in the LAV in our case. Therefore, sampling was performed at 3 positions of the peripheral portion of the LAV, and the 2 branches draining into the LRV and IVC. This revealed elevated aldosterone levels at all 3 positions. Therefore, the sampling positions should be carefully determined in patients with LAV anomalies.

The aldosterone level at the branch draining into the IVC (3B) was lower than those at the LAV (2) and the branch draining into the LRV (3A). The branch draining into the IVC formed a near-right angle to the IVC. Therefore, it is possible that the blood flow from the IVC refluxed into the branch and diluted the aldosterone level at position 3B. In patients with an

anomalous branch draining into the IVC, the direction of blood flow in the branch should be confirmed by angiography.

Conclusion

When performing adrenal vein sampling, we observed a rare case in which the LAV did not form a common trunk with the left inferior phrenic vein, and instead drained into both the LRV and IVC after the bifurcation. Therefore, it is important to understand the exact anatomy of both adrenal veins using preoperative contrast-enhanced CT to plan an appropriate sampling strategy.

Patient consent

Informed consent was obtained from the patient for publication of this case report.

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