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Case Report

Large intra-abdominal venous malformations in associated with inferior vena cava aneurysm ☆,☆☆

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ABSTRACT

Intra-abdominal venous malformations and inferior vena cava aneurysms are rare and difficult to diagnose because of their nonspecific clinical symptoms. These vascular anomalies are important entities due to the risk of thrombosis or rupture. According to the classification of International Society for the Study of Vascular Anomalies, venous malformations are classified as low-flow vascular anomalies, showing absence of arterial and early venous enhancement and slow gradual filling with contrast on delayed venous imaging. Phleboliths related to thrombosis and calcifications, are the key finding of venous malformations. In this article, we report an exceptional case of large intra-abdominal venous malformations in associated with an inferior vena cava aneurysm.

Introduction

Venous malformations are low-flow vascular malformations, which appear to derive from abnormal growth of the venous network, leading to dilated and dysfunctional veins [1,2]. These lesions are commonly found in the head, neck and extremities [1,3]. Intra-abdominal venous malformations are rare. IVC aneurysms are also quite rare. In the Montero-Baker's review, only 53 cases were identified [4]. Based on their anatomic characteristics and the presence of associated venous obstruction, inferior vena cava (IVC) aneurysms were classified into 4 types [5]. In addition, Thompson and Lindenauer [6] classified IVC aneurysms into 3 groups as congenital, acquired, and arteriovenous fistula dependent. Our article aimed to illustrate an extremely rare case of IVC aneurysm, which was accompanied by large intra-abdominal venous malformations.

Case report

A 36-year-old woman presented to hospital with epigastric pain for 1 day. She was alert and oriented with stable vital signs. Abdominal examination demonstrated a soft nontender abdomen without organomegaly. She had a history of gastritis and a caesarean delivery at local hospital 5 years ago. Other than that, no history of trauma or other surgery was

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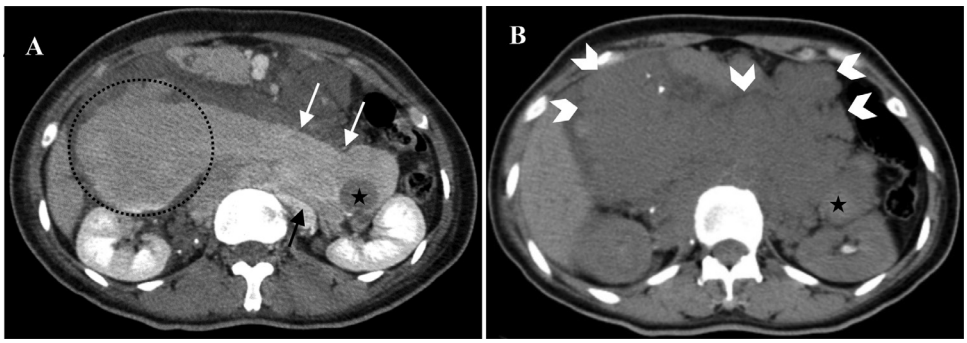

Fig. 1 – Large IVC aneurysm (A, dash circle) associated with dilated left renal veins (A, arrows) and left renal vein thrombosis (B, star). In addition, multifocal, lobulated, infiltrative, hypoattenuating lesions were also detected (B, arrowheads). IVC, inferior vena cava.

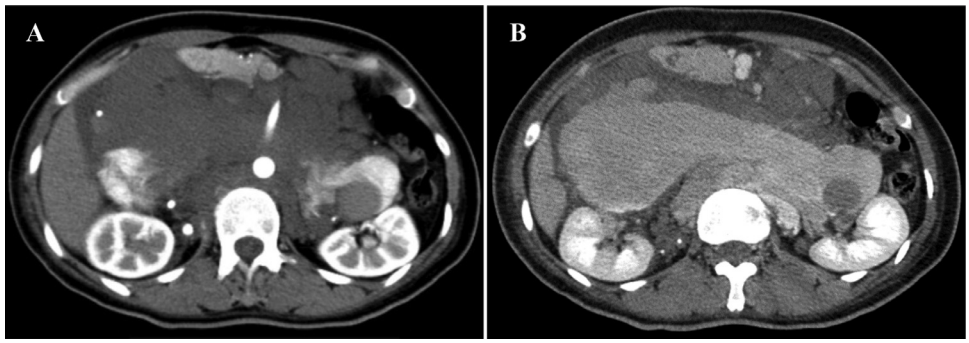

Fig. 2 – The above lesions show inadequate enhancement on arterial phase (A) and gradually filling in with contrast on later phase (B).

noted. Her family history is unremarkable. Laboratory studies showed a normal complete blood count. Coagulation tests result were prothrombin time 13.5 seconds, activated partial thromboplastin time 35.7 seconds and international normalized ratio 1.2. Elevated D-dimer level was noted (28,089 ng/mL). Echocardiography showed normal left ventricular systolic function with ejection fraction 75%. Abdominal ultrasound showed abnormal dilatation of the IVC, with turbulent intrinsic flow.

She underwent an abdominal computed tomography (CT) protocol with intravenous contrast enhancement (iohexol, Omnipaque 350 mgI/mL). Data was reconstructed with a slice thickness of 1.25 mm and analysed using Picture archiving and communication system (PACS) workstation. The abdominal CT displayed a large IVC aneurysm, in associated with dilated bilateral renal veins. There was a round, hyperdense structure causing intraluminal filling defect of the left renal vein proximal to the left renal hilum, approximately 2.6 cm in diameter, consistent with a thrombus (Fig. 1). In addition, large intraperitoneal and retroperitoneal multifocal lesions were also detected. These lesions were infiltrative, lobulated, hypoattenuating, gradually filled with contrast on multiphase contrast-enhanced images (Figs. 1, and 2). Scattered internal calcifications were also detected, consistent with multiple phleboliths of low-flow vascular malformations (Fig. 3). A digital subtraction angiography (DSA) and abdominal magnetic resonance imaging (MRI) were also performed.

The angiography procedure was achieved by a 5F pigtail catheter (Merit, 100 cm), advanced though a 6F sheath using a 0.035-inch guidewire (Terumo, 150 cm). The DSA images showed no abnormalities of the abdominal aorta or signs suggestive of an arteriovenous fistula. A large IVC aneurysm and dilated renal veins were clearly showed again (Fig. 4). The vascular malformations were not displayed, even on delayed DSA images, suggesting its low-flow dynamic feature.

The MRI protocol included triplanar T2-weighted images, volumetric interpolated breath-hold examination (VIBE) using 2-point Dixon fat-water separation (VIBE-Dixon) T1-weighted images (T1W), precontrast and multiphase postcontrast axial T1W images. The venous malformations were obviously showed on MRI study, with multiple hypointense phleboliths and gradual filling in with contrast on delayed phase images (Fig. 5). The patient opted for conservative treatment with oral anticoagulation, proton pump inhibitor, antacid, and analgesic. She was discharged after her epigastric pain resolved and advised to take a close follow-up.

Discussion

Venous malformations are low-flow vascular lesions according to the classification of International Society for the Study of Vascular Anomalies, based on their flow characteristics [7]. Although venous malformations are quite common, most of

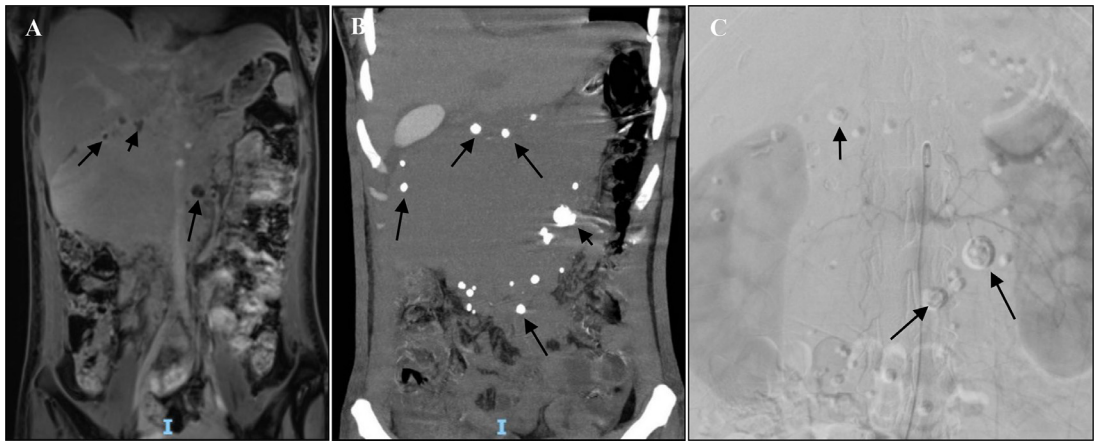

Fig. 3 – Phleboliths, the key finding of venous malformations, were demonstrated as multiple hypointense nodules on MRI, calcified nodules on CT and DSA images. CT, computed tomography; DSA, digital subtraction angiography; MRI, magnetic resonance imaging.

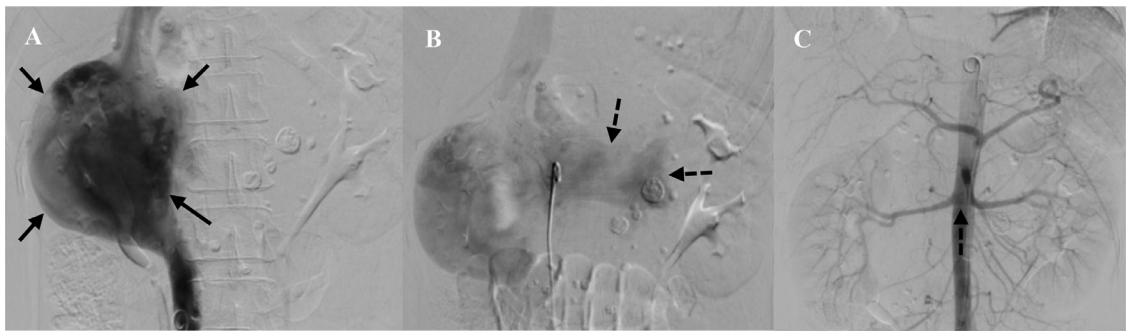

Fig. 4 – DSA images clearly demonstrated a large IVC aneurysm and dilated left renal vein (A, B). The abdominal aorta was normal, no sign suggestive of an arteriovenous fistula was showed (C). DSA, digital subtraction angiography; IVC, inferior vena cava.

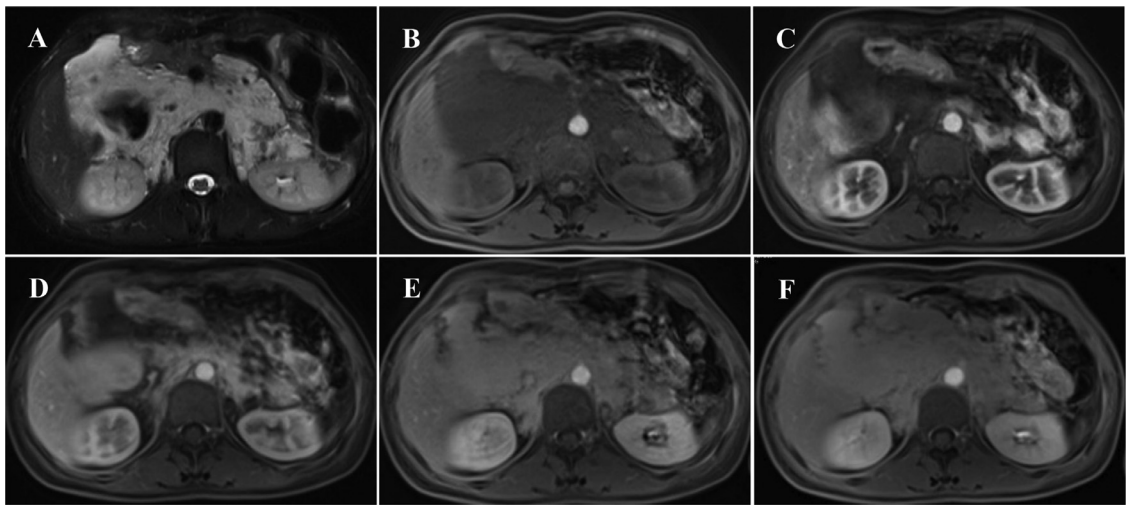

Fig. 5 – On MRI study, large, infiltrated lesions were detected within the intraperitoneal and retroperitoneal space, which is hyperintense on T2W (A). On precontrast T1W, postcontrast arterial phase, venous phase, 3-min and 10-min images (B-F), these lesions were gradually filling in with Gd. MRI, magnetic resonance imaging; T1W, T1-weighted images; T2W, T2-weighted images.

the lesions located on the head, neck or extremities [1,3]. These lesions may be focal, multifocal, or infiltrative. Venous malformations are attributable to aberrant development of the venous system, creating dilated and dysfunctional veins. Based on their connection with the venous system, venous malformations are further classified as (1) type I, isolated lesions without venous connection; (2) type II, lesions that drain into normal veins; (3) type III, lesions with drainage into dysplastic veins; and (4) type IV, lesions composed of venous ectasia [8]. Because of the low-flow feature, multiphase contrast-enhanced MRI becomes the optimal modality for evaluating venous malformations, providing valuable information about its characteristics, extension, as well as demonstrating associated thrombus. Phleboliths are the key finding, which are typically shown as multiple hypointense nodules on T1W and T2-weighted images images.

Venous aneurysms are defined as persistent focal venous dilatation twice the normal diameter [9]. In particular, IVC aneurysms are rare and the majority of cases were asymptomatic and diagnosed by chance [10]. The etiology of IVC aneurysm is not fully understood. Congenital weakness of venous wall was noted as one of the suggested causes [11]. Acquired aneurysms could evolve from trauma, arteriovenous fistula, IVC stenosis or obstruction. According to Thompson and Lindenauer [6], IVC aneurysms were classified as congenital, acquired, or arteriovenous fistula dependent. Based on their anatomic relation to renal veins and the presence of associated venous obstruction, Gradman and Steinberg [5] grouped IVC aneurysms as 4 types, including: type I, aneurysms involving the suprahepatic IVC with no venous obstruction; type II, aneurysms associated with obstruction of the IVC above or below the hepatic veins; type III, infrarenal aneurysms without venous obstruction; type IV, miscellaneous. In our case, the aneurysm mainly involved the inferior renal segment of IVC without any signs of an arteriovenous fistula detected on DSA images. Dilated bilateral renal veins associated with an intraluminal thrombus were also noted.

The choice of treatment method for venous malformations and IVC aneurysms should be considered appropriately on a case-by-case basis. For isolated, well circumscribed without visible venous drainage or draining to normal veins (type I and II according to Dubois/Puig classification), sclerotherapy offers a convenient and effective modality of treatment [8]. However, diffuse infiltrative venous malformations with drainage into ectatic dysplastic veins or composed of venous ectasia are difficult to treat and associated with more complications. Due to the rarity of IVC aneurysms, there is currently no consensus on treatment strategies. Surgical treatment such as ligation, resection with end to end anastomosis or synthetic graft interposition could be considered in symptomatic and low-risk asymptomatic cases [4,9]. In our case, a large infrarenal IVC aneurysm was surrounded by diffuse infiltrative venous malformations, making the surgery particularly difficult and risky. Conservative treatment with warfarin therapy and observation with serial imaging studies is an acceptable treatment option.

The association of large, diffuse, infiltrative intra-abdominal venous malformations and IVC aneurysm as in our case is exceptionally rare. The initial CT scan performed well for detecting intra-abdominal vascular malformations, aneurysm, as well as excluding other causes of acute abdominal pain. DSA remains gold standard imaging modality for accessing aneurysms and associated arteriovenous fistula, if present. MRI has advantages in evaluating low-flow malformations in general and venous malformations, in particular. On contrast-enhanced MRI, typical venous malformations show inadequate enhancement on arterial phase and gradually filling in with contrast on later phases. Multiple internal phleboliths are well demonstrated on the CT and MRI studies.

Conclusion

Intra-abdominal venous malformations, IVC aneurysms are rare. These vascular anomalies may be presented with non-specific symptoms, make it challenging to diagnose. Imaging modalities play an essential role in detecting, assessing the lesion's extension and its correlation with adjacent structures, which make a significant contribution to patient's management and follow-up.

Patient consent

Informed consent for patient information to be published in this article was obtained.

Ethical statement

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

REFERENCES

[1] Loose D. Surgical management of venous malformations. Phlebology 2007;22(6):276–82.
[2] Hussein A, Malguria N. Imaging of vascular malformations. Radiol Clin 2020;58(4):815–30.
[3] Olivieri B, White CL, Restrepo R, McKeon B, Karakas SP, Lee EY, et al. Low-flow vascular malformation pitfalls: from clinical examination to practical imaging evaluation—part 2, venous malformation mimickers. Am J Roentgenol 2016;206(5):952–62.
[4] Montero-Baker MF, Branco BC, Leon LL Jr, Labropoulos N, Echeverria A, Mills JL Sr, et al. Management of inferior vena cava aneurysm. J Cardiovasc Surg (Torino) 2015;56(5):769–74.
[5] Gradman WS, Steinberg F. Aneurysm of the inferior vena cava: case report and review of the literature. Ann Vasc Surg 1993;7(4):347–53.
[6] Thompson NW, Lindenauer SM. Central venous aneurysms and arteriovenous fistulas. Ann Surg 1969;170(5):852.
[7] ISSVA, *Classification of vascular anomalies© 2018 International Society for the study of vascular anomalies*. 2018.
[8] Puig S, Casati B, Staudenherz A, Paya K, et al. Vascular low-flow malformations in children: current concepts for classification, diagnosis and therapy. Eur J Radiol 2005;53(1):35–45.

[9] Gusani R, Shukla R, Kothari S, Bhatt R, Patel J, et al. Inferior vena cava aneurysm presenting as deep vein thrombosis– a case report. Int J Surg Case Rep 2016;29:123–5.
[10] Atalar M. Aneurysm of the inferior vena cava: imaging findings. Austin J Radiol 2016;3:1053.
[11] Mookadam F, Rowley VB, Emani UR, Al-Harthi MS, Baxter CM, Wilansky S, et al. Aneurysmal dilatation of the inferior vena cava. Echocardiography 2011;28(8):833–42.