Eur J Vasc Endovasc Surg 15, 365–368 (1998)

CASE REPORT

Idiopathic Aneurysm of the Inferior Vena Cava as a Cause of Massive Penile Bleeding

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Introduction

Aneurysms of the inferior vena cava (IVC) are very rare and only 16 cases have been reported[1–4] previously. Mostly these aneurysms are saccular, secondary to proximal stenosis or obstruction and association with congenital anomalies of the IVC. Thrombosis of an IVC aneurysm with severe oedema of the lower extremities and fatal pulmonary emboli are well described complications. We report for the first time the case of a 19-year-old man with an idiopathic fusiform aneurysm of the IVC, complicated by urethral varicose veins and penile bleeding. Surgical resection became mandatory because of recurrent, life-threatening, massive penile bleeding.

Case Report

A 19-year-old man, practising power training, was admitted a first time with massive penile bleeding and hypovolemic shock. The haemoglobin value was only 4 g/dl. Spontaneous, recurrent, urethral blood loss had occurred during the preceding year. Cystoscopy on admission demonstrated bleeding urethral varices which were coagulated, and the anaemia was corrected by blood transfusion. A few months later recurrent severe bleeding occurred and the patient was transferred to our institution for further investigation. Physical examination was normal. Cystoscopy confirmed the presence of urethral varices and the pelvic CT scan showed dilatated caval and hypogastric veins (Fig. 1a). Cavography showed a fusiform aneurysm of the IVC, beginning 3 cm below the level of the renal veins and extending to the left hypogastric vein, with extensively dilated pelvic veins (Fig. 1b). No associated congenital anomalies of the IVC or other causes of obstruction to bloodflow were found. Arteriovenous malformations and communications were excluded by selective intra-arterial arteriography. The diagnosis of an idiopathic fusiform aneurysm of the IVC was made.

In the first instance thrombo-occlusion of the left internal hypogastric vein was attempted by placing a coil percutanously in this vein. However, thrombosis did not take place and further bleeding occurred a few weeks later. Faced with a life threatening condition, surgery was undertaken. The aneurysm of the IVC, the renal, hypogastric and spermatic veins were exposed by a transverse abdominal incision and left visceral rotation (Fig. 2). Dissection of the venous aneurysm was carried out with great care because of the very thin wall of the caval and the iliac veins. A possible anterior wall resection to reduce its lumen was rejected because of the impossibility of making secure sutures. Therefore the dissection was cautiously continued proximally and distally until healthy venous wall was reached. The aneurysm was resected including the right common hypogastric vein, the left external and internal hypogastric, and the pelvic veins as distally as technically possible. The venous continuity was restored with a 16/8 mm PTFE bifurcated prothesis (Stretch Goretex®) implanted between the infrarenal IVC, the right common hypogastric and the left external hypogastric vein. In an effort to avoid

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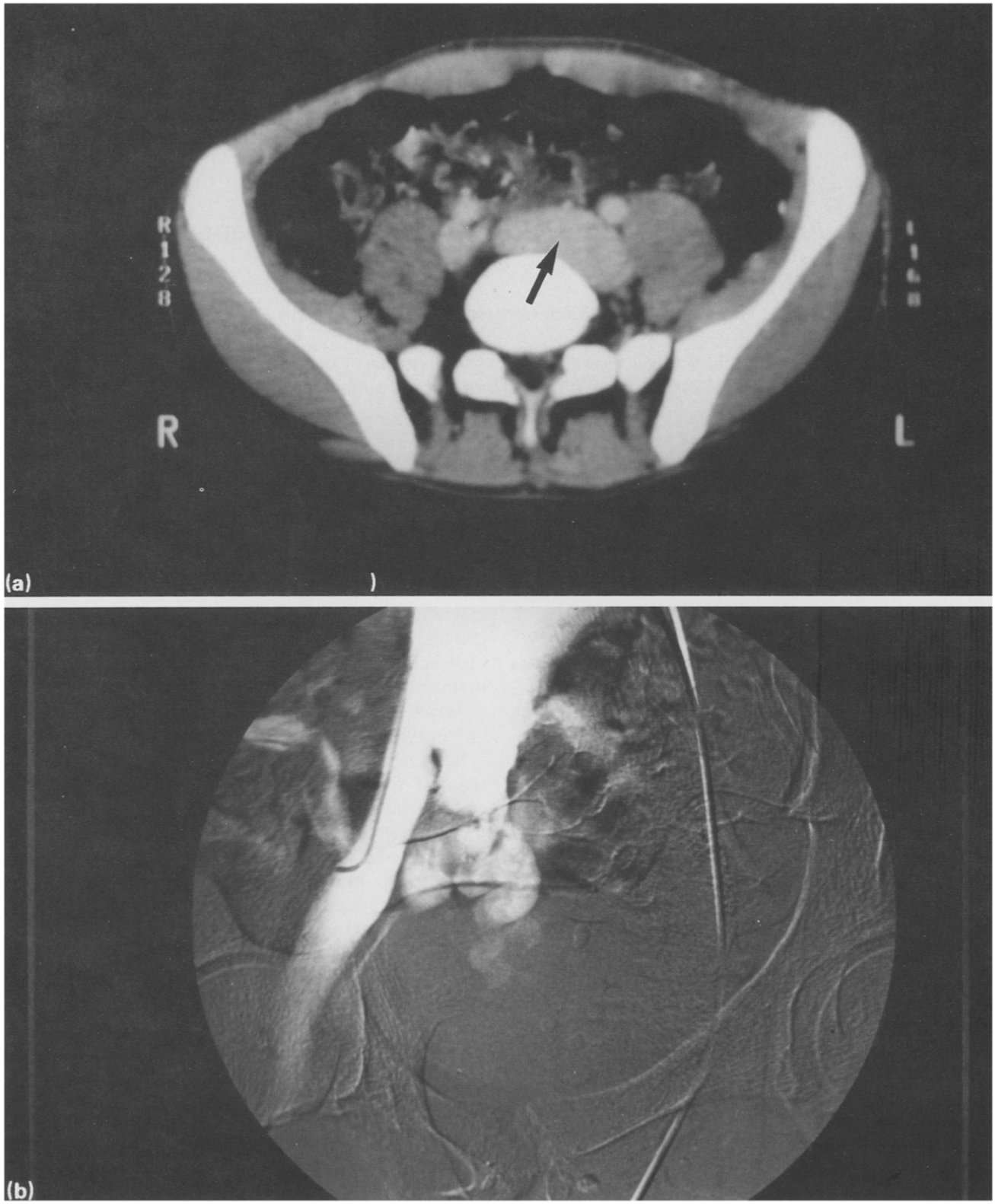

Figs 1a and b. Pelvic CT scan showing dilatated left hypogastric vein (a) and cavography demonstrating the aneurysm of the IVC extending to the left hypogastric vein with large varicosities (b).

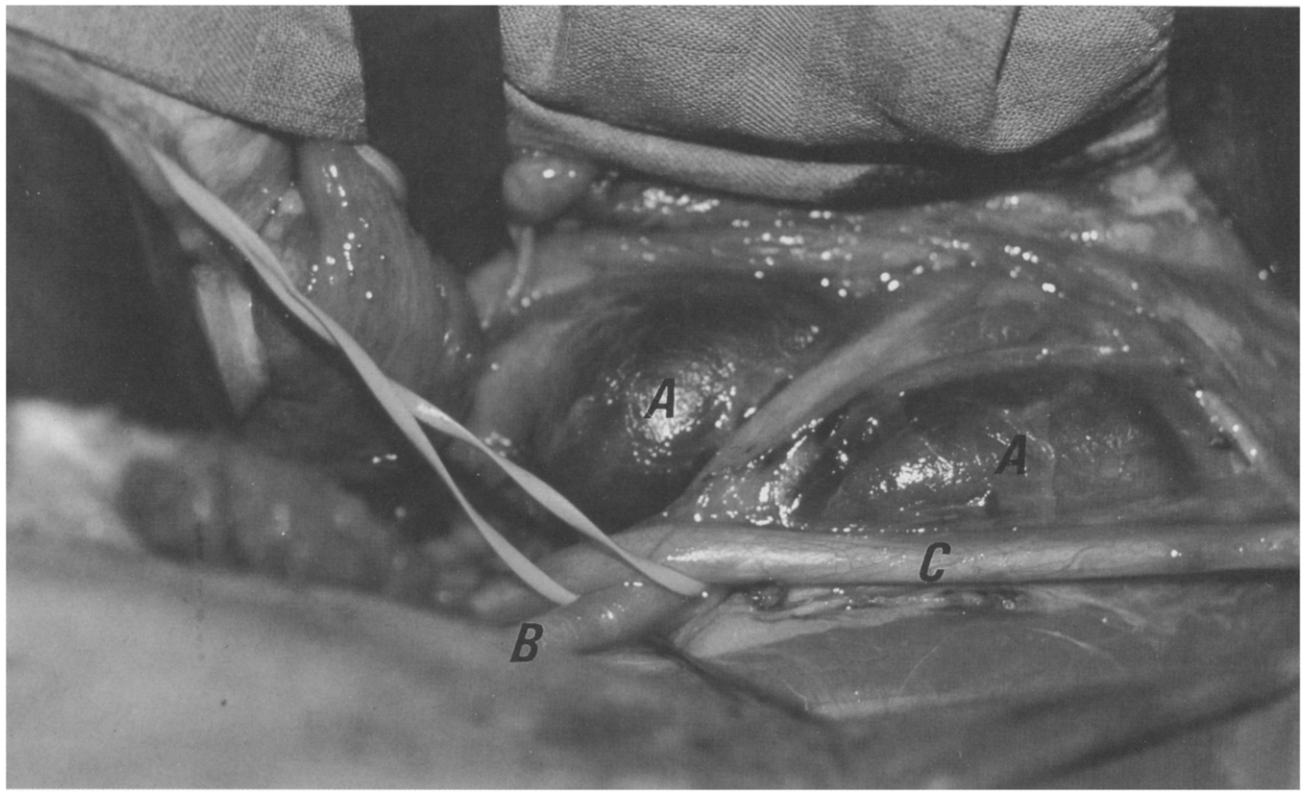

Fig. 2. Peroperative view after left visceral rotation, exposure of the aneurysm of IVC and left hypogastric vein (A), the right iliac artery (B) and the right ureter (C).

graft thrombosis, acenocoumarol and aspirin (100 mg daily) were administered orally from the first operative day, while elastic stocking was prescribed. The perioperative period was uneventful and the patient was discharged on the eighth postoperative day. However 1 month after surgery a deep vein thrombosis of the left leg occured, treated by surgical thrombectomy of the femoral and hypogastric vein. Optimisation of anticoagulant therapy was needed because the patient had taken his medication irregularly. At the present time, 7 months after the operation, the patient is asymptomatic, without complaints or clinical signs of penile bleeding or lower extremeity thrombosis.

Histopatological examination revealed a fusiform venous aneurysm consisting of a three-layered wall structure with a decrease of muscle fibres and an increase of fibrous tissue in the tunica media.

Discussion

In this patient a very unusual cause of urethral varicose veins was found. Mostly urethral dilated veins are caused by venous pelvic congestion during pregnancy.[5] We think that in our patient the enlargement of the aneurysm of the IVC to the internal hypogastric and pelvic veins and the repeated exaggerated Valsalva manoeuvres during power training led to repetitive high venous pressure, resulting in the development of urethral varicose veins and subsequent bleeding. The histopathological examination suggests a congenital cause of this aneurysm, especially as obstruction to flow, as well as collateral circulation were absent. Gradman *et al.*[1] has proposed a classification of these congenital venous aneurysms into four types consistent with their anatomic and embryologic characteristics: suprahepatic caval aneurysm without venous obstruction (type 1), aneurysms associated with suprahepatic or infrahepatic caval interruption (type 2), aneurysm in the infrerenal IVC without associated congenital anomalies (type 3), and aneurysm of iliac vein with left-sided IVC (type 4). The case we describe might correspond to the type 3 class proposed by these authors.

To our best knowledge only 16 cases of IVC aneurysm have been reported until now. Eight of these were associated with congenital anatomic anomalies (tricuspid stenosis, left-sided IVC, supra- and infrahepatic interruption and segmental agenesis of IVC). Eight others were of a probably congenital origin

without other anomalies. Only two of the 16 reported aneurysms were fusiform. In contrast to our case these two were located supra- and perihepatically. Nine patients were asymtomatic, six presented with IVC thrombosis and severe oedema of the lower extremities, and one died from fatal pulmonary emboli. Five saccular aneurysms were resected, while the others were treated conservatively.

Uncomplicated aneurysms might best be followed non-invasively, but when complications occur surgery may become necessary in selected cases. In our patient, life threatening, recurrent massive bleeding made resection and prothestic replacement necessary, in spite of his young age.

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Accepted 19 August 1997