Giant venous aneurysm associated with hypogastric arteriovenous malformation

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Venous aneurysms are extremely rare. They may be congenital or acquired in origin and occasionally related to arteriovenous communications. A 58-year-old man complained of dull left lower quadrant pain and constipation. On physical examination a soft deep mass was palpated. Ultrasonogram and CT scan revealed a cystic formation in the pelvic cavity. Angiograms disclosed an arteriovenous malformation (AVM) at the pelvic floor draining into a large cavity. The patient was successfully managed by intraoperative selective embolization of the AVM and partial resection of a 10.6 × 8 × 6.7 cm venous aneurysm. The histopathologic studies of the wall confirmed a venous structure. Venous dilatation has been reported in high flow vein grafts, blood access V fistulas and rarely, proximal to traumatic AV fistulas of the lower extremities. The etiology of the present case is probably congenital, being tc the best of our knowledge, the first case affecting the hypogastric territory, reported in the English literature. (*Ann Vasc Surg*, 1986, *1*, 143-146).

KEY-WORDS : Venous aneurysm. — Arteriovenous malformation.

Although clinically recognized by Osler [1] early this century, true venous aneurysms are seldom reported. They have been found isolated or associated with arteriovenous communications. Excepting those related to trauma, the etiology remains unknown. Clinical presentation depends on its location and associated complications due to local compression or pulmonary embolization. The present report describes our experience with a giant venous aneurysm of the hypogastric territory associated with a pelvic arteriovenous malformation (AVM).

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CASE REPORT

A 58 year-old Caucasian male presented with a 3 months history of dull pain of the left lower quadrant, occasionally colicky in character, radiating to the left lumbar area. Associated complaints included impotence and recent onset of constipation. He denied a history of palpitation, dyspnea or trauma. Sixteen years before he had passed a urinary stone. The physical examination disclosed a soft mass deep in the left lower quadrant. Peripheral pulses were normal and without bruits. On the rectal examination the mass was well defined, painless, slightly pulsatile and a thrill could be felt. An intravenous pyelogram (Fig. 1) showed extrinsic compression of the bladder and displacement of the terminal left ureter without dilatation. The ultrasonogram (Fig. 2a) revealed a cystic mass 10.6 × 6.7 × 8 cm occupying most of the pelvic cavity. A CT scan (Fig. 2b) confirmed the vascular nature of the cyst and its relation to the surrounding structures. An abdominal aortogram (Fig. 3a) showed mild dilatation and tortuosity of the left common iliac and hypogastric arteries. Selective left iliac catheterization (Fig. 3b) demonstrated an AVM at the pelvic floor fed by hypertrophied branches originating distal to the superior gluteal artery. Late films showed filling of an aneurysm enclosed by the arterial vessels. Selective films of the right iliac artery failed to show connections with the AVM. Emptying of the aneurysm could not be traced.

The patient underwent elective laparotomy. The pelvic anatomy was distorted by the tortuous left iliac vessels and the aneurysm which displaced the bladder and compressed the rectum.

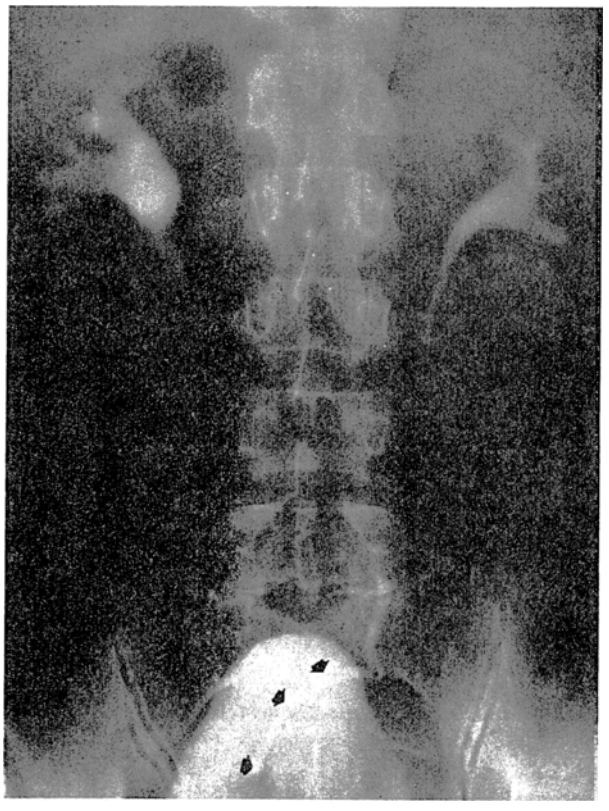
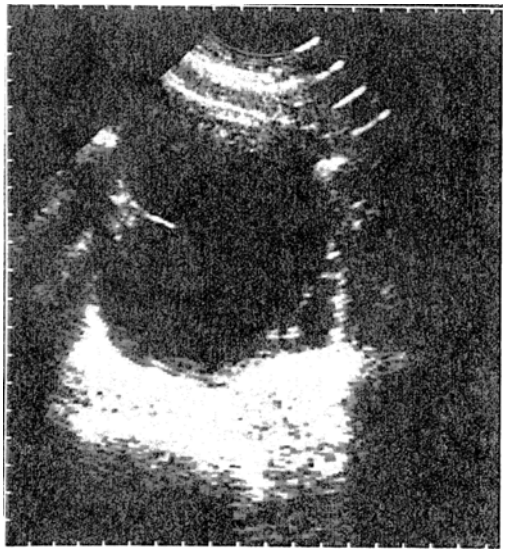


Fig. 1. — Intravenous pyelogram showing displacement of the bladder and left ureter by a mass located to the left in the pelvic cavity.



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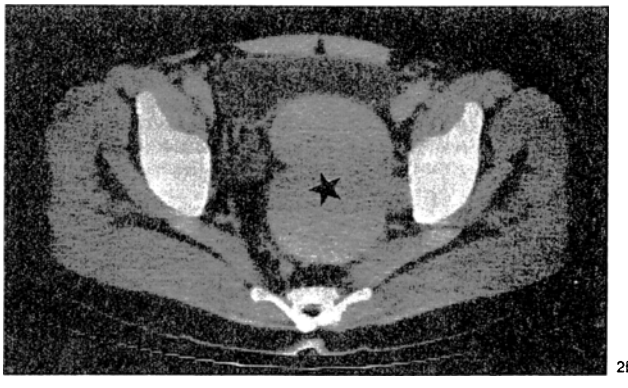

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Fig. 2. — a) Ultrasonogram of the pelvis. A cystic formation occupies most of the cavity. b) CT scan disclosing a large mass (*) filled with contrast, located between the bladder and rectum.

The mass was firm and slightly pulsatile. Temporary occlusion of the hypogastric artery was followed by disappearance of pulsatility, allowing compression and partial emptying of the aneurysm, facilitating dissection through its base where the AVM became evident. Direct intraoperative embolization was performed with gelfoam flakes injected distal to the superior gluteal artery. This maneuver permitted a safe opening of the aneurysm and to achieve a quick control of venous back bleeding from two large openings draining into the left iliac vein. No mural thrombus was found. The left hypogastric artery was then ligated. After suture ligation of the arterial and venous openings, the sac was partially resected. The wall of the aneurysm was thick, with venous aspect. The histopathologic study of the specimen revealed intimal thickening and phlebosclerosis (Fig. 4).

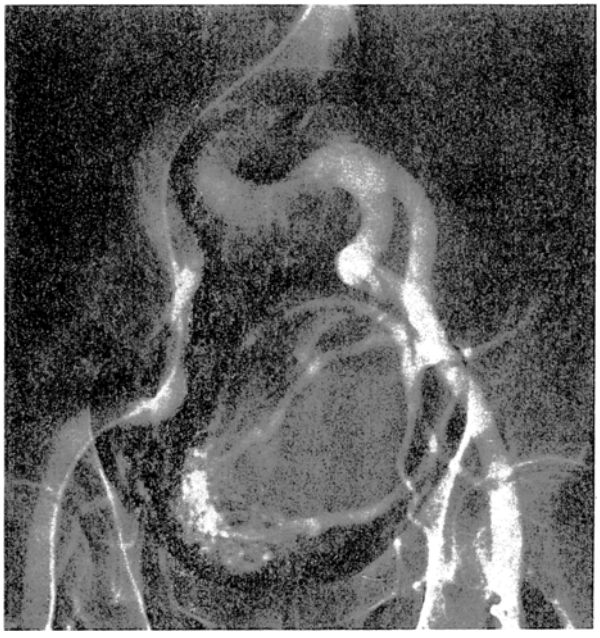
Recovery and follow-up for one year has been uneventful. Postoperative angiogram showed absence of residual lesions (Fig. 5).

DISCUSSION

The first venous aneurysm was reported by Beaumont [2] in 1867, following the autopsy of a patient who suffered a traumatic arteriovenous fistula of the femoral artery. However the first clinical report was done by Osler [1] almost fifty years later. Since then, sporadic cases of venous aneurysms affecting the extremities, visceral veins or superior vena cava have been reported [3]. Based on Abbott's suggestion, venous aneurysms are classified into congenital, acquired, isolated or associated with arteriovenous communications [4]. Most isolated venous aneurysms present in young people and involve the superior vena cava. They are usually fusiform and rarely reach more than 5 cm but may be complicated by pulmonary embolism from mural thrombus.

Thompson in 1969 reviewed the literature referring to central venous aneurysms associated to arteriovenous fistulas [5]. However, most of the cases were secondary to longstanding traumatic arteriovenous fistulas of the lower extremities and the venous aneurysm presented in the external iliac vein. In the recent review by Yao [3] only two cases presented in the upper extremity.

The present case, apparently congenital, would be the first case affecting the hypogastric territory. The factors involved in the progressive dilatation of the veins include the flow rate determined by the size and duration of the fistula, the compliance of the



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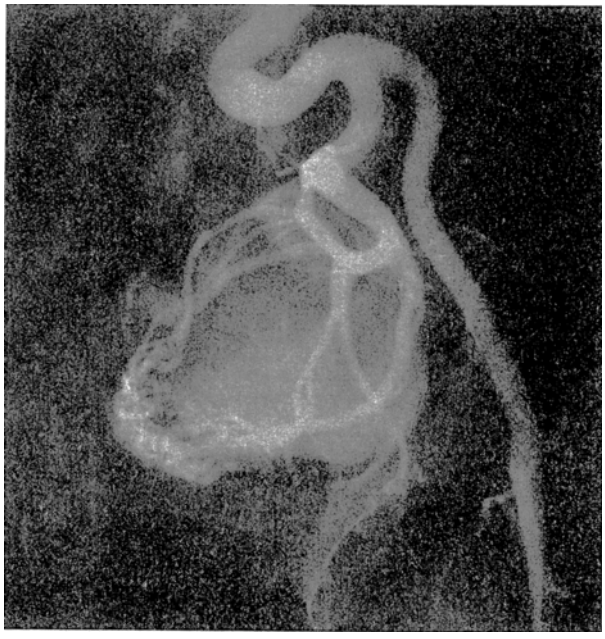

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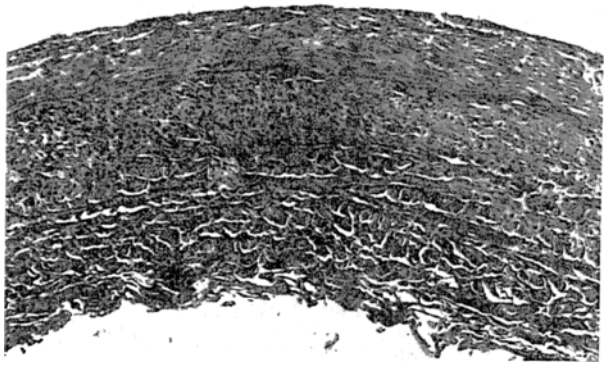

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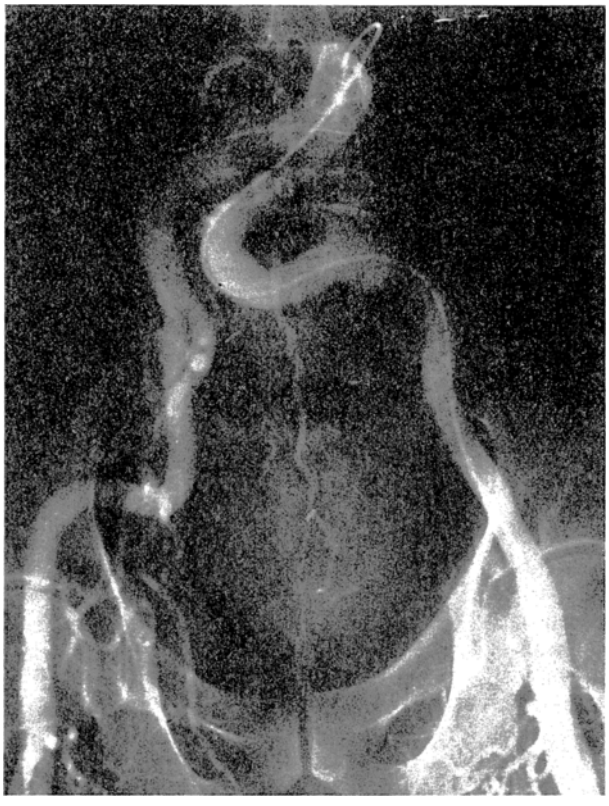

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Fig. 3. — a) Abdominal aortogram showing tortuosity and dilatation of the left common iliac and hypogastric arteries. Branches distal to the superior gluteal artery fill an arteriovenous malformation and a large aneurysmal sac. b) Selective catheterization of the left common iliac artery.

Fig. 4. — Microscopic view of the aneurysmal wall. Intimal thickening and increased fibrous tissue in the outer layers. (Elastic van Gieson stain ; original magnification × 80).

Fig. 5. — Postoperative abdominal aortogram. There is occlusion of the left hypogastric artery and complete disappearance of the AVM and venous aneurysm.

perianeurysmatic tissues and the hydrostatic pressure determined by the anatomical situation of the abnormality. It is interesting that although the fistula may be located in the popliteal area, the dilatation may present in the external iliac vein. Possibly local factors determined by the histologic structure of the vein are also involved. In our case, the aneurysm arouse in a vein draining into the common iliac, most likely the hypogastric vein. The increased fibrous tissue and intimal thickening of the aneurysmal wall are secondary to the longstanding increased pressure. In previous cases, the fistula was known

for up to 36 years [3]. Endophlebosclerosis and endophlebohypertrophy have been reported as pathologic features in cases of primary venous aneurysms [6]. Due to the slow growing rate, most of the cases have evolved asymptomatically becoming clinically evident as a mass or as an autopsy finding. Our case became evident due to compression of bladder and rectum. Spontaneous rupture has never been reported. Systemic complications are possible if the increased flow leads to high cardiac output failure or the aneurysm harbors thrombus which potentially could result in pulmonary embolism.

Currently, the treatment of choice of isolated AVM of the pelvis is intraarterial embolization [7]. The association of a venous aneurysm demands surgical excision, therefore a combined approach should be the therapy of choice. In the present case, symptomatic relief was achieved by direct intraoperative selective embolization and partial resection, allowing complete control of the arterial feeding as well as venous drainage.

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RESUMEN : Los aneurismas venosos son muy raros. Pueden ser de origen congénito o adquirido estando relacionados, frecuentemente, con comunicaciones arterio-venosas. Se trata de un varón de 58 años, afecto de dolor sordo en región iliaca izquierda, impotencia y constipación intestinal, con antecedentes de litiasis renal. En la exploración clínica se aprecia un masa blanda e indolora, con ligero latido y thrill a nivel de la fosa iliaca. Tanto la ecografía como el TAC ponen de manifiesto una formación quística en cavidad pelviana. La arteriografía muestra una pequeña dilatación de la arteria iliaca primitiva junto a una malformación arterio-venosa pelviana, que drena hacia una gran cavidad sin conexión con las arterias iliacas contralaterales. El paciente fue tratado quirurgicamente con éxito mediante embolización selectiva peroperatoria de la malformación arterio-venosa y resección parcial del aneurisma venoso que con un tamaño de 10,6 × 8 ×6,7 cm. ocupaba la mayor parte de la fosa iliaca con desplazamiento del ureter y de la vejiga, y compresión del recto. El estudio anatomopatológico de la pared confirmó la estructura venosa de la malformación. No se presentaron complicaciones postoperatorias. La posibilidad de dilatación venosa ha sido descrita en injertos venosos de alto flujo, en las fístulas A-V para accesos vasculares y raras veces en el sector proximal de las fístulas A-V traumáticas de los miembros inferiores. En el caso descrito la etiología es probablemente de origen congénito y creemos que se trata de la primera publicación en lengua inglesa de un caso de aneurisma venoso que afecta el territorio hipogástrico.

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