Congenital aneurysms of the deep veins of the lower extremities

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This is a report of our experience with seven children with congenital aneurysms of the deep veins of the lower extremities. All the patients had various clinical features of angiodysplasia. With the use of ascending functional phlebography the patients were divided into two groups: (1) four patients with fusiform aneurysms and (2) three patients with saccular aneurysms. Patients in group 1 had venous reflux and a mild degree of venous insufficiency, which were surgically treated. Three patients with saccular aneurysms (group 2) showed no signs of venous insufficiency and are still being carefully followed. Anatomic and functional assessment of the deep venous system by ascending functional phlebography is essential for planning an appropriate treatment for this rare clinical entity. (J VASC SURG 1987;5:765-8.)

Venous aneurysms, unlike arterial aneurysms, are extremely rare. Most venous aneurysms encountered in adults were associated with pulmonary embolism.1,3 The purpose of this article is to summarize our experience with seven children with congenital venous aneurysms of the deep veins of the lower extremities and to emphasize the main clinical and radiologic features of this rare entity.

MATERIAL AND METHODS

Eighty-eight children with congenital and acquired venous disorders of the lower extremities had complete workups: (1) physical examination including measurement of the length and circumference of both legs in recumbent and standing positions, (2) Doppler ultrasound examination of the main vessels, (3) determination of the partial oxygen tension in venous blood (arteriovenous connections), and (4) plain x-ray films of the limbs to demonstrate soft tissue changes such as phleboliths and hemangiomas.

All the patients underwent ascending functional phlebography, a method previously described.4 The posterior tibial vascular bundle, located behind the medial malleolus, was exposed after the patient was given a local anesthetic, and the vein was catheterized. Methylglucamine diatrizoate (Hypaque 50%) (1 ml/kg to a maximum of 20 ml) was injected rapidly by hand without tourniquet. Radiographs were obtained with the patient standing up and his leg internally rotated at 45 degrees and then at the following intervals: (1) immediately after injection; (2) 30 to 40 seconds later, after active functional stress consisting of lifting his heels four times; and (3) 3 minutes after the first radiograph (an empirically selected interval). When exploration of the posterior tibial vascular bundle failed to reveal the associated veins, phlebography was performed via a vein on the dorsum of the foot with a tourniquet applied above the ankle joint. Normally the contrast material is completely cleared from the venous system in 3 minutes. The degree of venous insufficiency and the anatomic abnormalities outlined by the three radiographs were classified according to the classification system preferred by us.4

RESULTS

Congenital venous aneurysms of the deep veins of the lower extremities were diagnosed in seven children. The main clinical and radiologic findings are summarized in Table I. All the patients had unilateral disease, five of them in the left limb. There was no significant difference in the length and circumference of the involved extremities. Various signs of angiodysplasia such as vascular nevi, port-wine stain, hemangiomas, and varicosities were found in all the patients. One patient had two episodes of hemorrhites of the left knee. None of the patients in this series had clinical or sonographic signs of arteriovenous malformations.

RADIOLOGIC FINDINGS

Ascending functional phlebography of the lower extremities demonstrated two types of deep vein an-
Fig. 1. Fusiform aneurysmatic dilatation of the deep venous system of the calf (A-C). Note venous reflux via typical communicants (arrow). Three minutes after injection (C), there is considerable retention of contrast material, indicating a second degree of venous insufficiency.

Table I. Clinical and phlebographic findings in patients with venous aneurysms

<table>
<thead>
<tr>
<th>Patient</th>
<th>Sex/age</th>
<th>Side</th>
<th>Clinical presentation</th>
<th>Venographic findings</th>
<th>Venous insufficiency*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>F/12 yr</td>
<td>Left</td>
<td>Intradermal venodilatation of buttck and thigh; painful varicosity at the popliteal fossa</td>
<td>Fusiform aneurysms of the deep calf veins</td>
<td>Mild reflux into the superficial venous system</td>
</tr>
<tr>
<td>2</td>
<td>M/14 yr</td>
<td>Left</td>
<td>Multiple subcutaneous hemangiomas of foot and calf</td>
<td>Fusiform aneurysms of posterior tibial vein</td>
<td>Mild reflux into intramuscular hemangiomas</td>
</tr>
<tr>
<td>3</td>
<td>F/8 yr</td>
<td>Right</td>
<td>Port-wine vascular nevus; venous cavernous hemangiomas of foot, calf, and thigh</td>
<td>Fusiform aneurysms in the calf veins</td>
<td>Single communicating vein (atypical) into intramuscular hemangioma of the thigh</td>
</tr>
<tr>
<td>4</td>
<td>M/7 yr</td>
<td>Left</td>
<td>Port-wine nevi and varicosities in knee and ankle regions</td>
<td>Fusiform aneurysms in the calf veins</td>
<td>Significant reflux into angiodyplastic formations of the knee region</td>
</tr>
<tr>
<td>5</td>
<td>F/7 yr</td>
<td>Left</td>
<td>Conglomerate of dilated veins in foot; recurrent haemorrhosis of the knee</td>
<td>Saccular aneurysm of popliteal vein</td>
<td>None</td>
</tr>
<tr>
<td>6</td>
<td>M/3 yr</td>
<td>Left</td>
<td>Intradermal venodilatation</td>
<td>Multiple saccular aneurysms of superior femoral vein</td>
<td>Mild reflux into intramuscular hemangiomas</td>
</tr>
<tr>
<td>7</td>
<td>M/7 yr</td>
<td>Right</td>
<td>Venous cavernous hemangiomas of leg</td>
<td>Multiple saccular aneurysms of popliteal vein</td>
<td>None</td>
</tr>
</tbody>
</table>

*Grade I, retention of less than 50% of injected contrast material in the superficial or deep venous system; grade II, retention of more than 50% in one system or less than 50% in both systems at once; grade III, simultaneous retention of more than 50% in both systems.

eurysms: fusiform and saccular. Patients 1 through 4 had multiple fusiform aneurysms. The aneurysmatic dilatation involved 50% to almost 75% of the length of the calf (Fig. 1). Dilatation of all the calf veins was found in patients 1, 3, and 4. In patient 2 only the posterior tibial veins were dilated. Ascending functional phlebography demonstrated venous reflux from the deep to the superficial venous system in all the patients with fusiform aneurysms. In patient 1 contrast material refluxed into the superficial
venous system via typical communicants (Fig. 1). In the other three patients, atypical venous communicants leading into intramuscular and subcutaneous angiodysplastic formations were demonstrated (Fig. 2). Patients 1, 2, and 4 had fusiform aneurysms and some degree of venous insufficiency (Fig. 1).

Saccular aneurysms were found in patients 5, 6, and 7. Two had popliteal aneurysms, and one (patient 6) had aneurysmatic dilatation of the superficial femoral vein. Patient 5 had a solitary aneurysm (Fig. 3), whereas the other two had multiple aneurysms. Patient 6 had mild venous reflux. None had clinical or radiologic signs of venous insufficiency.

**DISCUSSION**

The true clinical entity of venous aneurysm was not established until 1950, when Abbott first reported venous aneurysm of the superior vena cava and offered a classification of this abnormality. The cause of congenital venous aneurysms is still unknown; it may be related to weakness of the vein wall. Because venous aneurysms may be an integral part of other congenital vascular malformations, clinical findings of these anomalies, such as varicosities, hypertrophy of limb, port-wine stain, or hemangiomas, are usually found in this group of patients.

In an excellent review on aneurysms of the venous system, Yao et al. summarized the literature and their experience in the treatment of congenital and acquired types of this rare entity. The majority of congenital venous aneurysms were found in the territory of the superior vena cava, visceral circulation, face, and neck. Congenital venous aneurysms of the lower extremities have rarely been reported. In the present series all the patients had typical features of angiodysplasia. One may assume that venous aneurysms may be part of this anomaly. The traditional classification of venous dysplasia proposed by Malan and Puglioniisi and later by Lea Thomas and Andress used the term *phlebectasia* to describe patients in whom the principal abnormality consists of dilatation of the superficial venous system, rather than the deep venous trunks. In the present series the main disease was found in the deep venous system. Two different types of aneurysms were demonstrated by phlebography: fusiform and saccular. Their different anatomic and functional characteristics were accurately assessed by ascending functional phlebography. This assessment had a practical therapeutic application: four patients with fusiform aneurysms and phlebographic evidence of venous reflux and insufficiency underwent ligation of the incompetent
Fig. 3. Saccular aneurysm of the popliteal vein (A-C). No venous reflux is demonstrated. Minimal amount of contrast material is retained in superficial veins (C).

communicating veins and excision of dilated superficial veins and angiodysplastic malformations. Three patients with saccular aneurysms and no clinical or radiologic signs of venous insufficiency did not have surgery and are still being carefully followed.

Our approach is based on the fact that pulmonary embolism, secondary to popliteal vein aneurysm, was never reported in children and that the excisional and reconstructive venous surgery of this area is still resulting in a relatively high failure rate because of postoperative thrombosis.1,3,8

In conclusion, vascular surgeons confronted with this rare group of anomalies should base their therapeutic approach on morphologic and functional data obtained by the various diagnostic modalities. Ascending functional phlebography is the method we prefer to obtain reliable information essential for planning an appropriate therapy.

REFERENCES