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Case Report

Primary aneurysm of the greater saphenous vein: Case report and literature review

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Abstract. Primary venous aneurysm is a rare vascular abnormality that can occur throughout the venous system. We report a proximal greater saphenous vein aneurysm in a twenty one-year old male. A venous duplex ultrasonographic examination showed a fusiform venous aneurysm (5 cm in diameter) of the proximal greater saphenous vein containing occluding thrombus adherent to its wall and extending to saphenofemoral junction. Operation was performed due to apparent symptoms and the potential risk for thromboembolic complications. Aneurysm was exposed through a vertical left femoral incision. It was totally filled with thrombi. Histopathological examination of the aneurysmal wall demonstrated fibrocellular thickening with irregular vascular proliferation with multiple reduplications of internal elastic lamina. Because of the risk of thromboembolism, saphenous vein aneurysm containing occluding thrombus extending to saphenofemoral junction should be treated surgically.

Key words: Greater saphenous vein, venous aneurysm

1. Introduction

An aneurysm is a focal dilatation of a blood vessel. The term is most commonly applied to dilatations of arteries. However, aneurysms may occur in any part of the vascular system including veins (1). Venous aneurysms were first mentioned in the literature by Osler (2) in 1915, and thereafter Harris, in 1928, described an infant with congenital venous cyst of the mediastinum (2). In contrast to arterial aneurysms, venous aneurysms are quite rare. Hilscher suggested the term of venous aneurysms in contrast to other malformations of the venous system (2,3).

Generally trauma, inflammation, congenital weakness or degenerative changes in the venous wall resulting from a connective tissue disorder or local inflammatory process and cardiovascular abnormalities producing increased venous pressure should be considered as a possible cause of venous aneurysms (3,4).

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In this report, we describe a 21-year-old male with primary aneurysm of the proximal greater saphenous vein (GSV). We think that this case is a unique presentation. According to our knowledge this is the first report of such a case.

2. Case report

A twenty one-year old male was referred to Cardiovascular Surgery Department of Yuzuncu Yil University Hospital, Van, Turkey. He initially noted a painless swelling of a right subinguinal lesion, which diminished in size when the swelling depressed and become prominent when released. Over 6 months, this lesion increased in size and became painful. He had no history of trauma, chronic inflammatory disease or arteriovenous fistula. Physical examination revealed a subcutaneous painful mass, which measured 5 cm in diameter and regular borders located on the right subinguinal region. Redness, tenderness or other evidences of inflammation were not present. On auscultation, no bruit could be heard in this region. Results of physical examination were otherwise normal.

Electrocardiogram, echocardiogram, chest and right extremity radiographs all yielded normal findings. Results of routine biochemistry and blood investigations were also in normal limits.

A venous duplex ultrasonographic examination showed a fusiform venous aneurysm (5 cm in diameter) of the right GSV containing occluding thrombus adherent to its wall and extending to saphenofemoral junction (SFJ). No other venous pathologies were identified.

Operation was performed due to apparent symptoms and the potential risk for thromboembolic complications. At the operation, aneurysm was exposed through a vertical left femoral incision. Following intravenous heparin (5000 IU bolus) administration, a vascular clamp was placed tangentially across the SFJ to prevent thromboembolic event and the aneurysm was resected. It was totally filled with thrombi. The distal end of the vein was ligated and SFJ was closed with 5-0 prolene suture. He had an uneventful postoperative course. He was discharged on the seventh postoperative day and remained asymptomatic for two months after surgery.

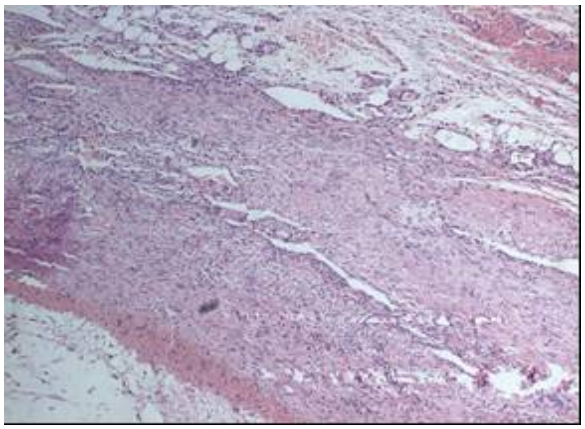

Fig. 1. Aneurysm wall shows fibrocellular thickening with irregular vascular proliferation. Fragment of recent intraluminal thrombus is present. (Hematoxylin-eosin stain; original magnification X 400).

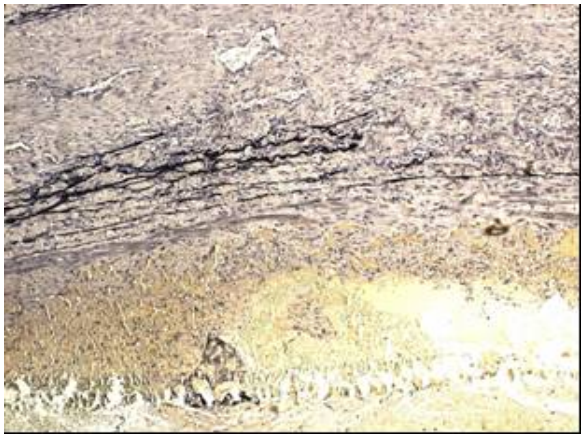

Fig. 2. Elastic tissue stain showing multiple reduplications of internal elastic lamina. (Verhoeff's and van Gieson's stain; original magnification X 400).

Histopathological examination of the aneurysmal wall demonstrated fibrocellular thickening with irregular vascular proliferation (Fig. 1) with multiple reduplications of internal elastic lamina (Fig. 2). Wall of the aneurysm was thinner than the vein wall of the normal segment. But its basic structure (intima, media and adventia) was preserved. There was no evidence of arterialization of the vein, which is characteristic finding of an arteriovenous malformation.

3. Discussion

A primary venous aneurysm is best described as a solitary area of venous dilatation that communicates with a main normal-size venous segment, and it must have no association with an arteriovenous communication or a pseudoaneurysm. Most importantly, it should not be contained within a segment of varicose vein (5). Morphologically venous aneurysms can be divided into 2 subtypes (fusiform, saccular).

Unlike secondary type venous aneurysms, the etiology of primary venous aneurysm remains unknown. However, several hypotheses have been proposed (3). Endophlebohypertrophy (intimal hypertrophy) and endophlebosclerosis are important factors in venous aneurysm formation, in a manner similar to the role of atherosclerosis in arterial aneurysm formation (6). A marked decrease in the number and size of the muscular and elastic fibers in the aneurismal segment of the vein wall is a very common finding, although these changes cannot be affirmed by some authors (7). In contrast, Gilbert et al.(8) found a histologically normal venous structure and a normal component of elastic tissue. The histopathological discrepancies between different reports could suggest different etiologies and differences in the location or in the chronicity of the lesion (7).

Although duplex scanning, CT scanning, and MRI are important noninvasive modalities, venography is mandatory to precisely define the venous anatomy before surgical repair, particularly in patients with a prior history of deep venous thrombosis (9). However, the proximal GSV and the SFJ are especially difficult to assess with venography (10). Additionally, when partial or complete thrombosis exists, the venography could not suggest the existence of venous aneurysm. Duplex scanning can be performed quickly and easily and also measures true size of the venous aneurysm, and permits knowledge of the presence and extent of thrombus within the venous lumen with a complete functional assessment of both

superficial and deep venous systems (7). Aldridge et al. (11) suggest that all patients with suspected or confirmed pulmonary emboli (PE) should undergo venous duplex imaging. It also delineates the thrombotic extent of the aneurysm. Additionally, the chest radiograph or the angio-CT-scan is useful to diagnose a concomitant PE (12).

In the differential diagnosis of the GSV aneurysms, it is important to consider varicose veins, inguinal hernias, lymphadenopathies or some subcutaneous tumors. Also, the existence of arteriovenous fistula or hemangiomas and other vascular malformations should be ruled out (7). In this case, the absence of any connective tissue disorder, infection, trauma, or other arteriovenous malformation supports the primary venous aneurysm.

The deep venous system aneurysms, especially the popliteal and iliofemoral veins may cause pulmonary embolism. Similarly, the proximal GSV aneurysms containing thrombi adjacent to SFJ may cause PE. Therefore these aneurysms should be operated on to avoid risk of pulmonary embolism, as was done in our patient.

Once a venous aneurysm is diagnosed, it seems important to exclude it from the circulation, especially when an intraluminal thrombus mass is present (13). Several operative techniques have been used in venous aneurysm of the limbs: ligation and simple excision, and venorrhaphy; excision and vein patching; excision with interposition vein grafting; tangential excision with lateral venorrhaphy; autologous vein patch; and complete resection. The method of treatment is usually dictated by the anatomical location. It is suggested that tangential aneurysmectomy with lateral venorrhaphy is safe and offers excellent result in saccular venous aneurysms without intraluminal thrombi (2). If a tangential aneurysmectomy with lateral venorrhaphy is performed, the venous endothelium injured by the vascular clamp should be excluded from the circulation, which is important. Excluding the endothelial damage caused by the clamp should reduce the need for postoperative anticoagulation and improve patency (11). However, we believe that risk of recurrence cannot be prevented by tangential aneurysmectomy and lateral venorrhaphy. Therefore, we suggest that all venous aneurysms (fusiform or saccular) should be treated by excision with ligation or by excision with graft interposition if required.

Recently, the endoluminal laser, endovenous radiofrequency energy or sclerotherapy should also be used as an alternative to surgery. It is suggested that sclerotherapy may be a simple, effective method to treat primary venous aneurysm of the superficial venous system (14). However, large and extensive thrombosed venous aneurysms should be treated surgically, as was done in this case.

A ligation of the aneurysm in the deep venous system (popliteal vein) should be avoided and venous continuity should be achieved to prevent possible complications. However, the aneurysmatic GSV (superficial vein) may be ligated, if required.

Histopathological findings help to distinguish a venous aneurysm from a varicose vein. Histological findings of venous aneurysm may be loss of elastic layers and hypertrophy of the connective tissue, absence of the media and adventitia, absence of smooth muscle cells, endophlebohypertrophy, endophlebosclerosis, and the thinning of vein wall without congenital anomaly (4). Endophlebohypertrophy consists of progressive intimal proliferation of elastic, muscle, and connective tissue; it occurs as a normal response to mechanical stress. Endophlebosclerosis is a degenerative pathologic process characterized by alterations in group substance, disruption and loss of elastic fibers, loss of medial muscle cells, and fibrosis (11).

In contrast to venous aneurysm, fibrous tissue is increased in varicose vein. Particularly, muscle layers and the vessel wall are thickened. In varicose vein, ectasia is also accompanied by prolongation of the vein, whereas venous aneurysm is not associated with prolongation of the vein (15). In view of these differences, it may be reasonable to regard venous aneurysm and varicose vein as different entities (16).

Thrombus formation, post-thrombotic syndrome, PE, and rupture are the known complications of the venous aneurysms. On rare occasions GSV aneurysms can be associated with a thrombophlebitic process and in exceptional circumstances with PE risk, especially when an ascending thrombus reaches the deep venous system (17). Additionally, focal peripheral neuropathy has also been reported as a complication of primary venous aneurysm (18).

4. Conclusion

In conclusion, duplex ultrasonography should be required for diagnosis in totally thrombosed aneurysm in proximal greater saphenous vein. Because of the risk of PE, progressive enlargement and rupture, patient presenting with the GSV aneurysm containing occluding thrombus extending to SFJ warrants surgical intervention. Additionally, protective

measurements should be taken during its surgical manipulation to prevent PE.

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