Spontaneous intra-operative rupture of an undiagnosed internal iliac vein aneurysm during laparotomic lymph node dissection for endometrial cancer: a unique case report

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ABSTRACT

Iliac vein aneurysms are rare, but they can be associated with major surgical complications. Our case report deals with an undiagnosed internal iliac vein aneurysm that caused intra-operative major bleeding during lymphadenectomy for endometrial cancer. Surgical manipulation of the vessel, although gentle, caused rupture of the vein that proved impossible to repair and the patient died. We suggest that the etiology of vessel injuries should be investigated in more depth in situations where surgical trauma is not commensurate with vessel damage.

KEYWORDS

Aneurysm; iliac vein; tunica media; hemorrhage; lymphadenectomy.

Introduction

A 75-year-old woman came to the Sant'Anna Hospital in Turin (Italy) in April 2016 complaining of abnormal uterine bleeding in menopause. She had no history of medical diseases other than hypertension, which was well controlled with enalapril, lercanidipine and amiloride, and lower leg phlebitis. She had no history of trauma or surgery. In the past, she had had two pregnancies with spontaneous delivery. She was moderately overweight (BMI 26 kg/m^2) and presented a slight dorsal kyphosis. She did not smoke.

Case presentation

Pelvic exploration revealed a cervical canal polyp measuring 15 mm, protruding from the external uterine orifice; furthermore, second-degree anterior and central pelvic organ prolapse was found. The patient underwent an ultrasound scan: both ovaries were normal in dimension and morphology, but the scan revealed endometrial thickening (20 mm).

Hysteroscopic polypectomy and endometrial biopsy were performed. Histological examination revealed the presence of endometrioid adenocarcinoma cells with moderately differentiated squamous metaplasia areas on endometrial biopsy; instead, the removed cervical polyp consisted of ectropion tissue and showed chronic inflammation. Total hysterectomy with bilateral salpingo-oophorectomy was indicated. Intra-operative histological examination was planned to assess myometrial involvement. The surgical approach was laparotomic. The bilateral salpingo-oophorectomy was performed without surgical complications. The specimen was sent to pathology for intra-operative analysis, which reported neoplastic invasion of more than 50% myometrial thickness, therefore pelvic lymphadenectomy was indicated according to our institutional guidelines. After the removal of a bulky right external iliac lymph node (maximum diameter 2 cm), the dissection proceeded, and during the ligature of a very small venous vessel, located in the adipose tissue above the right internal iliac vein, a spontaneous internal iliac vein rupture occurred, even though only gentle traction was applied to right hypogastric vein.

The vascular surgery team on call was consulted intra-operatively: three attempts at vascular suture were made, unsuccessfully. The rupture extended only a few millimeters, but during the effort to repair it, the vessel broke again two centimeters above the first lesion, at the common trunk level, after just a gentle and atraumatic touch with an atraumatic forceps. The vein wall appeared thin, unable to hold stitches; after each suture attempt, the size of the injury increased. Surgical and medical management was unable to control the blood loss and

Article history
Received 10 Dec 2019 - Accepted 25 Feb 2020

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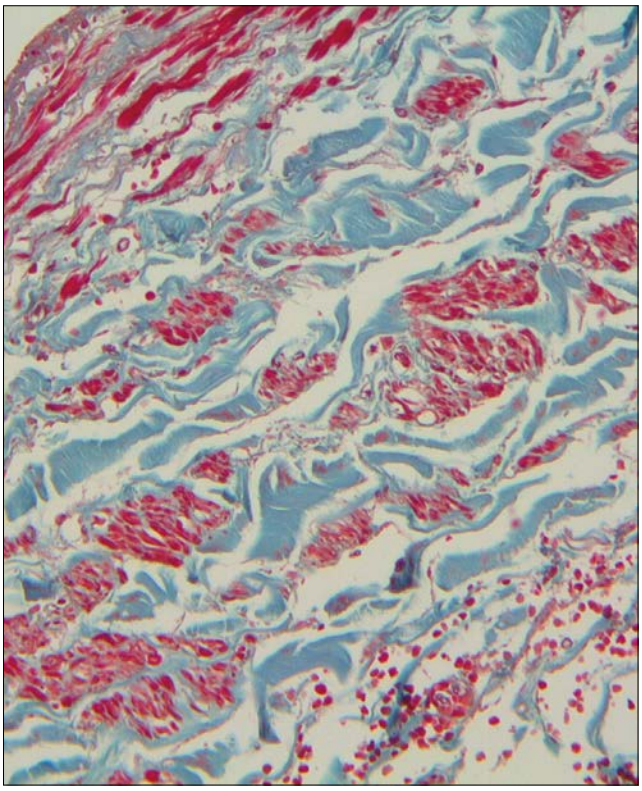
the patient died, despite undergoing cardiopulmonary resuscitation. The delicate blunt dissection made by Kelly clamp could not explain the massive vein wall injury.

The extremely easy rupture of the vein wall and the impossibility of performing surgical suturing, which worsened the vein damage, suggested a pre-existing pathological condition. In fact, autopsy revealed the absence of a continuous muscle layer of the right internal iliac vein, consistent with venous aneurysm (Fig. 1).

Figure 1 Histologic section of the internal iliac vein (trichrome staining, magnification x125). The muscle cells (red) do not continue to form a sleeve (tunica media) but are dissociated from the connective tissue (blue) and present regressive phenomena with cellular necrosis and poorly conserved nuclei.



Discussion

A venous aneurysm is traditionally defined as a dilation of a localized segment of vein to >1.5 times its normal size. However, this definition is incomplete because the venous segment in question may sometimes be part of a vein that is abnormal throughout a long varicose course. Certainly, a venous aneurysm is better defined by features of histological examination: in fact, on microscopic examination it is characterized by thinning or absence of its vascular muscle layer. This finding can be associated with elastin fiber fragmentation and an increase in fibrinous connective tissue [1]. Aneurysms are classified as primary or secondary [2] (to trauma, chronic compression of the vein wall, for example) according to their etiology.

Iliac vein aneurysms are rare. Patients are asymptomatic in 16.7% of cases; if symptomatic, limb swelling or pain, pulmonary embolism, signs of venous insufficiency, abdominal mass effect, back pain, testicular pain, and urinary tract symptoms [3] can be reported. Thrombosis, rupture, embolization and mass effects are the most frequent complications of iliac vein aneurysm [4]. Aneurysms in female patients are primary and asymptomatic in most cases. Primary aneurysms are very infrequent with few cases reported in the literature [5-12]. The largest literature review on iliac vein aneurysm was published by Zarrintan et al. in 2019 [13]. The mean age of the reported patients was 41.7±17.8 years (min=13; max=70). Iliac vein aneurysm is more usual in male patients (only 35.4% were female) and usually involves the external iliac vein. Iliac vein aneurysm is more common on the left side and is usually associated with an atypical relationship between the left common iliac vein and the right common iliac artery. Contrariwise, our patient was a woman and the affected vessel was the right internal iliac vein.

Pelvic vein iatrogenic operative injuries occur during oncological resection in 65% of patients sustaining such vessel injuries; lymphadenectomy is performed in 58% of them, and the iliac vein is the injured vessel in 68%. The injured vessel was frequently mobilized and debrided to allow tension-free anastomosis on healthy tissue. Internal iliac vein rupture causes a mean blood loss of 7300 ml (range: 1200-20 000 ml) and the mean red blood cell transfusion requirement is 20 units (range: 6-58 units). Injuries are repaired by venorrhaphy or primary ligation; patients die in 25% of cases [14].

Our patient's internal iliac vein had a macroscopically normal wall, so no atypical dilatation was found on pre-operative computed tomography or intra-operatively.

On the other hand, her hypogastric vein, although not dilatated (Fig.s 2 and 3), lacked the muscle layer that is normally present, formed by one or two layers of smooth muscle cells, and contributes to the vein wall strength. Surgical manipulation of the vessel, although gentle, caused vein rupture that proved impossible to repair.

Figure 2 Pre-operative computed tomography image showing the right internal iliac vein, which appears macroscopically normal.

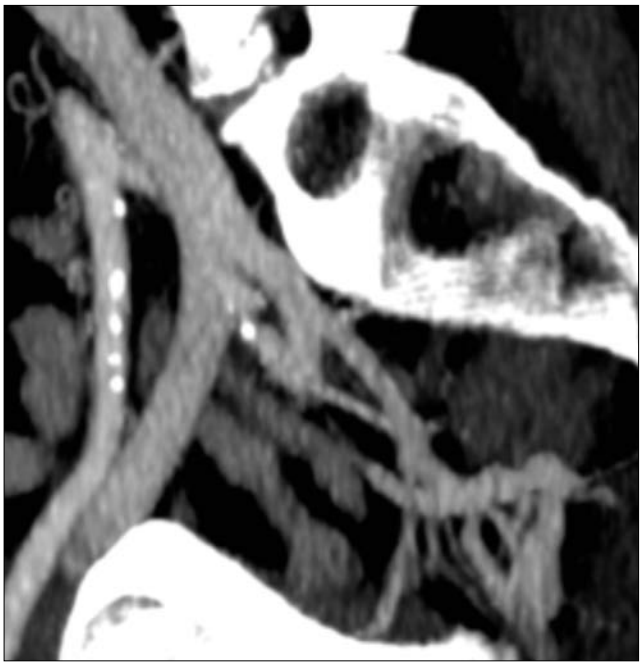
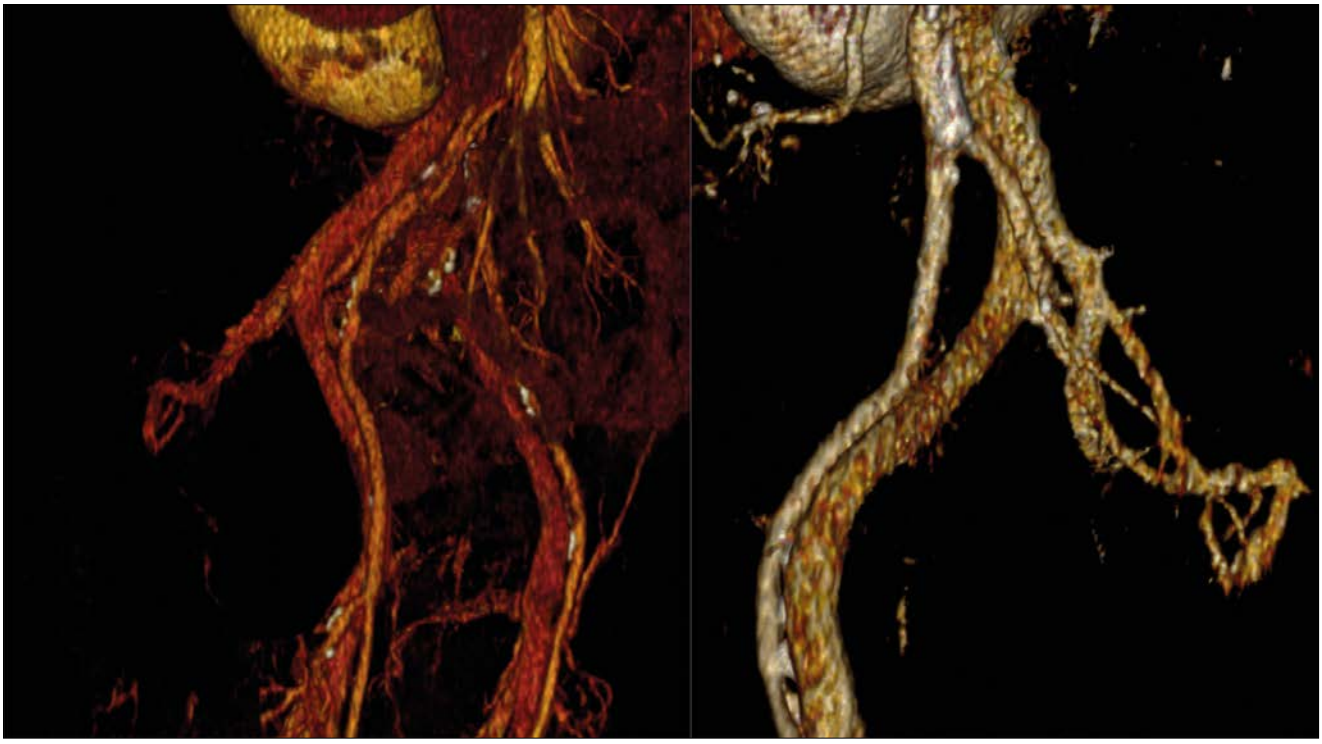


Figure 3 3D CT iliac vein reconstruction.



Conclusion

Our case report deals with an unexpected iatrogenic venous injury that occurred during non-vascular surgery and resulted in the patient's death. We suggest that the etiology of vessel injuries should be investigated in more depth in situations where surgical trauma is not commensurate with vessel damage.

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Acknowledgements: *this research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.*
Conflicts of Interest: *the authors declare that there is no conflict of interest.*