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En bloc resection of the external iliac vein along with broad ligament leiomyosarcoma: A case report

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ABSTRACT

Outcomes following the excision of the external iliac vein during gynaecological oncology surgery are poorly documented. This is because most gynaecological oncologists consider tumours with vascular involvement inoperable. We describe a patient whose right external iliac vein was transected during the removal of a large broad ligament leiomyosarcoma invading the right external iliac vein. The patient's recovery following surgery was uneventful, and she remained disease-free 6 months postoperatively, with minimal morbidity. In describing this case, we hope to educate and inform other gynaecological oncologists facing a similar surgical challenge. We also propose that resection of the external iliac vessels in such cases is safe and feasible, and summarise the anatomical course of venous collaterals, which develop when the external iliac veins are obstructed.

1. Introduction

According to the modern principles of surgical oncology, the aim should be not only to resect the primary tumour mass along with sufficient tumour-free margins to prevent disease recurrence, but also to minimise surgical morbidities so that physiological functions can be preserved [1]. Large and complex pelvic tumours pose a particular surgical challenge to gynaecological oncology surgery, as these tumours are frequently in close proximity to vital organs, in particularly the iliac vasculatures, which supply and drain the pelvic organs and lower limbs. In the past, tumours that invade the iliac vessels would be deemed inoperable because of potential surgical morbidities and mortality risks [2]. Nevertheless, an improved knowledge in surgical anatomy coupled with the availability of modern imaging technology and electrosurgery equipment have allowed surgeons to continue to push the boundaries in surgical oncology. Today, a pelvic tumour which invades the iliac vessels is no longer viewed as a limitation to surgery; these vessels can safely be excised en bloc to achieve negative tumour margins [2–5]. Whether these vessels should be repaired, either through primary anastomosis, using grafts, or simply ligated and left alone, remains a source of debate [6,7].

While extensive literature describes the surgical management of non-gynaecological tumours invading the iliac vessels, comparatively little is known about the surgical management of pelvic tumours of Mullerian origin which invade the iliac vessels. Here, we present the case of a patient with a large uterine sarcoma to highlight the success of surgery in order 1) to demonstrate the feasibility of doing so, 2) to inform other gynaecological oncologists facing a similar challenge, and 3) to add to the pool of data available for future research into this area.

2. Case Report

A 62-year-old woman, with a WHO performance status of 1, and a past history of non-Hodgkin's lymphoma, presented with increasing abdominal pain. An ultrasound scan revealed a 28 cm pelvic mass. Carcinoma antigen 125 (Ca – 125) was slightly elevated at 48kU/L. A computed tomography (CT) scan showed a uterine sarcoma compressing both the ureters, resulting in bilateral hydronephrosis. A magnetic resonance imaging (MRI) scan was also performed to further delineate the mass and plan management (Fig. 1). The MRI demonstrated that the mass was merely "in contact" with the right external iliac vasculature. Her case was discussed at the gynaecological oncology multidisciplinary team (MDT) meeting, and a decision was made for laparotomy at a tertiary gynaecological oncology centre to debulk the uterine mass.

Intra-operatively, a 28 cm solid pelvic mass of uterine origin was noted. The mass was immobile and densely adherent to vital structures of the pelvis and abdomen: omentum, the bladder dome, sigmoid colon and rectum. Both ovaries appeared normal but were buried within the main tumour mass. Marked bilateral hydronephrosis was noted, a finding consistent with the CT scan. In addition, the mass had also invaded the right external iliac vessels. However, there was no apparent extracorporeal disease spread.

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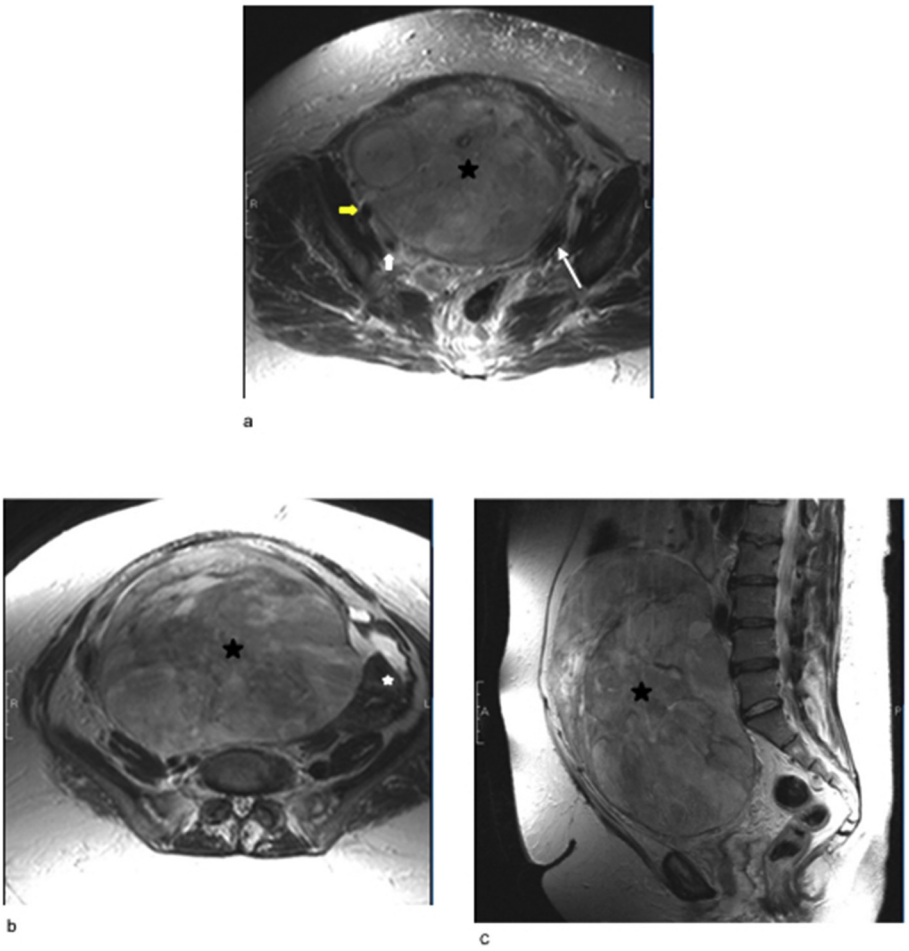

Fig. 1. (a) Axial T2-weighted MRI of pelvis demonstrates a bulky, heterogeneous, intermediate to high signal, mass (black star) in contact with the right external iliac artery (yellow arrow) and the right external iliac vein (short white arrow). The cervix (thin white arrow) is displaced towards the left by the tumour. (b) Axial T2-weighted MRI of the pelvis more superiorly shows the uterus (white star) displaced to the left iliac fossa by the right adnexal mass. (c) Sagittal T2-weight MRI demonstrates the superior extent of the pelvis mass (black star).

In short, she underwent a laparotomy and resection of the pelvic mass, which included: extensive adhesiolysis to free the primary tumour from the pelvic sidewalls, sigmoid colon and rectum; partial cystotomy to remove the bladder dome and primary closure of the defect; hysterectomy and bilateral salpingo-oophorectomies; and excision of the right external iliac vein. Enlarged lymph nodes were removed from both sides of the iliac vessels. The greater omentum was mobilised and brought into the pelvis to act as a biological spacer in preparation for potential adjuvant radiotherapy. A vascular surgeon advised against grafting the right external iliac vein that had been removed en bloc along with the tumour. Complete macroscopic disease clearance was successfully achieved.

Postoperatively, a catheter was left in situ for 14 days and a cystogram was performed to confirm bladder integrity before its removal. The right leg was compressed with class 2 compression hosiery, and the patient was started on low molecular weight heparin. As a consequence of transection of the right external iliac vein, she developed a right femoral deep vein thrombosis (DVT) and was commenced on lifelong warfarin. This was expected, as a large volume of residual blood had stagnated in her right femoral-popliteal veins following transection of her right external iliac vein. Her postoperative recovery was otherwise uneventful and she was discharged home after 2 weeks.

The final histology confirmed a diagnosis of a broad ligament leiomyosarcoma. The patient had adjuvant radiotherapy and remained well 6 months postoperatively. She developed chronic right leg lymphedema, which was managed with class 2 compression hosiery and regular physiotherapy. Longer term, the patient was referred to the regional sarcoma multidisciplinary team meeting for consideration of adjuvant treatment.

3. Discussion

We successfully achieved en bloc resection of a broad ligament leiomyosarcoma along with the right external iliac vein. Postoperatively, the patient developed stage 2 right leg lymphedema [8] (Fig. 2) with minimal compromise to function, demonstrating the feasibility of resecting the external iliac vein in such rare cases. Our vascular surgical colleagues advised against grafting the external iliac vein to restore lower limb blood flow due to possible neovascularisation, where collateral blood vessels would have developed to drain the lower limb.

The femoral vein, which drains the lower limb, continues as the external iliac vein once it crosses beneath the inguinal ligament [9]. It ascends along the brim of the lesser pelvis, alongside and medial to its corresponding artery, before joining the internal iliac vein to form the common iliac vein anterior to the sacroiliac joint [10]. It drains the inferior epigastric and deep circumflex iliac veins [9] and courses along the medial border of the psoas muscle [5].

In pelvic venous obstruction, which develops in patients such as ours, neovascularisation is not necessarily observed; rather, the existing venous channels become dilated to provide adequate collateral drainage [11–13]. As a result of restricted blood flow following the obstruction of the external iliac vein, collateral pathways develop by either

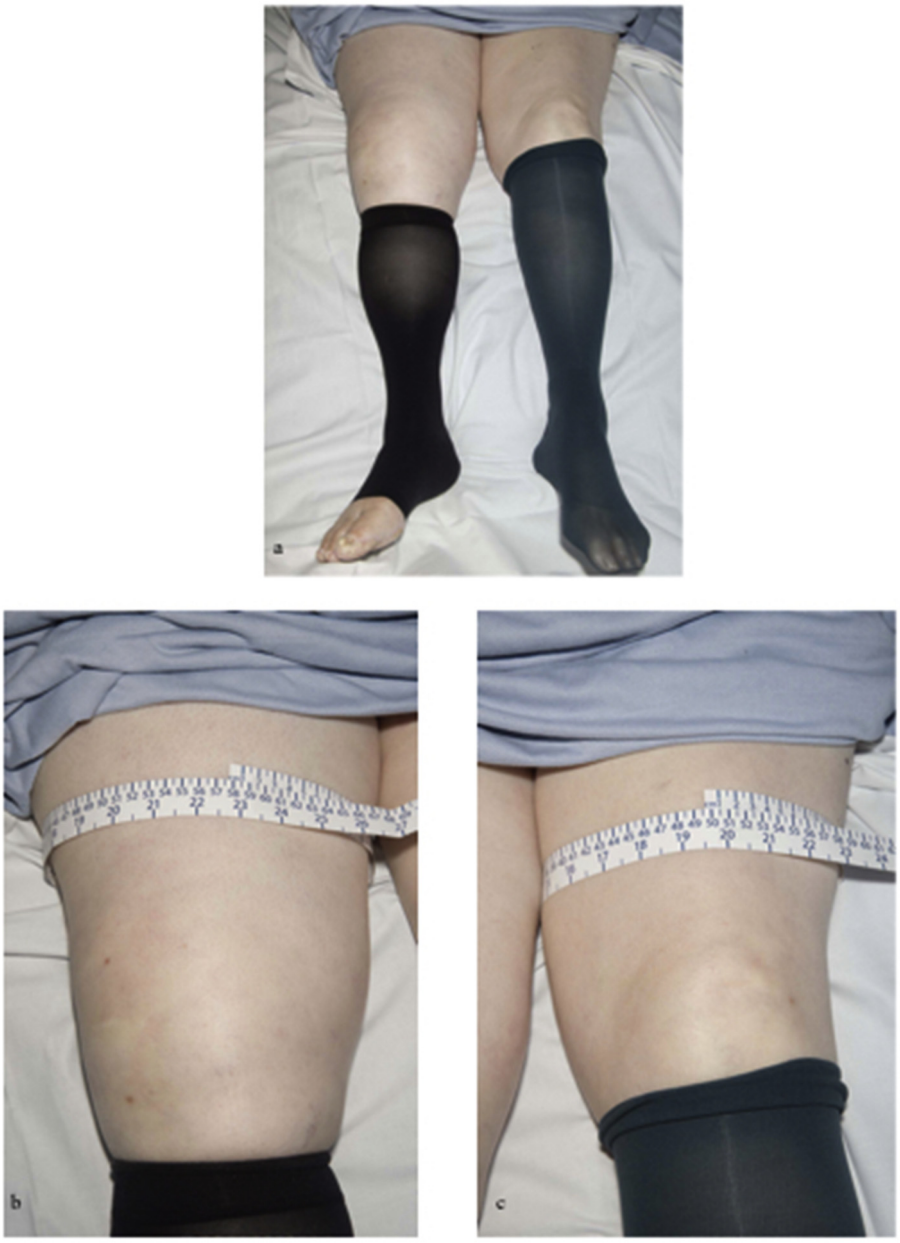

Fig. 2. (a) Image demonstrating overall difference between the right and left lower limbs 3 days post-operatively. The black stocking is the G2 elastic compression stocking. (b) Measurement of the right thigh (26 cm). (c) Measurement of left (unaffected) side (23 cm). Overall difference of 3 cm or 13% following surgery.

(1) bypassing the obstruction to join the common iliac vein on the ipsilateral side of the obstruction via the deep external pudendal, deep circumflex iliac, obturator, and internal iliac veins or (2) running through the veins of the lower anterior abdominal wall, i.e. the superficial external pudendal and pubic veins, and via the parametrial and pre-sacral plexuses to the contralateral, unaffected, side [11,12]. When veins of the lower anterior abdominal wall are utilised as collaterals, they become visible as varicosities on the abdominal wall or across the pubic bone [11–13]. In addition, the internal pudendal vein can also become dilated to serve as an additional collateral pathway [11] in cases of external iliac vein occlusion. If the level of obstruction is below the external iliac vein, the collateral pathways observed between the femoral and internal iliac veins are via the obturator, inferior gluteal, and internal pudendal veins and the pelvic venous plexuses [11].

We decided against re-anastomosing the transected vessel or using grafts for repair given the patient's history and intra-operative findings, as we believed significant collateral circulations, described above, would have formed. Therefore, taking the advice from our vascular surgeon colleague, we felt that vessel repair was not indicated and could have led to unnecessary complications such as graft occlusion and infection. Indeed, others have reported similar outcomes following sacrifice of the external iliac vessels involved in pelvic tumours [14], and recommend no vascular reconstruction is needed where there is no impaired collateral venous drainage [15]. Whether resected iliac veins should be repaired remains a source of much debate [6,7]. A study by Song and colleagues [16] showed that there was 78% venous patency at 65 months post venous reconstruction, thus supporting the view of undertaking venous reconstruction. However, the sample was small (8 patients) and included other major veins such as the femoral vein and vena cava [16], so it could be argued that the results cannot be applied to the iliac veins specifically. Furthermore, recovery following venous reconstruction with grafts was complicated by wound dehiscence and infection. Importantly, the authors did not attempt venous reconstruction in cases where the affected vein was chronically occluded [16].

This is an important consideration because venous reconstruction was performed in cases where they did not believe collateral channels had time to develop. Ghert et al., on the other hand, demonstrated that patients who had vascular reconstruction were more likely to experience post-operative leg oedema than those without reconstruction and were at an increased risk of post-operative complications, including limb amputation [18]. A review by Adelani and colleagues suggested no difference in lower limb oedema in those who had venous reconstruction versus those who did not. There was also no difference in infection and thrombosis risk in those reconstructed with synthetic grafts versus those re-constructed with autogenous saphenous vein grafts [17]. Therefore, it was posited that the incidence of post-operative lower limb oedema is secondary to disruption of lymphatic channels rather than venous insufficiency [19]. Furthermore, it was also suggested that leg oedema could be associated with the degree of disruption to the collateral channels during surgery [7]. On the face of current evidences, it remains debatable whether reconstruction of the iliac veins is of benefit, particularly in our patient, who may require further adjuvant treatment. Any post-operative complications following reconstruction surgery can potentially delay starting adjuvant therapy in oncological patients and this may affect survival outcome.

Our case highlighted the feasibility of complete resection of pelvic tumours involving critical structures such as the external iliac veins; thus negative tumour margins can be safely achieved with minimal morbidity to the patient [2,4,5]. We advocate an MDT approach to managing such rare and complex cases, which should involve a radiologist and vascular surgeon. Most importantly, a frank discussion with the patient should take place to highlight the potential risk of significant surgical morbidities associated with major vascular injuries such as lower limb ischaemia, amputation, nerve damage, and lifelong lower limb oedema, which can adversely affect function and quality of life. We believe that the advancement of three-dimensional (3D) radiological imaging techniques coupled with the increasing use of additive technology in aiding the pre-operative planning of surgery will not only aid surgeons in planning for surgery but will also allow anticipation of additional potential surgical risks, and thus crucially improve pre-operative patient counselling.

Contributors

Fatima Ahmed was responsible for preparation of the original draft.
Rachel Pounds contributed to the review and editing of the draft.
Hong-Giap Teo contributed to the review and editing of the draft.
James Nevin contributed to the review and editing of the draft.
Kavita Singh was responsible for project administration, and contributed to the review and editing of the draft.
Jason Yap was responsible for supervision, conceptualisation, and methodology, and contributed to the review and editing of the draft.

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Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

Declaration of Competing Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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