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Case report

Phlegmasia cerulea dolens and external iliac vein disruption after revision total hip arthroplasty

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ABSTRACT

We present a unique case of phlegmasia cerulea dolens and compartment syndrome secondary to external iliac vein disruption after revision total hip arthroplasty. To our knowledge, this complication has not yet been described following revision total hip arthroplasty. We conclude that although vascular complications are fortunately rare after hip arthroplasty, they can have significant morbidity and mortality. Surgeons should have a thorough understanding of pelvic and hip anatomy for screw and retractor placement and know how to appropriately and expeditiously manage vascular complications should they occur.

Introduction

The incidence of vascular injury is an uncommon complication following total hip arthroplasty (THA), with a reported incidence of 0.2%-0.3% [1-4]. The external iliac artery (48%) or the femoral artery (23%) is the most frequently injured vessels [4]. The etiology of these vascular injuries is multifactorial. Direct injuries can occur via penetration, transection, or trauma from retractor placement [4-6], while indirect injuries may be secondary to patient positioning, dislocation, and relocation maneuvers which cause elongation and/or torsion of vessels [4,5]. The most common complications include thromboembolic events (46%), lacerations (26%), pseudoaneurysms (25%), and arteriovenous fistulas (AVFs) (3%) [4]. In this care report, we review a rare case of an external iliac vein (EIV) injury presenting acutely as phlegmasia cerulea dolens following revision total hip replacement.

Case history

The patient is a 60-year-old male who underwent bilateral hip replacements 20 years ago at an outside institution. He presented with progressive left worse than right groin pain, bilateral startup groin pain, as well as some bilateral thigh pain and subjective instability in the left hip over 4 years without any significant trauma. He denied fevers or any distant history of infection. His physical examination was significant for a limp, as well as a 1-2 mm leg length discrepancy involving the left leg. He did not have any significant restriction of range of motion and had preserved abductor strength bilaterally. The patient had an unremarkable neurovascular examination. His medical history was significant for cardiomyopathy with an ejection fraction of 35%. He is a construction worker and is a nonsmoker. Radiographs revealed diffuse periacetabular and peritrochanteric osteolysis bilaterally with concern for loosening of the acetabular component on the left hip (Fig. 1a and b). All 3 DeLee and Charnley [7] zones were involved in the left side.

The patient was taken to the operating room for revision of the left hip via a posterior approach. The acetabular component was found to be loose with minimal bone attached medially. There was significant periacetabular osteolysis, consistent with a Paprosky 3B defect [8] with a very thin medial wall, as well as a contained defect in the posterior femoral neck and calcar. There was no pelvic discontinuity and no loosening of the stem. The acetabulum and

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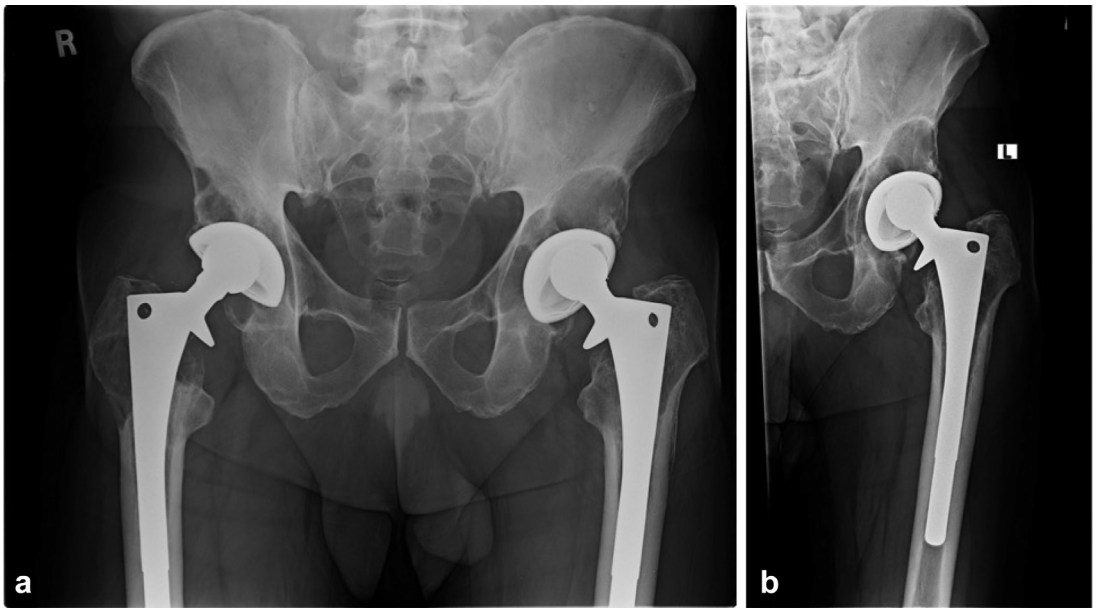

Fig. 1. Preoperative anteroposterior pelvis (a) and left hip (b) demonstrating diffuse periacetabular and peritrochanteric osteolysis bilaterally.

femur were grafted with morsellized fresh frozen femoral head allograft and demineralized bone matrix. A 68-mm trabecular metal cup (Continuum; Zimmer Biomet, Warsaw, IN) was implanted and secured with 1 screw into the ischium and 3 screws into the ilium. A 36-mm highly cross-linked polyethylene liner was utilized (Longevity; Zimmer Biomet). During the case, there was no significant blood loss nor was there any obvious bleeding. The patient's vital signs remained stable throughout the entire procedure. Tranexamic acid was administered per our institution's protocol (1 g prior to incision and 1 g at closure).

Shortly after arrival to the postanesthesia care unit, the patient was found to have a cyanotic left leg (Fig. 2a and b) and complained of significant pain below the knee. He had no motor or sensory function below the knee, but Dopplerable pulses were present in the left foot. Postoperative radiographs were concerning for intrapelvic screw penetration (Fig. 3). An emergent vascular surgery consult was obtained. Within 15 minutes, the vascular attending evaluated the patient. Bedsides ultrasound showed extensive venous thrombosis extending from the popliteal vein to the common femoral vein. With a working diagnosis of acute deep venous thrombosis causing phlegmasia cerulea dolens, the patient was started on a heparin drip. Within an hour, the patient was taken to the operating room for placement of an inferior vena cava filter (IVCF) and thrombectomy. During initial placement of the IVCF, no contrast dye was found to fill the left common iliac vein (Fig. 4a and b). It was presumed that the thrombosis involved the entire left iliac venous tree. After placement of the IVCF, the patient was placed prone and a venogram was performed via access from the left popliteal vein. The EIV was found to be completely disrupted with extravasation of contrast caudal to the acetabular cup (Fig. 4c and d). At this time, the patient became suddenly hypotensive. The patient was then urgently placed supine to anticipate resuscitative efforts. It was then noticed that the abdomen was severely swollen and tense. An emergent exploratory laparotomy was performed to stop intrapelvic hemorrhaging. The EIV was ligated and the abdomen was packed and left open. The patient's leg continued to become more swollen, tense, and cyanotic (Fig. 5) as the case progressed over 2 hours. The decision was made to perform a 4-compartment fasciotomy. All muscles appeared healthy and viable. The wounds were packed and left open. Postoperative examination in the postanesthesia care unit revealed better perfusion of the left lower extremity, as well as some return of gross motor and sensory function to the leg and foot.

On postoperative day (POD) 2, the patient was taken back to the operating room by vascular surgery for irrigation and debridement of the abdomen and left leg, as well as abdominal wound closure and vacuum assisted closure of the leg. On POD 4, the patient was noted to have improved perfusion in the leg, sensation was intact in the tibial, sural, and saphenous nerves, while decreased in superficial and deep peroneal nerves. Motor examination revealed intact gastroc-soleus complex and flexor hallucis longus, while extensor hallucis longus (EHL) and tibialis anterior (TA) muscles were weak. On POD 6, a computed tomography (CT) scan of the pelvis was obtained confirming intrapelvic screw placement (Fig. 6a and b). A discussion was held with the patient regarding the aberrant screw and its proximity to the lumbosacral nerve possibly causing the peroneal nerve palsy. However, given the continued improvement in the patient's neurological examination, no further intervention was performed. The patient's hospital course was further complicated by a pulmonary embolus for which the patient was restarted on therapeutic anticoagulation. The fasciotomy sites were closed with a DermaClose device (SYNOVIS Micro Alliance Companies, Inc, Birmingham, AL) and the patient was discharged on POD 13 with slowly improving EHL and TA function, but still diminished superficial and deep peroneal nerve sensation.

On POD 26, the patient displayed improved TA and EHL function, as well as improved sensation in the superficial peroneal nerve distribution, but continued to have diminished sensation in the deep peroneal nerve distribution. At most recent follow-up 1 year postoperatively, the patient was asymptomatic in the left hip but still has an antalgic gait due to his right hip. He regained active plantar and dorsiflexion of the foot. However, he has chronic pain, swelling, and diffuse paresthesias and dysesthesias in the left foot as sequelae from compartment syndrome. He requires narcotics for his chronic pain. Additionally, he has been receiving local wound care to a portion of the grafted medial fasciotomy wound which has not healed. He has not been able to return to work and is currently on disability.

Fig. 2. Clinical pictures of cyanotic left foot (a) and left leg (b) immediately postoperative in the recovery room.

Discussion

This case represents an unusual complication following revision THA, specifically the acetabular component. To our knowledge, there have been no recent reports of phlegmasia cerulea dolens following revision hip replacement [9]. We hypothesize that the intrapelvic screw placement or extrusion of bone graft through the medial acetabular wall caused the venous injury, which led to venous congestion of the entire limb and phlegmasia cerulea dolens. We also believe that tranexamic acid played a role in preventing profuse bleeding from the venous injury, which eventually became acutely hemorrhagic subsequent to intravenous heparin administration.

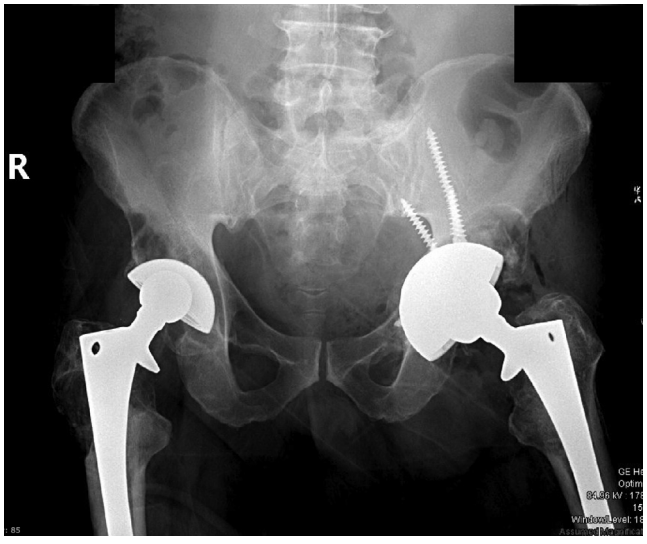

Fig. 3. Postoperative anteroposterior pelvis radiograph demonstrating ilium screw placement into pelvis.

The incidence of vascular injury is an uncommon complication following THA, with a reported incidence of 0.2%-0.3% [1-5]. The difference in incidence of injury between primary and revision surgery has not been shown to be significant [4,5,10]. When vascular injuries do occur, they occur twice as often in women [4,6] and most often affect the external iliac artery (48%) or the femoral artery (23%) [4]. Thromboembolic events (46%) are the most common vascular injury, followed by lacerations (26%), pseudoaneurysms (25%), and AVFs (3%) [4]. Vascular surgery interventions consist of bypass grafting, primary repair, thrombectomy, embolectomy, and/or ligation [4,5].

Vascular injuries can occur by one of several ways. Direct injuries can occur via penetration, transection, or trauma from pressure from retractor placement [4-6]. Indirect injuries may be due to patient positioning, dislocation, and relocation maneuvers which cause elongation and/or torsion of vessels [4,5]. In a review of 16 patients with vascular injuries after total joint arthroplasty, Parvizi et al [5] found that patients undergoing total knee

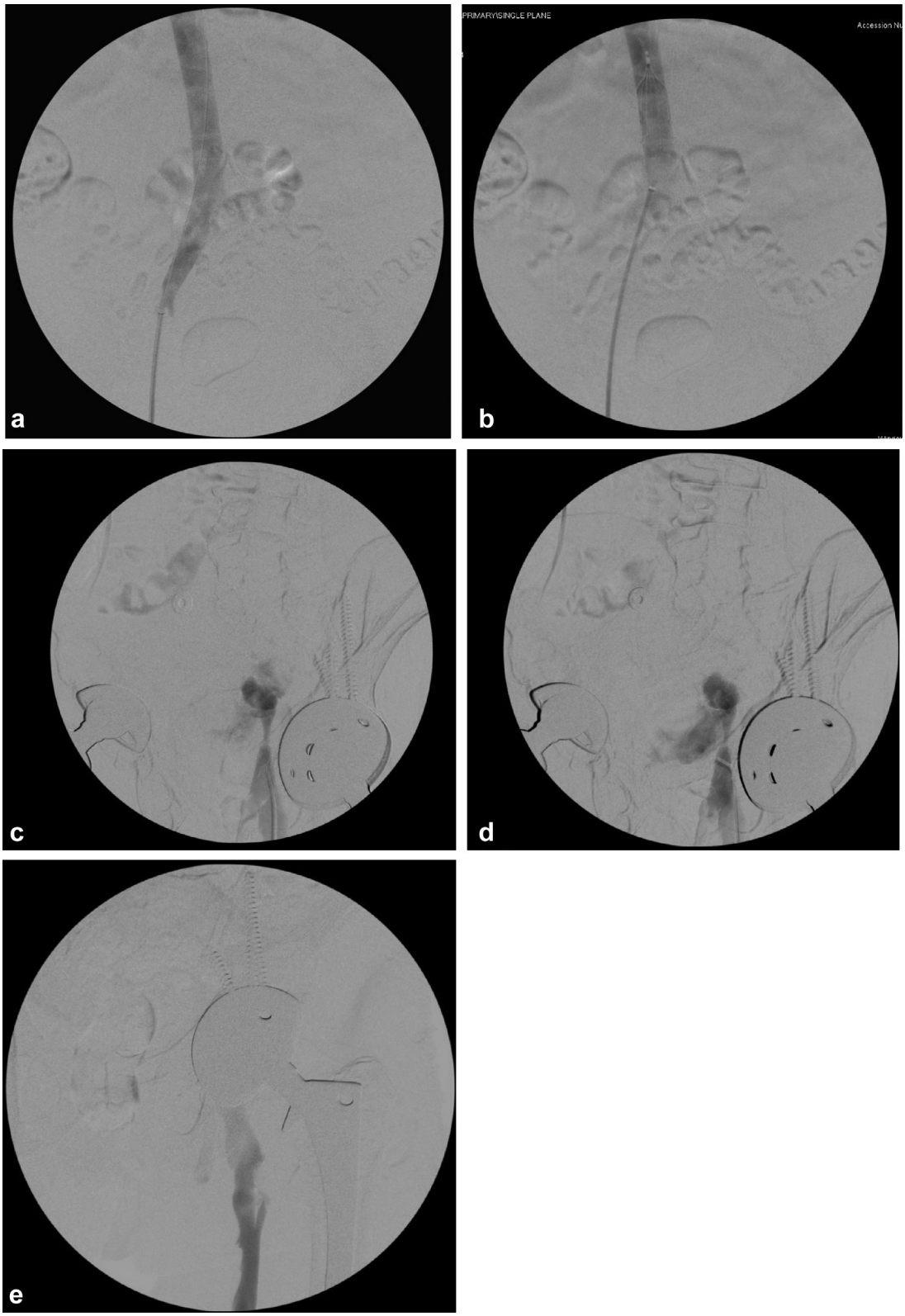

Fig. 4. Intraoperative fluoroscopy of venogram and IVC filter placement. Note absence of contrast in the left common iliac vein (a) during filter placement (b). There is extravasation of contrast caudal to the acetabular cup (c-e) indicating the location of external iliac vein disruption.

arthroplasty were more likely to have indirect injuries to the popliteal vessels (popliteal artery thrombosis), whereas THA patients were more likely to sustain a direct vascular injury (femoral or external iliac artery lacerations). Cementing can also lead to vascular injury secondary to compression and thermal injury [4]. Cement spicules can erode through and perforate vessels, resulting in pseudoaneurysms and AVFs [4]. Acute thrombus formation manifests intraoperatively or postoperatively as limb

Fig. 5. Intraoperative clinical picture demonstrating increasing cyanosis of left lower extremity during the venogram.

swelling (due to venous congestion) or ischemia, as was the case for our patient. Risk factors for vascular injury include acetabular dysplasia, congenital dislocations, acetabular protrusion medial to Kohler's line resulting in a thinned medial wall, pelvic fractures, and prior pelvis/hip surgery resulting in extensive scar tissue formation and atherosclerotic vessels [4,11].

In phlegmasia cerulea dolens, there is complete occlusion of venous outflow. In the milder version, phlegmasia alba dolens, collateral venous outflow of the limb remains despite the venous thrombosis [12]. For orthopaedic surgeons, it is important to know that phlegmasia cerulea dolens and phlegmasia alba dolens can present as and lead to compartment syndrome [12].

EIV injury is a known vascular complication with total hip replacement [11,13-17]. However, there are no prior reports describing acute phlegmasia cerulea dolens as the presenting sign of venous injury. A prior case report noted that EIV injury during total hip arthroplasty could result in intraoperative shock despite minimal bleeding during the surgery [11]. In that report, a large retroperitoneal hematoma was identified on abdominal echography and a laparotomy was undertaken 50 minutes after initial hypotension, which revealed a 2-cm long laceration of the left EIV. In another report, a patient was admitted 4 days postoperative with a large right-sided iliofemoral deep venous thrombosis. A pelvic radiograph showed that the acetabular construct had migrated medially into the pelvis with a CT scan showing compression of the external iliac vessels by one of the acetabular screws [13]. The EIV appears to be more vulnerable than the artery because of its more medial position and the paucity of interposed tissue along the pelvic brim, which protects the artery [11,18].

Some authors have suggested preoperative vascular examination prior to THA [1,4,19]. The risk of vascular problems is reported to be higher in patients with pre-existent vascular insufficiency as exhibited by leg claudication, atherosclerosis, prior coronary bypass surgery, and decreased distal pulses [1,20,21]. However, vascular injuries after total joint arthroplasty can occur in patients with unrecognized or undetected vascular problems [5]. If arterial insufficiency is suspected, the ankle-brachial index (ABI) may help determine the need for further evaluation. Vascular consultation has been recommended for patients with ABI <0.5, and CT angiography for ABI <0.4. Additionally, preoperative arteriography can minimize ischemia time by identifying distal arteries that can be used for bypass procedures, if needed. Additionally, when planning for revision surgery, CT angiography has been recommended in some patients with acetabular protrusion, particularly if the component has been cemented. CT angiography allows the surgeon to identify the relative proximity of the external iliac vessels and determine if they have been incarcerated in cement [1,22].

The placement of acetabular fixation screws poses a known risk of vascular injury. The commonly used quadrant system has been developed to help surgeons intraoperatively identify safe zones for screw placement [2,23]. In this system, the acetabulum is divided into anterior and posterior halves by a line originating from the anterior superior iliac spine going through the center of the acetabulum. A perpendicular line centered over the acetabulum then divides each half into a quadrant. The external iliac and obturator vessels are at risk in the anterior superior and anterior inferior quadrants, respectively. The inferior gluteal and internal pudendal vessels are located adjacent to the posterior inferior quadrant. The "safe zone" is the posterior superior quadrant, because it has had the thickest bone allowing for secure screw fixation (>25 mm). According to Wasielewski et al, the other 3 quadrants only allow for

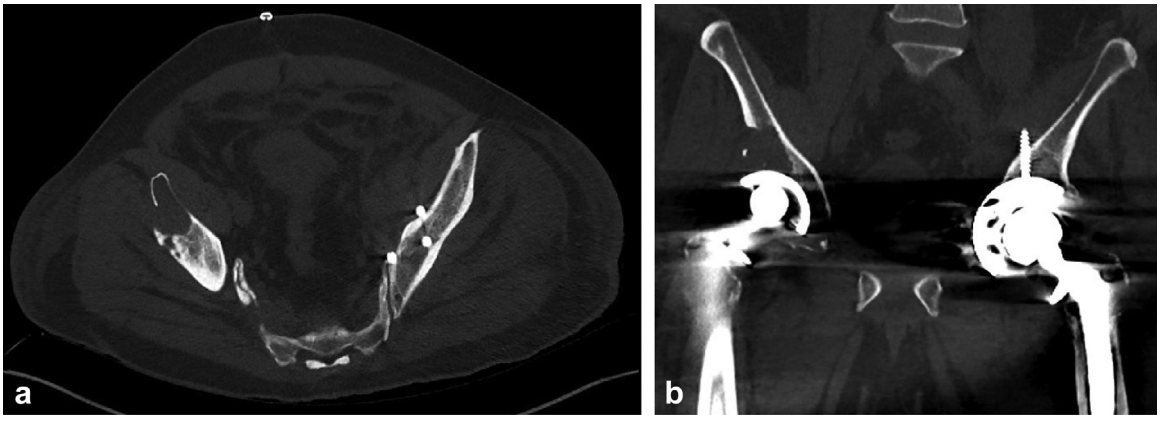

Fig. 6. Postoperative CT of pelvis demonstrating intrapelvic placement of iliac screws on axial (a) and coronal slices (b).

screws <25 mm. Caution must still be used when placing screws into the safe zone, as the superior gluteal vessels and sciatic nerve are located beyond the safe zone. However, in cases of revision THA, distorted anatomy and significant bone loss and deficient bone quality often require placement of screws outside the safe zone. When this is the case, care must be taken to drill safely and secure screws within bone.

Multiple studies have investigated anatomic variability in the iliac vein tree within the pelvis [18,24,25]. In a study using computed tomographic angiography, Kawasaki et al found that the EIV was closer to the pelvis than expected, especially on the left side. In 36 of their 100 patients, the vein rested directly on the inner table of the pelvis [25].

In a study on vascular injuries associated with total hip arthroplasty, Shoenfeld et al [4] found that vascular injuries more often occurred on the left side (66%) and during primary cases (39%). In their study, the EIV was most commonly injured, often due to cement extrusion into the iliac venous system. Overall, their study demonstrated a 7% mortality rate and 15% of patient required some level of amputation.

One study on high-risk patients found that the use of a preoperative screening protocol and aggressive vascular intervention once an injury was identified led to limb salvage in all patients [1]. The authors concluded that thrombectomy alone, which had a 28% success rate, might not be sufficient. More aggressive measures, such as bypasses, were needed for adequate revascularization, and completion arteriography should be performed after bypass to rule out any underlying intimal flap.

Summary

To our knowledge, this is the first case of EIV disruption presenting acutely as phlegmasia cerulea dolens following revision total hip arthroplasty. EIV disruption was likely caused by intrapelvic screw placement and/or extrusion of bone graft through the medial acetabular wall. This case was further complicated by abdominal compartment and leg compartment syndromes. Overall, vascular injury during THA is a rare but potentially devastating complication. Prompt diagnosis and vascular intervention have demonstrated improved outcomes. Overall mortality rate has been reported to be 7%, with a 15% incidence of major amputation (above the ankle joint) and 4% incidence of minor amputation (portion of the foot) [4]. Orthopaedic vascular injuries often result in legal action, with approximately 50% of patients initiating litigation against the surgeon [5]. Thus, despite the rare occurrence of vascular injury, the consequences are severe enough to warrant discussion in the informed consent process.

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