Pseudoaneurysm rupture with hemorrhagic shock in a patient with periprosthetic hip joint infection A case report

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Abstract

Rationale: Although pseudoaneurysm (PA) formation following primary and revision total hip arthroplasty (THA) is rare, PA rupture may lead to severe complications that can result in a threat to life and limb.

Patient concerns: A 65-year-old man presented with acute hemorrhagic discharge for one day from the chronic hip sinus secondary to revision THA that had been performed 6 years ago, for which he had received multiple courses of debridement, antibiotics, and implant retention procedures owing to periprosthetic joint infection (PJI).

Diagnoses: Radiographs showed septic loosening of both the femoral and acetabular components, with medial migration of the component beyond Kohler's line. Contrast-enhanced computed tomography angiogram of the abdomen and pelvis of the patient demonstrated a large PA of the right external iliac artery (EIA), measuring 6.1 cm × 7.7 cm in diameter and 9.1 cm in length.

Interventions: A ball-shaped antibiotic-loaded cement spacer (ALCS) was used to tamponade a bleeding PA, treat the coexisting PJI, and thus facilitate endovascular stent-graft repair (ESGR) later on.

Outcomes: The ESGR resulted in complete exclusion of the PA and successfully controlled the bleeding. The patient underwent a successful revision THA 6 months after stent insertion. Neither stent-graft infection nor recurrent PJI were detected at 18 months.

Lessons: Given the potential of a PA for causing significant morbidity and mortality, the surgeon should have an elevated index of suspicion in the presence of intrapelvic migration of the acetabular component. While facing a PA rupture with/without hemorrhagic shock in patients with coexisting hip PJI, ALCS ball implantation and subsequent ESGR might be an effective method to save the patient's life and limb.

Abbreviations: ALCS = antibiotic-loaded cement spacer, BP = blood pressure, EIA = external iliac artery, ESGR = endovascular stent-graft repair, PA = pseudoaneurysm, PJI = periprosthetic joint infection, THA = total hip arthroplasty.

Keywords: endovascular, hemorrhagic shock, periprosthetic joint infection, pseudoaneurysm, total hip arthroplasty

1. Introduction

Pseudoaneurysm (PA), a lesion arising from a disruption in arterial wall continuity, is a rare but serious complication in primary and revision total hip arthroplasty (THA).^[1,2] Coexisting periprosthetic joint infection (PJI) may result in further aneurysmal degeneration of the arterial wall, complicating this condition and worsening its prognosis.^[3] The PA is unstable and prone to rupture because of poor support of the PA wall. Once PA

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Received: 28 February 2018 / Accepted: 18 May 2018 http://dx.doi.org/10.1097/MD.000000000011028 rupture occurs it can have serious consequences for both patients and doctors.^[2] We present here a case of spontaneously leaking PA with hemorrhagic shock in a patient with chronic hip PJI, which has never been reported in the literature.

2. Case presentation

In 2016, a 65-year-old man presented to the emergency department with acute hemorrhagic discharge for 1 day from the chronic hip sinus secondary to revision THA that had been performed 6 years ago, for which he had received multiple courses of debridement, antibiotics, and implant retention procedures owing to chronic hip PJI. Radiographs showed septic loosening of both the femoral and acetabular components, with medial migration of the component beyond Kohler's line (Fig. 1A). The patient appeared ill and was lethargic but cooperative on physical examination. He had a temperature of 35.7°C, heart rate of 103 beats/min, respiratory rate of 20 breaths/min, and blood pressure of 72/46 mm Hg. Physical examination showed extensive blood discharge from the sinus during active and passive hip motion. The extremities were neurologically intact, with palpable distal pulses. Laboratory investigations yielded the following values: hemoglobin, 10.0 g/dL; hematocrit, 31.8%; white blood cell (WBC) count, 22.8×10^3 cells/ µL (61.5% neutrophils, 33.0% lymphocytes, and 4.5% monocytes); platelet count, 507×10^3 platelets/µL; prothrombin time, 10.5 s (international normalized ratio, 1.02); partial thromboplastin time, 24.3 s (normal control, 29.2 s); serum glucose, 169 mg/dL;

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Figure 1. (A) Anteroposterior hip radiograph shows septic loosening of both the femoral and acetabular components, with medial migration of the component beyond Kohler's line. (B) Contrast-enhanced computed tomography angiogram reveals a large pseudoaneurysm of the right external iliac artery (white arrow) with a diameter of $6.1 \text{ cm} \times 7.7 \text{ cm}$ and length of 9.1 cm. A huge ball-shaped antibiotic-loaded cement spacer (asterisk) in situ is used not only as a pressure-packing filler for provisional bleeding control but also as an antibiotic carrier for sustained antibacterial treatment.

serum creatinine, 1.60 mg/dL; aspartate aminotransferase, 31 U/L (reference, 0-34 U/L); serum sodium, 133 mEq/L; serum potassium, 3.8 mEq/L; and C-reactive protein, 50.4 mg/dL (normal range, 0-5 mg/dL). After hemodynamic stabilization with fluid resuscitation and blood transfusion, the patient was brought to the operating room for debridement, removal of the infected hip prosthesis, and surgery to control the bleeding, followed by definitive vascular reconstruction.

An anterolateral approach was adopted to treat the hip lesions. The femoral component was confirmed to be grossly loose and was removed without difficulty. While attempting to remove the loose antiprotrusio cage, pulsatile bleeding along with an evident drop in blood pressure (BP) was encountered from behind the component. A ball-shaped antibiotic-loaded cement spacer (ALCS) was then prepared to replace the cage, and to be used not only as a pressure-packing filler for provisional bleeding control but also as an antibiotic carrier for sustained antibacterial treatment. Thereafter, the patient remained hemodynamically stable; however, pulsatile bleeding with a marked drop in BP was

again encountered once the ALCS ball was removed, and the source of arterial bleeding was not accessible via the defect in the medial acetabular wall. Then, the wound was provisionally closed with the ALCS ball in situ. At this point, total blood loss was estimated to be 2,500 mL, and the repeat hemoglobin level and platelet count were 8.9 g/dL and $69 \times 10^3 \text{ platelets/}\mu\text{L}$, respectively. Cultures of the excised vascular tissue grew the rare bacteria *Stenotrophomonas maltophilia*.

Contrast-enhanced computed tomography angiogram of the abdomen and pelvis of the patient demonstrated a large PA of the right external iliac artery (EIA), measuring 6.1 cm × 7.7 cm in diameter and 9.1 cm in length (Fig. 1B). After consultation with experts in vascular surgery, a decision was made to control hemorrhage using endovascular techniques. Endovascular stentgraft repair (ESGR) was performed under general anesthesia and in an operating room equipped with C-arm fluoroscopy. Pelvic angiography confirmed a PA of the right EIA with active bleeding (Fig. 2A). Thereafter, the right superficial femoral artery was surgically exposed, and a Gore Viabahn Endoprosthesis (W.L. Gore and Associates, Inc., Flagstaff, AZ) (8 mm × 100 mm) was implanted to exclude the PA by overlapping the stent graft. The ESGR resulted in complete exclusion of the PA and successfully controlled the bleeding (Fig. 2B). The post-ESGR course was uneventful. All arterial pulsations in the foot were present. At 2 weeks after ESGR, the wound oozing ceased, and the follow-up hemoglobin level, platelet count, WBC count, and C-reactive protein were 11.0 mg/dL, 357×10^3 platelets/µL, 6×10^3 cells/µL (64% neutrophil), and 27.5 mg/dL, respectively. The patient underwent a successful revision THA 6 months after stent insertion (Fig. 3). Neither stent-graft infection nor recurrent PJI were detected at 18 months.

This report was approved by the ethic committee (Institutional Review Board) of the Chang Gung Memorial Hospital in Taiwan (Reference number: 201701864B0). We did not obtain informed consent from the patient due to a statement of this committee, that analyzing patient data retrospectively requires no informed consent.

3. Discussion

PA, a lesion arising from a disruption in arterial wall continuity, is a rare but serious complication in hip arthroplasty.^[1,2] Although the EIA is the most frequently affected owing to its proximity to the acetabular roof, the common femoral artery and its principal branches are also at risk.^[1] Under the influence of sustained blood pressure, blood leaks through the vessel wall and forms a perfused sac between the 2 outer layers of an artery, the muscularis propria and the adventitia, which communicates with the arterial lumen.^[2] Owing to poor support of the PA wall, the risk of rupture is higher than that of a true aneurysm. Morbidity and mortality rates from a PA are high, with an estimated 11% incidence of limb loss and a 6% of mortality rate.^[1] When vascular repair had to be performed as an emergency procedure, the mortality rate increased to 33%.^[4] Coexisting infection may result in further aneurysmal degeneration of the arterial wall, complicating this condition and worsening its prognosis.^[3] Given the potential of a PA for causing significant morbidity and mortality, the surgeon should have an elevated index of suspicion in the presence of high-risk factors. Previous studies have demonstrated that revision surgery, female sex, left-sided as opposed to right-sided surgery, and intrapelvic migration of the acetabular component with or without PJI are associated with an increased risk of vascular injury.^[1,5]



Figure 2. (A) Intraoperative fluoroscopic pelvic angiography shows a pseudoaneurysm of the right external iliac artery (black arrowhead). There is brisk contrast medium leak from the center of the external iliac artery (black arrow). (B) Angiography after endovascular stent graft repair shows complete exclusion of the pseudoaneurysm and good angiographic flow through the stent graft (black arrowhead). Note the ball-shaped antibiotic-loaded cement spacer in situ (asterisk).

PAs after hip arthroplasty commonly develop subclinically; although they may be present in the acute phase of surgery, their presentation is often delayed and has been reported up to 16 years following surgery.^[6] They are usually detected incidentally during surgery or during radiographic studies of other conditions, unless infection, local compression on neurovascular structures, or rupture occurs.^[2] PJI may result in a destructive osteolytic process, with dissolution of the interfaces and progressive loss of bone structure, thus facilitating the protrusion of acetabular prosthesis. In fact, intrapelvic migration of the acetabular component itself has



Figure 3. Anteroposterior radiograph of the pelvis shows a custom-made hip prosthesis after definitive right revision total hip arthroplasty.

been associated with PJI.^[7] The development of the PA might have been secondary to the underlying infection or mechanical injury of the arterial wall by the protruding acetabular components, screws, osteophytes, or by cement that has been leaked into the pelvis through a breach of the acetabular cortex.^[7–11] The infected PA is unstable and highly prone to rupture, which is the most serious cause of morbidity and mortality from PA.^[2] The current case, to our knowledge, might be the first reported case of a spontaneously leaking infected PA with blood discharging through a chronic hip sinus, and hemorrhagic shock. Emergency bleeding control surgery was required to save this patient.

Control of the massive hemorrhage by using an ALCS ball was the key in this case. The size of the infected PA and the difficult access for proximal vascular control via the defect in the medial acetabular wall prompted the use of such a pressure-packing filler. This allowed time for patient stabilization, computed tomography angiogram, and definitive vascular reconstruction. The ALCS ball not only functioned as a pressure-packing filler for provisional bleeding control, but also was utilized for direct local antibiotic delivery for sustained infection control. Alternatively, a Sengstaken-Blakemore tube had ever been used to effectively tamponade a bleeding PA but not valid for treatment against PJI while awaiting definitive vascular reconstruction.^[12] In general, there are 3 main options for definitive vascular reconstruction of an infected PA: open approach with in situ or extra-anatomic bypass grafting, endovascular embolization, and ESGR.^[13] When the perfused sac of an infected PA communicates with the external milieu, massive bleeding occurs, increasing the risk of death. This emergency condition has conventionally been treated with open surgery, which carries a high morbidity and mortality rate.^[4,11] Owing to its low invasiveness, ESGR has been proposed as an alternative method of management, not only as a temporary measure before interval surgical repair but also as a definitive procedure.^[11,14] Despite concerns about the feasibility and safety of inserting a covered endovascular stent into an infected vessel,^[15] successful treatment of an infected PA of EIA with ESGR in the presence of chronic infection has been reported.^[11-14] Our patient

was at high operative risk for open vascular reconstruction, and this successful outcome further supports the effectiveness of ESGR, even in the presence of chronic PJI and during life-threatening hemorrhage.

4. Conclusion

Emergent vascular repair and debridement was required for such a patient who suffered from PJI with spontaneously leaking PA and hemorrhagic shock. Here we show that an ALCS ball could be used to effectively tamponade a bleeding PA, treat the coexisting PJI, and thus facilitate ESGR later on.

Author contributions

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References

- Shoenfeld NA, Stuchin SA, Pearl R, et al. The management of vascular injuries associated with total hip arthroplasty. J Vasc Surg 1990;11:549–55.
- [2] Saad NE, Saad WE, Davies MG, et al. Pseudoaneurysms and the role of minimally invasive techniques in their management. Radiographics 2005;25(Suppl 1):S173–89.

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- [5] Macbeth GA, Rubin JR, Mchityre KEJF, et al. The relevance of arterial wall microbiology to the treatment of prosthetic graft infections: graft infection vs. arterial infection. J Vasc Surg 1984;1:750–6.
- [4] Richardson JW, Greenfield LJ. Natural history and management of iliac aneurysms. J Vasc Surg 1988;8:165–71.
- [5] Wera GD, Ting NT, Della Valle CJ, et al. External iliac artery injury complicating prosthetic hip resection for infection. J Arthroplasty 2010;25:660.e1–4.
- [6] Smith GH, Nutton RW, Fraser SC. Iliac artery pseudoaneurysm rupture following excision of an infected hip prosthesis. J Arthroplasty 2011;26:977.e13–5.
- [7] Stiehl JB. Acetabular prosthetic protrusion and sepsis: case report and review of the literature. J Arthroplasty 2007;22:283–8.
- [8] Nachbur B, Meyer RP, Verkkalla K, et al. The mechanisms of severe arterial injury in surgery of the hip joint. Clin Orthop Relat Res 1979;122–33.
- [9] Hopkins NF, Vanhegan JA, Jamieson CW. Iliac aneurysm after total hip arthroplasty. Surgical management. J Bone Joint Surg Br 1983;65:359-61.
- [10] O'Toole GC, Madhavan P, Shanik G, et al. Purulent bloody drainage after infected total hip arthroplasty: a sign of a leaking mycotic aneurysm. J Arthroplasty 2004;19:391–4.
- [11] Warren MJ, Fabian S, Tisi P. Endovascular PTFE-covered stent for treatment of an external iliac artery pseudoaneurysm in the presence of chronic infection. Cardiovasc Intervent Radiol 2007;30:770–3.
- [12] Makar RR, Salem A, McGee H, et al. Endovascular treatment of bleeding external iliac artery pseudo-aneurysm following control of hemorrhage with Sengstaken tube during revision total hip arthroplasty. Ann R Coll Surg Engl 2007;89:W4–7.
- [13] Sawbridge D, O'Connor OJ, MacEneaney P, et al. Successful endovascular treatment of an infected external iliac pseudoaneurysm with hemorrhage at total hip arthroplasty. J Vasc Interv Radiol 2010;21:1135–6.
- [14] Clarke MG, Thomas HG, Chester JF. MRSA-infected external iliac artery pseudoaneurysm treated with endovascular stenting. Cardiovasc Intervent Radiol 2005;28:364–6.
- [15] Ishida M, Kato N, Hirano T, et al. Limitations of endovascular treatment with stent-grafts for active mycotic thoracic aortic aneurysm. Cardiovasc Intervent Radiol 2002;25:216–8.