

CASE REPORTS

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Urethral pseudoaneurysm life-threatening bleeding: a case report and literature review of etiological and treatment options

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Abstract

Background: Pseudoaneurysm (PA) of pudendal artery's distal branches is an extremely rare clinical scenario. We report a case of life-threatening urethrorrhagia due to PA of the internal pudendal artery (IPA).

Case presentation: A 35-year-old man presented with massive urethral bleeding after traumatic urethral catheterization. Failed attempts of conservative management necessitate an emergency intervention with angiography which shows an active extravasation from the left IPA, which was embolized with gel foam, and complete hemostasis was achieved.

Conclusion: PA of the pudendal artery's distal branches is serious, and majority of cases need an emergency intervention. CT angiography is the best diagnostic imaging, and angioembolization of the bleeding site is considered the first-line treatment. Other treatment options such as US-guided compression, endoscopic coagulation, and open surgery are alternative treatments.

Keywords: Urethrorrhagia, Urethral bleeding, Angioembolization

1 Background

Continuous or intermittent active urethral bleeding independent of urination is defined as urethrorrhagia, which is an uncommon urological condition [1]. Pseudoaneurysm (PA) of the pudendal artery's distal branches is a very rare but devastating disorder. It can result following traumatic catheterization with urethral false passage formation [2–5], iatrogenic urethral injury following internal optical urethrotomy [6], straddle injury [7], and traumatic pelvic fracture [1]. Bleeding in such condition may be massive (> 1000 ml), associated with rapid hemoglobin (Hb) level drop, and the patient becomes hypovolemic requiring an emergency intervention [7].

We present a rare case of massive urethrorrhagia due to internal pudendal artery (IPA) PA caused by iatrogenic traumatic urethral catheterization that was managed successfully by an angioembolization with gel foam.

2 Case Presentation

A 35-year-old male complaining of hypertension, anemia, and end-stage renal disease presented to the ER with complaints of shortness of breath for three days and generalized body swelling. Patient was initially unstable, so he was admitted to the ICU. However, during urethral catheter insertion there were traumatic urethral injury and profuse urethral bleeding. Urology on call-doctor was called, and urethral catheter was fixed properly and with perineal compression bleeding stopped.

On the 3rd day before discharge and after urethral catheter removal, bright-red severe urethral bleeding restarted again. A urology doctor was consulted,

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he inserted a 3-way 20F Foley's catheter with good urethral lubrication, traction on thigh, and manual perineal compression was performed for the patient with close monitoring and observation. Urine in bag became clear; however, continuous massive urethral bleeding came out around the catheter that stopped only with penile compression and bandage that was placed for 24 h. After removal of the penile bandage, there was massive re-bleeding again from the urethral meatus around the catheter. Urgent CT angiography of the pelvis was done and showed an active extravasation of contrast in the corpus spongiosum/periuethral area which is due to PA in posterior aspect of the penile urethra (Fig. 1). The patient's Hb levels dropped steadily from 7 to 5.1 over the past 48 h, and he received three packed RBCs in the ICU. The patient became hypotensive and tachycardic. Following

an emergency multidisciplinary meeting, discussing with the patient treatment options available, an agreement and informed consent from the patient were obtained to do a pelvic angiography that was conducted under ultrasound and fluoroscopic guidance to gain access to the right femoral artery. Angiogram of the left internal iliac artery revealed active extravasation of the left IPA, which was embolized with gel foam, and complete hemostasis was achieved (Fig. 2).

The patient tolerated the treatment well and had no complications during or after the embolization. The urethral bleeding stopped at the end of the day after the source of bleeding was embolized, and the Hb level remained stable in the days after the treatment. After a four-month follow-up in the OPD, the patient was voiding normally and had normal erectile function and sexual

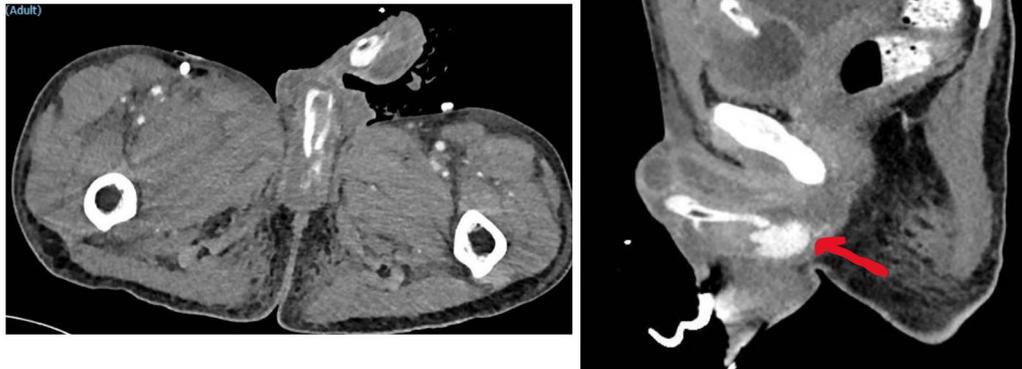


Fig. 1 CT angiography (axial and sagittal cuts) of the pelvis showing an active contrast extravasation in the corpus spongiosum/periuethral area, which is due to pseudoaneurysm in the posterior aspect of penile urethra (red arrow)

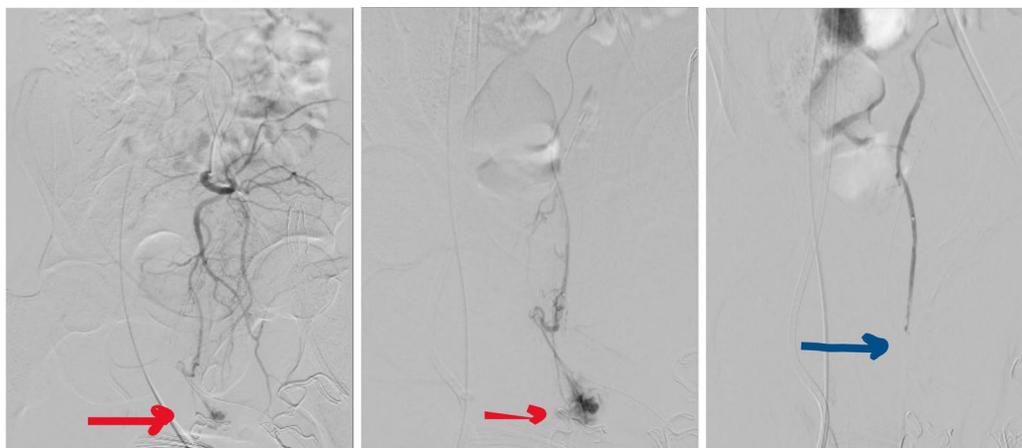


Fig. 2 Angiography confirmed a left internal artery pseudoaneurysm, which was successively treated by embolization: An active extravasation of contrast (red arrow) postembolization angiography revealed cessation of flow to the pseudoaneurysm (blue arrow)

activity. The manuscript report adheres to CARE guidelines for reporting of case reports.

3 Discussion

External trauma is the commonest cause of urethral injuries that primarily affects men. Clinically the patient is presented by urethrorrhagia, penis ecchymosis, and scrotal/perineal hematoma. The majority of urethral bleeding is small and resolves spontaneously or with conservative treatment including external perineal compression, gentle urethral catheter insertion, antibiotics, close monitoring, stop anticoagulation, and antiplatelet therapy [1].

Massive urethrorrhagia due to PA formation is an extremely rare but devastating event and can lead to hypovolemic shock if one of the pudendal artery's distal branches is involved, requiring emergency intervention [1–7]. Different etiological mechanisms have been described for the formation of pseudoaneurysms including traumatic pelvic fracture [1], straddle injury [7], and iatrogenic urethral injury during difficult urethral catheterization [2–5] or after endoscopic treatment of urethral stricture [6]. The most common involved arteries are the internal pudendal artery [1, 4–7], the bulbar artery [2], and the obturator artery [3]. Pseudoaneurysm diagnosis is usually made by radiological imaging such as Doppler ultrasound, angio-computed tomography, or angiography [1–7].

Literature data about this critical condition are sparse, and thus, protocol of management has been based on surgeons' experience. If conservative therapy is failed and bleeding persists, there are different therapeutic options either by angiography, open, or endoscopic therapy.

Angioembolization is the first-line therapy for arterial bleeding as usual. Previous reports have shown that it is safe, feasible, and effective in controlling severe urethral bleeding due to rupture pseudoaneurysms [1–5]. On the other hand, Attri et al. reported a successful unique management technique of US-guided compression repair for bulbourethral artery pseudoaneurysm following optical internal urethrotomy. They recommended before going for invasive procedure, and this simple technique can be tried specially in small pseudoaneurysm [6]. Furthermore, urethroscopy and direct transurethral coagulation of the bleeding artery in the bulbar urethra have been reported by Yuki and colleagues. In their case, the penile artery was terminal and narrow and they prefer endoscopic management directly as super-selective embolization for targeted artery would be difficult [8]. It is worth to mention that bilateral internal iliac artery ligation for management of massive recurrent urethral bleeding following urethral dilatation has been reported successfully [9]; however, in the era of minimally invasive therapeutic

options such as angiography and endoscopy open surgery can be last option if all previous methods failed.

4 Conclusion

Massive life-threatening urethrorrhagia due to PA of the pudendal artery's distal branches is very rare but devastating condition and needs emergency intervention. CT angiography is the best diagnostic imaging, and angioembolization of the bleeding site is considered the first-line treatment. Other treatment options such as US-guided compression, endoscopic coagulation, and open surgery are alternative treatments.

Abbreviations

PA: Pseudoaneurysm; IPA: Internal pudendal artery.

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Author contributions

DT: contributed to evaluation and treatment of index patient, study design, and drafting the manuscript. AH: contributed to literature search, scoping review and data acquisition. SB: contribute to evaluation and treatment of index patient, study design, and critical review of the draft manuscript. MO: contributed to study design, literature search, and drafting the manuscript. IA: contributed to literature search, scoping review and data acquisition. AAR: contributed to evaluation and treatment of index patient, study design, scoping review, drafting the manuscript, literature search, and drafting the manuscript and data acquisition. All authors read and approved the final manuscript.

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Consent for publication

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Competing interests

The authors declare that they have no competing interests.

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