Massive Hematuria and Shock Caused by Ilio-Ureteral Fistula in a Patient with an Isolated Internal Iliac Artery Aneurysm

Takeshi Soeda, MD, Yuhei Saito, MD, Shuji Setozaki, MD, and Hisao Harada, MD

An emergent operation was performed on a 73-year-old woman with massive hematuria and serious shock. A computed tomography (CT) revealed that the cause of the shock was hemorrhage from an aneurysm into the ureter, with resultant massive hematuria. During surgery, we observed that the ureter was encased into the wall of the aneurysm, with exposure of the pre-positioned ureteric stent inside the aneurysmal space. Reconstruction of the ureter was performed by wrapping the tissues with the ureteric stent inside. Postoperative recovery was uneventful, and CT angiography showed complete exclusion of the right internal iliac artery with the in situ ureteric stent.

Keywords: massive hematuria, ureter, iliac artery aneurysm

INTRODUCTION

An isolated iliac artery aneurysm can cause serious problems since it grows unnoticed deep within the pelvic space and sometimes becomes large enough to rupture spontaneously or affect nearby organs, such as the bowels, the urinary system, veins, and gynecological organs via progressive compression or invasion.1 Here, we report the invasion of the ureter by an aneurysm, finally resulting in a massive hematuria.

CASE

A 73-year-old woman with recent relapsing fever was admitted to our hospital for suspected pyelonephritis and sepsis. Her past history documented mixed collagen diseases, and 2 mg of oral betamethasone had been administered daily. At admission, a plain CT revealed a mass, 62 mm in diameter, inside the pelvis (Fig. 1A) compressing the right ureter to cause hydronephrosis. While her hemodynamics was improving, ureteric stenting was performed to alleviate urinary retention and infection. Enterococcus faecium was revealed to be the causative agent by urine and blood culture, and antibiotic therapy was continued. Two weeks after admission, since an enhanced CT definitively demonstrated the pelvic mass to be a huge internal iliac artery aneurysm, 68 mm in diameter, (Fig. 1B), an operation was then considered to avert rupture of her rapidly growing aneurysm. However, difficulty in controlling the urinary tract infection deferred the surgical intervention. About a month later, the sudden onset of massive hematuria and serious shock necessitated an emergent operation, because a heavy clot formation identified by CT was at this time thought to be produced by aneurysmal rupture directly into the bladder (Fig. 2). The operation was performed through a right extra-peritoneal approach. While tracing upstream along the external iliac artery with simultaneous exposure of the aneurysm, the proximal neck of the aneurysm was successfully ligated. After an aneurysmectomy, suture closure of the distal exit branch orifices was performed by an endoaneurysmal route. Inside the exposed aneurysmal space, the encased right ureter was revealed to have a broad fistulous communication with the lumen of...
erosion, and infection in situ or indirectly to remote organs can all take place with a poor prognosis even if an operation is attempted.\textsuperscript{3)} Among the various complications, the urinary tract organs, especially the ureter, seem to be the most susceptible to obstruction by aneurysmal compression. Sometimes the ureter can be incorporated into the aneurysmal wall (so called encasement) frequently producing urinary tract infection and bacteremia, and micro-

the aneurysm. Reconstruction of the urinary route was performed by wrapping it as well as filling the space with a recto-abdominal muscle flap to prevent abscess formation or urine leakage (Fig. 3). She was doing well and was discharged 27 days after the operation. However, she required periodic replacement of the ureteric stent at an outpatient clinic thereafter.

\section*{Discussion}

Concerning the possibility of spontaneous rupture, surgical intervention for iliac artery aneurysms exceeding 30 mm in diameter has unanimously been recommended.\textsuperscript{2) However, these aneurysms are located deep within the pelvis which enables their latent growth. As a result, they sometimes become big enough to affect adjacent organs by the time they become symptomatic or are detected incidentally through imaging. In such situations, the vulnerable organs include the iliac venous channels, colon, urinary system, gynecological organs, and pelvic nerves. Occlusion, rupture, fistula formation,
Massive Hematuria Caused by Ilio-ureteral Fistula

Massive Hematuria Caused by Ilio-ureteral Fistula

achieved by ligating its neck, plus obliteratorative endoan-
eurysmorrhaphy. With regards to the ilio-ureteric fistula,
this case required additive surgical procedures. Since
natural repair of the ureter was difficult, an alternative
method of reconstruction was applied using plastic surgery
in addition to the influence of the pulsatile movement of the
aneurysmal wall.7) Whatever the cause, the bleeding
resulted from the fistula, which put the patient in a state
of serious shock. Prolonged control of the infection
should not have been prioritized given the potential for
fatal complications.

Endovascular therapy seems a promising technique
for iliac aneurysms.8) However, while exclusion of the
aneurysm is critical, it is also important to block back-
flow from the distal branches. Another drawback of per-
manent stenting in this particular case is the patient’s
tendency toward chronic bacteremia that risks infection
during routine and emergent operations related to
deployed devices.7) On the other hand, an iliac stent or
balloon could be used as an effective temporary solution
before emergent open surgery. However, as described in
the literature,7) only the presence of a ureteric stent itself
can have a local hemostatic effect, as in our case, with
tolerable hemodynamics. Thus, we were able to perform
the emergent operation without the need for additional
endovascular techniques.

Surgical therapy has already been well-established
in complex and symptomatic cases9) and should be an
absolute indication in lesions with accompanying infec-
tion. The extra-peritoneal surgical approach without the
use of artificial vessels seemed, in our case, accessible
and less invasive. Decompression of the aneurysm was
achieved by ligating its neck, plus obliteratorativeendo-
eurysmorrhaphy. With regards to the ilio-ureteric fistula,
this case required additive surgical procedures. Since
natural repair of the ureter was difficult, an alternative
method of reconstruction was applied using plastic surgery
to maintain the route of urinary flow. Aggravation of
the infection was less likely at the repair site. As a result,
the urinary stent became indispensable in this case.

In conclusion, huge isolated internal iliac artery aneu-
rysms with significant compression of the adjacent organs
should alert one to various catastrophic outcomes. Our
specific case of irreversible ureter involvement and even-
tual rupture resulting in massive bleeding is therefore
presented as an example of this serious condition requir-
ing early surgical intervention.

DISCLOSURE STATEMENT

The authors disclose no conflict of interest.

REFERENCES

1) Dix FP, Titi M, Al-Khaffaf H. The isolated internal
iliac artery aneurysm—A review. Eur J Vasc Endo-
3) Parry DJ, Kessel D, Scott DJ. Simplifying the internal
83: 302-8.
4) Krupski WC, Selzman CH, Floridia R, et al. Contem-
porary management of isolated iliac aneurysms. J Vasc
5) Grime PD, Wilmshurst CC, Clyne CA. Spontaneous
iliac artery aneurysm-ureteric fistula. Eur J Vasc Surg
6) Thiry AJ, Struyven J, Van DeCasseye M. Spontaneous
rupture of right iliac arterial aneurysm into ureter.
7) Darcy M. Uretro-arterial fistulas. Tech Vasc Interv
management of isolated iliac artery aneurysms. J Vasc
9) Brin BJ, Busutil RW. Isolated hypogastric artery