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Letter

Arterioureteral fistula: An unusual cause of haematuria 10 years after the implantation of a synthetic iliac-femoral stent

Dear Editor,

The patient had a history of intermittent self-resolving episodes of gross haematuria and was referred to our department for a massive bleeding. At presentation, haemoglobin was 8.6 g/dL and creatinine was 1.37 mg/dL. He was hemodynamically stable, but suffered an important anemization in the subsequent days requiring multiple blood transfusions. In his medical history, it was reported that he had type II diabetes mellitus and the implantation of a right iliac-femoral stent 10 years before.

After bladder catheterization and irrigation, computed tomography (CT) scan was performed. No urological malignancies or other findings apparently explaining haematuria were detected. CT scan and angiography failed to demonstrate extravasation of contrast agent in the ureter. Of note, CT scan showed an oedematous right ureter at the crossing with common iliac artery precisely at the level of the proximal extremity of the vascular stent (Fig. 1). A flexible cystoscopy showed an active bleeding from the right ureteral meatus. In order to confirm the cause of bleeding, the patient underwent ureterorenoscopy under general anaesthesia. During the operation, an oedematous, erythematous, and pulsating area in the mucosa of the iliac ureter, was consistent with an iliac arterioureteral fistula (AUF). A 6 Charrière × 26 cm double-J ureteral stent was positioned as the patient preoperatively refused an eventual nephrostomy placement. Subsequently the vascular surgeon placed an iliac endovascular stent (Endurant™ Stent Graft System, Medtronic Inc., Minneapolis, MIN, USA) to exclude the fistulous communication. In addition to endovascular stent placement, the internal iliac artery was embolised using three 7-mm coils. Neither postoperative complications (Clavien-Dindo 0) nor further episodes of haematuria in the perioperative period were reported. The ureteral stent was removed 1 month later in order to guarantee an adequate healing of the ureteral lesion. After 1 year follow-up, the patient had no further episodes of haematuria. Unfortunately, he developed a right ureteral stricture and was proposed nephrostomy to avoid the risk of a further AUF secondary to ureteral stenting. The patient again refused the nephrostomy and received a ureteral stent. He required periodical ureteral stent replacement, but did not present new episodes of haematuria.

AUF is an uncommon and challenging clinical scenario involving skills and experience of urologist, vascular surgeon, and interventional radiologist. As reported in a recent review by Kamphorst et al. [1], 470 cases in 445 patients were described in literature: despite its low incidence, nowadays AUF maintains a 7% mortality rates. As in the majority of patients, the only symptom is haematuria, a presentation common to various urological disorders [2]; AUF is a challenging condition to recognize and often the diagnosis is delayed or even missed leading to serious and potentially lethal complications like haemorrhagic shock [3].

Secondary AUFs account for 97.5% of cases [1], can develop after pelvic oncologic surgery (usually urologic or gynaecologic) [4], and often are in association with pelvic irradiation causing retroperitoneal fibrosis. Other surgical procedures that may be associated with AUF are ureteral stenting and, as described in our case, vascular surgery with synthetic grafting [5]. The remaining 2.5% of AUFs can be classified as primary and are caused by aneurysms, pseudoaneurysms, or arteriovenous malformations [1].

The exact pathophysiology behind AUFs is not completely understood: probably external radiotherapy and surgical procedures lead to vasa vasorum disruption, alterations of media, and adventitia of the arteries and ureteral ischemia. The result is an eroding inflammatory process with fibrosis, adhesion of the ureter to the iliac vessels (or rarely aorta) and friction between the pulsatile artery and the ureter itself, thus predisposing to AUF development [1,6].

Detecting the presence of an AUF is challenging. Pyelography, ureterorenoscopy, and flexible cystoscopy can easily miss the diagnosis, while provocative angiography can reach a 100% sensitivity rate but may expose the patient to the risk of uncontrollable bleedings [1,7,8]. Of note, even if it permitted to reach a conclusive diagnosis in our case, we do not recommend ureterorenoscopy as a standard procedure as it could further damage the fistula and dislodge a tamponing clot with the risk of a massive haemorrhage.

Although CT scan has been used successfully in previous reports, its sensitivity is very low; in our patient and in line

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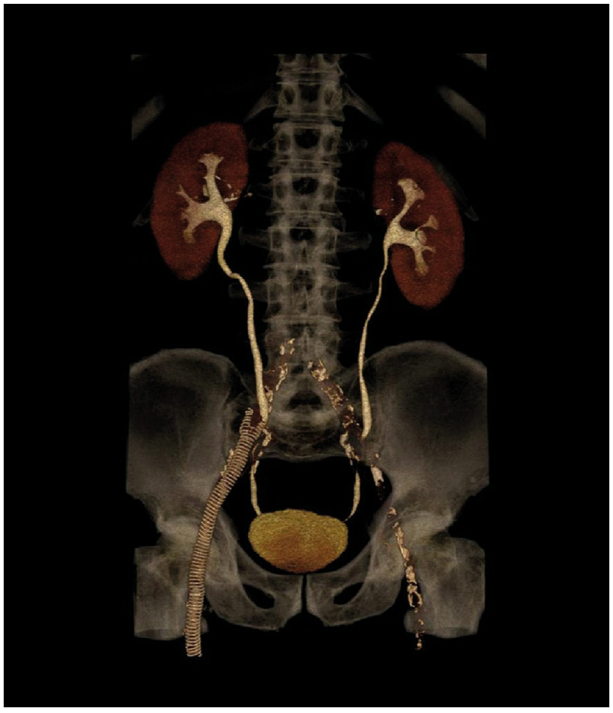

Figure 1 Computed tomography scan showing the proximal extremity of the endovascular stent laying at the crossing with the right ureter.

with previous experiences [9], it showed only indirect and not specific signs of ureteral injury as oedema, failing to identify extravasation of the contrast agent. Of note, the oedema of the right ureter just at the crossing with common iliac artery and precisely at the level of the proximal extremity of the vascular stent, triggered the suspicion of an AUF.

Our patient was successfully treated with the placement of an endovascular stent and a ureteral double-J stent. In this way, we were able to abolish the arterial inflow to the ureter, providing an adequate urinary drainage at the same time.

In the majority of reports, a combined urologic and vascular treatment has been used even if some authors claim that while arterial lesion treatment is mandatory, urological manoeuvres (either endoscopic, percutaneous or surgical) are not always necessary but should be evaluated in relation to the surrounding circumstances, as AUF will close spontaneously once the arterial inflow to the ureter is abolished [6].

According to Van den Bergh et al. [3], endovascular approaches demonstrated optimal results with a 0% AUF-specific mortality rate. Remarkably, if a combined strategy is adopted, after endovascular stent placement nephrostomy may be the preferable choice, even if in patients with discussed risk factors a smaller and softer ureteral stent can be considered [10].

In conclusion, AUF remains a rare but potentially lethal condition that clinicians should be aware of. Our experience shows that in patients with haematuria not otherwise explainable and previous history of vascular surgery with endovascular stent placement, the presence of an AUF should be suspected, especially if CT shows an extremity of the vascular stent laying in close proximity to the ureter.

The present study was carried out according to the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Patient's written informed consent for treatment and publication was obtained. All anamnestic, clinical, and laboratory data containing sensitive information about the patient were de-identified in order to ensure analysis of anonymous data only.

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