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SUCCESSFUL SURGICAL TREATMENT FOR RUPTURE OF AN EXTERNAL ILIAC ARTERY PSEUDO ANEURYSM INTO A URETER AND REVIEW OF LITERATURE.

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Abstract:

Grosshematuria is a manifestation of various urological condition such as Nephrolithiasis, urological malignancy, urinary tract infection and trauma. Most cases ofgross hematuria are manageable and are seldom life-threatening, however, rarer diagnoses can be potentially lethal. Pseudo Uretero-iliac aneurysm fistula (UIAF) are rare and one of such lethal emergency which require a multidisciplinary approach for urgent diagnosis and management. Clinical picture varied from asymptomatic to flank pain, hematuria and shock and condition need urgent diagnosis by angiography, cystoscopy, Computed Tomography of abdomen and sometimes surgical exploration.

We have come through an interesting case of ruptured case of pseudo iliac aneurysmal into ureter. HenceWe will review the causes, risk factors, pathophysiology, clinical presentation, and various approach for diagnosis and treatment and our experience of approach of one such cases of pseudo Uretero-iliac aneurysmal fistula.

Keywords: Gross Hematuria.Uretero-iliac aneurysm fistula (UIAF),Computed Tomography.

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OUR CASE:

40 years male came to our emergency room with gross intermittent haematuria and passage of clots since 15 days. Generalised weakness since 5 days and shortness of breath since 1 day.

Patient had a history of right open ureterolithotomy following failed ureteroscopy and intracorporeal pneumatic lithotripsy for right impacted mid ureteric calculus just one month prior to current admission. Following the procedure patient remain asymptomatic for 15 days but later (after 15 days from the surgery) he developed gross intermittent haematuria, which was managed locally with five units of packed blood transfusion and he was then referred to us.

On examination Blood pressure was 80/50mmHg ,pallor was present pulse was 112per minute, respiratory rate was 26per minute and SpO2 was 90%. Investigation revealed Haemoglobin was of 3.6 gm/dl, WBC count was 12200 /cmm, urea 46 mg/dl, serum creatinine 0.9mg/dl. Patient was resuscitated with multiple unit of blood transfusion, oxygen support. Ultrasound abdomen was done which showed right mild hydroureteronephrosis with urinary bladder clots. Cystoscopy was done in local anaesthesia which showed clot in bladder and profuse oozing of blood from right uretericorifices. Possibility of bleeding from major artery suspected. We then planned computed urography and angiography and urgent exploration. CT urography and angiography revealed 11x9mm aneurysm arising from right external iliac artery, adjacent to and closely related to thickened wall of ureter at the level of its crossing over iliac vessel,(Figure 1 & 2).

Diagnosis of Pseudo aneurysm of right external iliac artery communicating with right ureter(Possibly fistulous communication) was made(figure 1). As we don't have any facility of interventional radiology at our centre and knowing ongoing bleeding and life threatening nature of the condition, patient was taken for urgent surgical exploration and cystoscopy and ureteroscopy for any bladder clot evacuation and placement of double J stent in right ureter.

Under general anaesthesia, Right hockey stick incision was given and retroperitonealy right External iliac artery approached. Due to extensive and dense fibrosis, approach was very difficult and tedious but finally artery was almost mobilised and pseudo aneurysm ligated. Intraoperatively transient loss of arterial pulsation noticed may be due to arterial spasm. Bleeding in ureter stopped immediately . Double J stent was placed in right ureter and bladder clot evacuated. In postoperative period we noticed decrease pulsation in right dorsalispaedis artery . However movement and pain sensation was present in right limb. Gradually a good volume pulsation felt in right dorsalispaedis artery.

Patient was discharged on 10^{th} post operative day. Follow up was done after one month with CT angiography of both lower limbs which showed occlusion at site of Pseudoneurysm and well formed collaterals which preserved lower limb circulation(Figure 3 & 4). There was no bleeding in urine and haemoglobin was 12gm/dl. Patient is ambulatory and doing his all normal physical

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activity. In our case urgent availability of surgical services and presence of ipsilateral collateral ultimately prevented mortality and morbidity. Our case emphasize the importance of emergent availability of any intervention to treat ureteroarterial fistula as this disease is very lethal without treatment. Here we will review ureteroarterial fistula in detail.

Introduction:

Uretero-iliac aneurysm fistula (UIAF) due to Rupture of aneurysm or pseudo aneurysm are very rare but life-threatening causes of gross hematuria. Majority of (Approximately 90%)cases of Uretero-iliac aneurysm fistula (UIAF) have been reported in the English literature; and nearly two thirds of such cases are reported recently [1]. Predisposing factors forUretero-iliac aneurysm fistula (UIAF) include history of major genitourinary surgical procedure,long dwelling stents, and pelvic irradiation [2]. Pseudo Uretero-iliac aneurysm fistula generally occurs following some injury to iliac vesselsespecially during genitourinary surgery. UIA fistulas are seen nowadays because of easy availability of ultrasound and computed tomography. However, diagnosis of UIAF still challenging because of low index of suspicion [3].However, availability of Color Doppler and CT angiography can easily help in early diagnosis of case and also Intervention radiology play significant role in emergency management of these patients. But the lack of interventional radiologist causes high mortality rates. Therefore, a high index of suspicion for cause of intermittent or life threateninghematuria is required for rapid diagnosis and treatment.

Uretero-iliac aneurysm fistula (UIAF) are due to communications between a iliac artery and ureter. The common or external iliac artery is usually involved, but the internal iliac artery and aorta [4] have also been found to be involved rarely. The name of this entity is variable in literature and the terms “arterioureteral fistulas” and “ureteroarterial fistulas” have been used interchangeably [5]. This may be due to two important factors, the direction of flow and the cause of primary etiology. The direction of flow is always from the vessels to the ureter, which is responsible for the primary symptom of hematuria. In addition, because of the emergency associated with first treatment of thearterial parts of fistula and prevent any ongoing blood loss, many authors prefer to use term as arterioureteral fistulas. Others prefer the term “ureteroarterial fistulas” because mostof fistulas between the artery and ureter result from some pathology in ipsilateral ureter. [1]. In our review we will use this entity as ureteroarterial fistulas(UAF)or Uretero iliac aneurysmal fistula(UIAF) throughout this article, although both names are appropriate.

Risk Factors, Causes and Pathophysiology:

Uretero-iliac aneurysm fistula (UIAF)can be broadly classified as either primary or secondary. Primary fistulas are mainly due to inflammation and disease process of artery and account for less than 15%. [6]. Sometimes congenital arteriovenous malformation may be causative. (7)

Majority ofUretero-iliac aneurysm fistula (UIAF) are secondary in nature and generally had a prior history of pelvic surgery, ureteric intervention in the form of chronic double J stenting and

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pelvic radiotherapy. Median period for the onset of symptoms vary with nature of previous intervention. Some study showed that median period from time of intervention to hematuria was 2 years (range, 2 months–30 years) in those who had a pelvic malignancy surgery and 10 years (range, 3 months–25 years) in those who underwent vascular surgery that include use of a synthetic graft. [8]. A review of 23 patients with UIAF [9] found that patients with ureteral stenting developed fistulas in a median time of 4 months (range, 15 days–12 years). In a recent literature review of 80 cases of arterioureteral fistulas, Bergqvist et al. found that 42% of patients with arterioureteral fistulas had some type of urinary diversion surgery. [1].
Three cases ofarterioureteral fistulas related to gravid uterus are reported [10] in literature; all three patients presented with gross hematuria, urinary tract infection and sepsis. Two of these patients died of shock due to bleeding; the third patient died of complications related to sepsis. In each case, the diagnosis was made postmortem. It is unclear how the patients' pregnancies contributed to the development of fistulas, but the pregnancies led to urinary retentions, which required stents placements in two patients. Fistulas due to gravid uterus arenowadays rare due to wide prevalent use of soft catheters. flexible stents and effective treatment for urinary tract infections.

Some rare causes of Uretero-iliac aneurysm fistula (UIAF)include presence of small ureteric stump following nephrectomy; pressure necrosis at ureteroneocystotomy, retroperitoneal fibrosis, iliac artery manipulative procedure, surgery for uterine cancer or transitional cell cancer of the bladder; intervention for ureterocoele,periureteral hematoma following renal transplantation andureter lithotomy. [1].

The pathophysiology of Uretero-iliac aneurysm fistula (UIAF)is not well defined. The most prevalent theory assumed with development of Uretero-iliac aneurysm fistula (UIAF) is that they are related to injury to the ureters, iliac vessels, or both either due to inflammation or ischemia [11].The most common location is place where ureter crosses over theiliac artery, with the ipsilateral common iliac artery being the most commonly involved followed by the external and internal iliac arteries [11]. Most UIAF are due to persistence of fibrosis following surgery or radiation which immobilizes the ureter to the artery at the point of intersection. Atherosclerotic aneurysms can produce perivascular inflammation andfibrosis, the presence of an indwelling stent act as a pressure point in a such condition. The pulsatile waves of the iliac artery is then transmitted to the already damaged ureter. This pulsatile waves produce pressure necrosis with eventual fistula formation. Factors that promote tension and damage to ureters (hydroureteronephrosis, infection, interruption of blood supply and innervations second to surgery, hypotension, and radiation) generate further compromise [12]. The presence of these conditions can weaken wall strength of both structures, and precipitate or worsen fistula hemorrhage during ureteral stent exchange

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Previous genitourinary Surgery, pelvic radiation, and urine leakage are generally responsible for the intense Necrofibrotic inflammatory response that fixes the ureter to an artery or vascular graft [13]. Pressure necrosis, surgical intervention, irradiation, chronic infection, and fibrosis may also result in ureteral ischemia. Some inherentabnormality of the iliac artery musculature such as native aneurysmal disease, further predispose patients to arterioureteral fistulas [1].

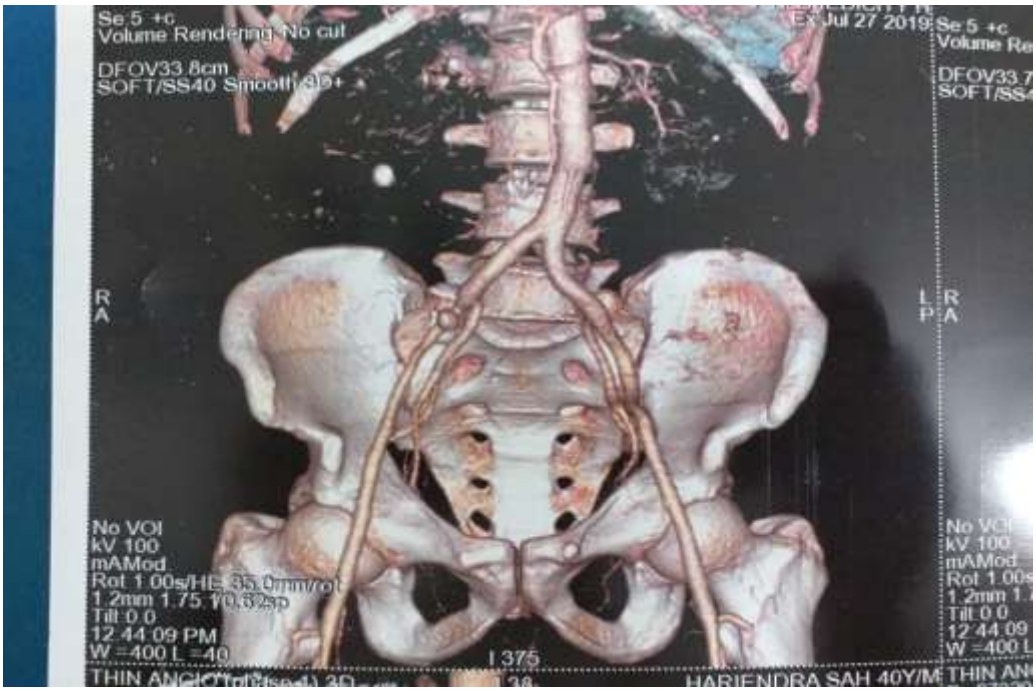

Figure :1

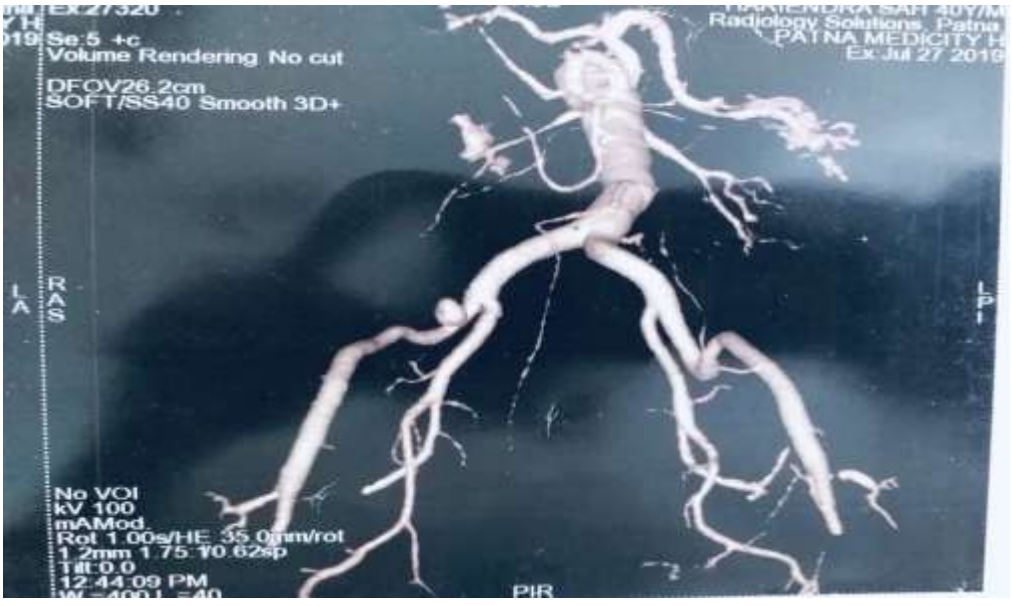

Figure:2

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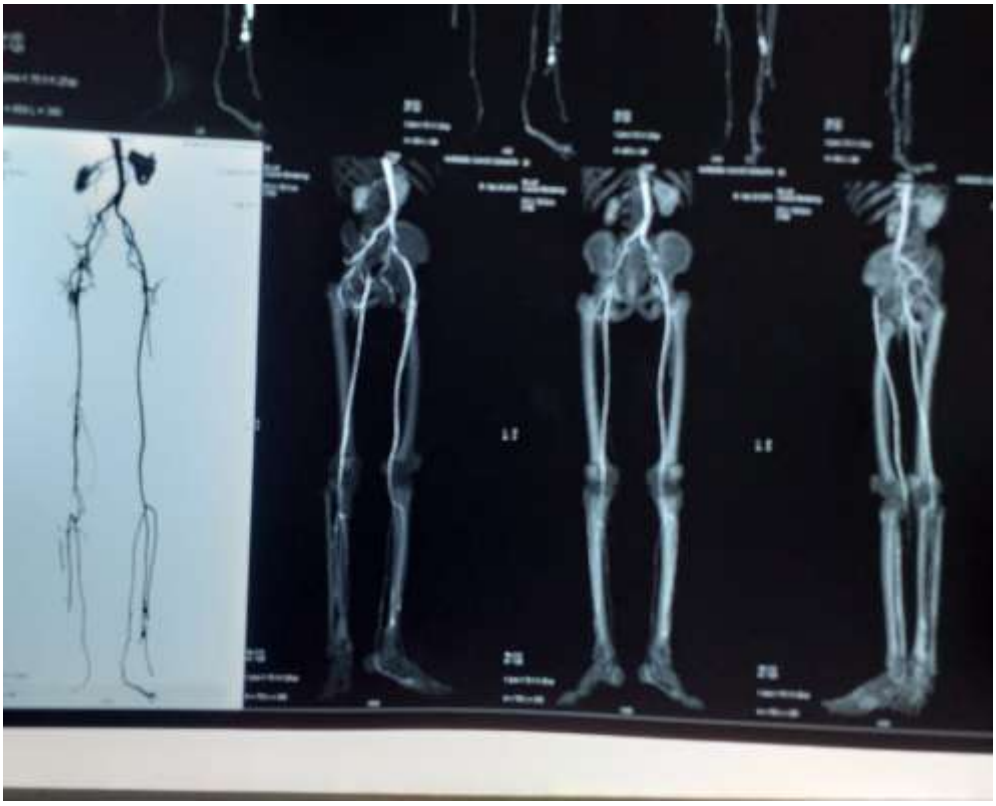

Figure:3

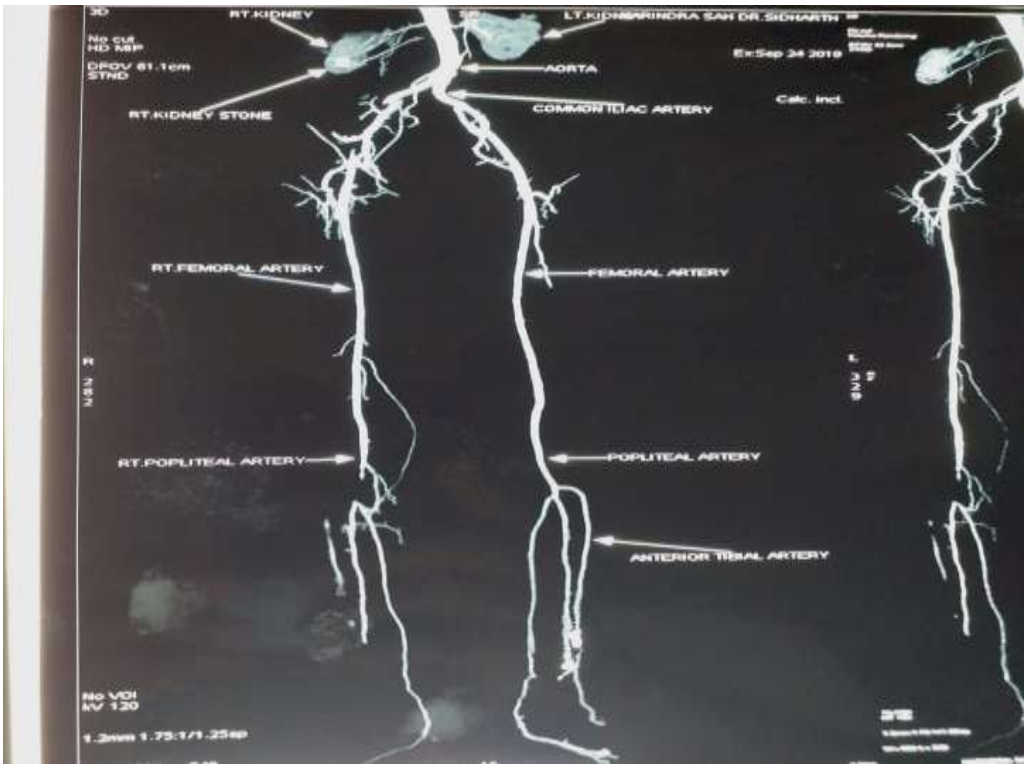

Figure4

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Clinical Presentation:
Uretero-iliac aneurysm fistula (UIAF is generally considered a life threatening condition and non intervention of this condition leads to death and sometimes critical ischemia of limbs. Hematuria especially gross, intermittent or continuous, pulsatile, painless or painful due to clot colic may be presentation. [14] Severe bleeding generally leads to shock and severe shortness of breath. Other symptoms include dull aching flank pain, dysuria due to urinary tract infection are usual presentation.

Sometimes, for patients with ureteral stents, bleeding may be provoked or exaggerated when the stents are exchanged [15]. If the hemorrhage produced during a stent exchange is massive and pulsatile, the diagnosis of arterioureteral fistulas should be considered.

Diagnosis:
Demonstrating a fistula can be very challenging, and a high index of suspension is needed due to the lack of sensitive diagnostic tools. Clotting and valvular action of ureteral stents can result in intermittent hemorrhage making visualization difficult. Surgical exploration is traditionally, well defined approach for diagnosis of arterioureteral fistulas.However, procedure itself is very technically challenging because operating through previously dissected fibrotic tissue planes and the risk of injury to adjoining structures such as, ureters, and blood vessels [9,13]. Thus, various techniques have been used to establish the diagnosis of UIAF before surgery. The widespread use of gray scale ultrasonography of pelvis can detect various ureteric and kidney abnormality. Doppler study can easily detect aneurysm of lilac vessels. CT scans of the abdomen and pelvis are usually negative or nonspecific for arterioureteral fistulas because they show bleeding only rarely and the fistulous communications are almost never seen. CT findings may include pseudo aneurysms (Fig. 1), signs of graft infection, and hydronephrosis with hydroureter. In addition, CT urography may help in excluding the more common causes of hematuria [9, 13]. cystoscopy is more accurate and can localize bleeding to one of the ureteral orifices. If a ureteral catheter or stent is in place, its extraction may provoke bleeding. If the orifical bleeding is pulsatile, an arterioureteral fistula is likely to be present. Advantage of the cystoscopy is that massive bleeding from the ureter can be temporarily blocked using a balloon catheter [16]. Excretory urography and ureterography (antegrade, retrograde, or both) reveal only nonspecific findings such as intraluminal blood clot and irregularity of the mid to distal ureter. [17] Selective iliac arteriography is considered the most sensitive technique, but its sensitivity rate is less than 50% [9]. Arteriograms and ureterograms fail to reveal the fistula during quiescent times, probably because the fistula is occluded by a thrombus. However, when angiographic findings are present they include arterial pseudo aneurysms at the point where the ureter crosses the iliac artery (Fig. 1) or gross extravasation of contrast material into the ureter. Obtaining multiple oblique projections during arteriography helps to identify small pseudoaneurysms that may otherwise be overlooked [18]. Contrast material extravasation and fistulous communication are seen only

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rarely. Provocative techniques such as stent or arterial catheter manipulation, and direct fistula catheterization have been used to induce or increase bleeding. Due to the risk of complications including hemodynamic compromise, provocative maneuvers should be undertaken with the support of a multidisciplinary team to manage complications and allow for emergent treatment [19]. The use of intravascular ultrasound (IVUS) to demonstrate a pulsatile flow pattern in the ureter, may aid in diagnosis during acute hemorrhage despite negative angiography, however, no reports in the literature have determined the sensitivity of this tool.

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Therapeutic Options:

Numerous vascular and urologic interventions are reported with varying degrees of success. Both arterial and ureteral components must be considered in the treatment plan, an accurate preoperative diagnosis is essential to decrease morbidity and mortality rates. In patients who were explored surgically without a preoperative diagnosis, the mortality rate has been reported to be as high as 64% [20]. However, when the correct diagnosis is made before surgery, the mortality rate decreased to 8% [23]. At present, no consensus has been reached about the best treatment option butAdvances in therapeutic techniques have reduced the mortality rate for UAF in recent years. Various treatment options for UIAF exist addressing both the arterial and ureteral components of the fistula. The immediate goal of treatment is to secure a hemodynamic control. Patients with a picture of evolving shock may require emergent ureteral or arterial embolization, endovascularstenting and/or open exploration with delayed reconstruction until stabilization is achieved. Open surgery was considered the treatment of choice and has demonstrated excellent results in the past but need for emergent vascular control and extensive fibrosis in surgical plane made this approach less appealing in recent years [22]. When open surgery is chosen, then repair of the both the artery and ureter are done on single setting, with omental wrapping demonstrating a benefit in preventing reoccurrence [1].

Open surgery is indicated when endovascular approaches fail to identify the location of the fistula, little renal function remains in the affected kidney or emergent vascular control is needed to stop blood loss. Due to the risk of severe hemorrhage, vascular management is the deciding factor when selecting treatment, and is dependent on the presence of infection or abscess, aneurysm or occlusive arterial disease and the availability of collateral blood flow to the ipsilateral lower extremity [23]. An alternative treatment method for the Management of the arterial component of arterioureteral fistulas varies and is influenced by associated local infections, the presence of associated aneurysmal or occlusive disease, the available collateral circulation to the ipsilateral leg, and the presence of an arterial graft. Vascular surgical procedures include local reconstruction (i.e., arteriorrhaphy, patch closure, interposition graft, bypass), ligation with or without extraanatomic bypass (if arterioureteral fistulas arise from either common or external iliac artery), and ligation of the internal iliac artery [1]. In 1908, Moschowitz [24] reported the first successful treatment of bilateral arterioureteral fistulas in which the external iliac arteries were ligated bilaterally. The patient did well after the procedure,

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experiencing only transient lower extremity ischemia with no additional hematuria. The treatment options for arterioureteral fistulas remained unchanged until 1965, when Arap et al. [25] reported the use of a prosthetic graft for primary reconstruction of a diseased iliac artery. Nine years later, Shultz et al. [26] reported a similar case in which the patient underwent nephrectomy for an arterioureteral fistula without experiencing recurrent bleeding. In the 1980s and early 1990s, several reports of successful treatment of these lesions using surgical ligation, intra- operative balloon occlusion, or radiologic embolization of the iliac artery followed immediately by extraanatomic bypass were published [27]. Some authors have described limb ischemia requiring delayed arterial bypass or limb amputation after common iliac artery ligation, and others have reported death during open vascular repair [3, 28]. Similarly, the ureteral component of the fistula can be managed with various procedure like nephroureterectomy, nephrostomy tube placement with or without simultaneous stenting, ureteroureterostomy and percutaneous nephrostomy with ureteral ligation[29]

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The recent addition of endovascularstentgrafts as a therapeutic alternative holds great promise in providing patients with a less invasive but effective method of treating arterioureteral fistulas and provides many of the essential features of an ideal therapy. These features include complete closure of the fistula, maintenance of antegrade blood flow through the iliac artery, no need for direct arterial or ureteral surgery, and avoidance of subsequent procedures for revascularization of the lower extremity [30]. Currently endovascularstenting has become the treatment of choice due to its minimally invasive approach, rapid control of bleeding, and shorter hospital stay. Additionally, it can be rapidly initiated following provocative angiography [31]. Previously balloon- expandable autologous vein covered grafts were used for the treatment, however they have largely been replaced by self-expandable synthetic stents with good results.In 2002, two reports described successful management of arterioureteral fistulas using newly released commercially available polyethylene terephthalate– and polytetrafluoroethylene-covered self-expanding stents [16].

The long-term outcome of endovascularstent treatment forarterioureteral fistulas is not known. Of note is its potential for stent occlusions and graft infections. More important, the 12-month primary patency for stent-grafts in occlusive aortoiliac disease is 70% [32]. Surgical revision in this already compromised setting would be difficult, so antiplatelet agents (to maintain stent-graft patency) and prophylactic antibiotics may be of use, but this is currently unknown [3]. If an infection later develops or if the stent-graft fails and causes persistent hemorrhage, occlusion, or refistulization, extraanatomic vascular reconstruction may be required [33]. One patient experienced stent-graft occlusion at 8 months after deployment necessitating a femoral–femoral bypass. However, nearly 10 similar cases have been reported in the literature, and no mortality using covered-stents in the treatment of arterioureteral fistulas has been reported.

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As mentioned before, both the ureteral and vascular components of arterioureteral fistulas must be addressed to have a successful out- come, and the treatment should be based on the patient's clinical situation. The arterial component must be treated expeditiously be- cause failure to do so can lead to exsanguination.

Conclusion

Ureteroarterial fistulae(UIAF) are a rare cause of both intermittent and continuous hematuria. Lack of definitive diagnostic tool, and the high rate of mortality associated with delays in diagnosis, clinicians must maintain a high index of suspicion when evaluating patients with gross hematuria and other risk factors that are concerning for UAF. Whenever predisposing factors are present and diagnosis is doubtful then emergent laparotomy may be diagnostic as well therapeutic. Open surgical repair can potentially address both the ureteral and vascular components of arterioureteral fistulas, but simple ureteral and arterial repairs are usually not possible because most patients are considered poor candidates for surgery. Minimally invasive techniques such as stent-grafts are currently being used and may represent the best therapeutic options. Further studies of endografts with evaluation for long-term follow-up are necessary before definitive conclusions can be drawn, but so far those techniques look promising.

Conflict of interest: No

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