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SUCCESSFUL SURGICAL TREATMENT FOR RUPTURE OF AN EXTERNAL ILIAC ARTERY PSEUDO ANEURYSM INTO A URETER: A CASE REPORT.

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ABSTRACT:

Background:
Gross hematuria can indicate various urological conditions, including nephrolithiasis, malignancies, infections, and trauma. While most cases are manageable and not life-threatening, certain rare conditions like pseudo-uretero-iliac artery fistula (UIAF) are emergent and potentially fatal, requiring prompt multidisciplinary intervention for diagnosis and treatment.

Case:
This paper presents a unique case of a ruptured pseudo-iliac aneurysm in the ureter. The patient's clinical presentation, diagnostic process, and treatment approach, encompassing angiography, cystoscopy, CT imaging, and possibly surgical exploration, are discussed to highlight the complexity and urgency of managing such cases.

Conclusion:
Pseudo-UIAF represents a critical emergency with varied clinical manifestations from asymptomatic to severe shock. The successful management of this case underscores the importance of a comprehensive diagnostic strategy and a multidisciplinary approach to treatment. Further discussion on the causes, risk factors, and pathophysiology of pseudo-UIAF, along with a review of diagnosis and treatment modalities, enriches our understanding and management of such rare but lethal emergencies.

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

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

Uretero-iliac aneurysm fistula (UIAF) is due to communications between an iliac artery and a 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

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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 the first treatment of the arterial parts of the fistula and preventing any ongoing blood loss, many authors prefer to use the term arterioureteral fistulas. Others prefer the term "ureteroarterial fistulas" because most fistulas between the artery and ureter result from some pathology in the 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.

CASE REPORT.

40 40-year-old male came to our emergency room with gross intermittent haematuria and passage of clots for 15 days—generalized weakness for 5 days and shortness of breath for 1 day.

The patient had a history of right open ureterolithotomy following failed ureteroscopy and intracorporeal pneumatic lithotripsy for right impacted mid-ureteric calculus just one month before current admission. Following the procedure patient remained 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 112 per minute, respiratory rate was 26 per minute and SpO2 was 90%. Investigation revealed Haemoglobin was 3.6 gm/dl, WBC count was 12200 /cmm, urea 46 mg/dl, and serum creatinine 0.9mg/dl. The patient was resuscitated with multiple units of blood transfusion, and oxygen support. Ultrasound abdomen was done which showed right mild hydroureteronephrosis with urinary bladder clots. Cystoscopy was done with local anesthesia which showed a clot in the bladder and a profuse oozing of blood from the right ureteric orifices. Possibility of bleeding from major artery suspected. We then planned computed urography angiography and urgent exploration. CT urography and angiography revealed an 11x9mm aneurysm arising from a right external iliac artery, adjacent to and closely related to the thickened wall of the ureter at the level of its crossing over the iliac vessel,(Figures 1 & 2).

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

Under general anesthesia, a Right hockey stick incision was given, and the retroperitoneal right External iliac artery approached. Due to extensive and dense fibrosis, the approach was very difficult and tedious but finally artery was almost mobilised and pseudo aneurysm ligated. Intraoperatively transient loss of arterial pulsation may be due to arterial spasm. The bleeding in the ureter stopped immediately. A Double J stent was placed in the right ureter and the bladder clot was evacuated. In the postoperative period, we noticed decreased pulsation in the right dorsal paedis artery. However, movement and pain sensations were present in the right limb. Gradually a good volume pulsation felt in the right dorsalis paedis artery.

The patient was discharged on the 10th postoperative day. Follow-up was done after one month with CT angiography of both lower limbs which showed occlusion at the 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. The patient is ambulatory and doing his all normal physical activity. In our case, the urgent availability of surgical services and the presence of ipsilateral collateral ultimately prevented mortality and morbidity. Our case emphasizes the importance of the emergent availability of any intervention to treat ureteroarterial fistula as this disease is very lethal without treatment. Here we will review the ureteroarterial fistula in detail.

DISCUSSION:

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 processes of the artery and account for less than 15%. [6]. Sometimes congenital arteriovenous malformation may be causative. (7)

The majority of Uretero-iliac aneurysm fistula (UIAF) are secondary and generally had a prior history of pelvic surgery, ureteric intervention in the form of chronic double J stenting, and pelvic radiotherapy. The median period for the onset of symptoms varies with the nature of the previous intervention. Some studies showed that the median period from the 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

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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 of arterioureteral fistulas related to the gravid uterus are reported [10] in the 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 stent placements in two patients. Fistulas due to gravid uterus are nowadays rare due to the 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 the 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 and ureter lithotomy. [1].

The pathophysiology of Uretero-iliac aneurysm fistula (UIAF) is not well defined. The most prevalent theory assumed with the 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 a place where the ureter crosses over the iliac artery, with the ipsilateral common iliac artery being the most commonly involved followed by the external and internal iliac arteries [11]. Most UIAFs are due to the 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 and fibrosis, and the presence of an indwelling stent acts as a pressure point in a such condition. The pulsatile waves of the iliac artery are then transmitted to the already-damaged ureter. These 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 the wall strength of both structures and precipitate or worsen fistula hemorrhage during ureteral stent exchange

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 inherent abnormalities of the iliac artery musculature such as native aneurysmal disease, further predispose patients to arterioureteral fistulas [1].

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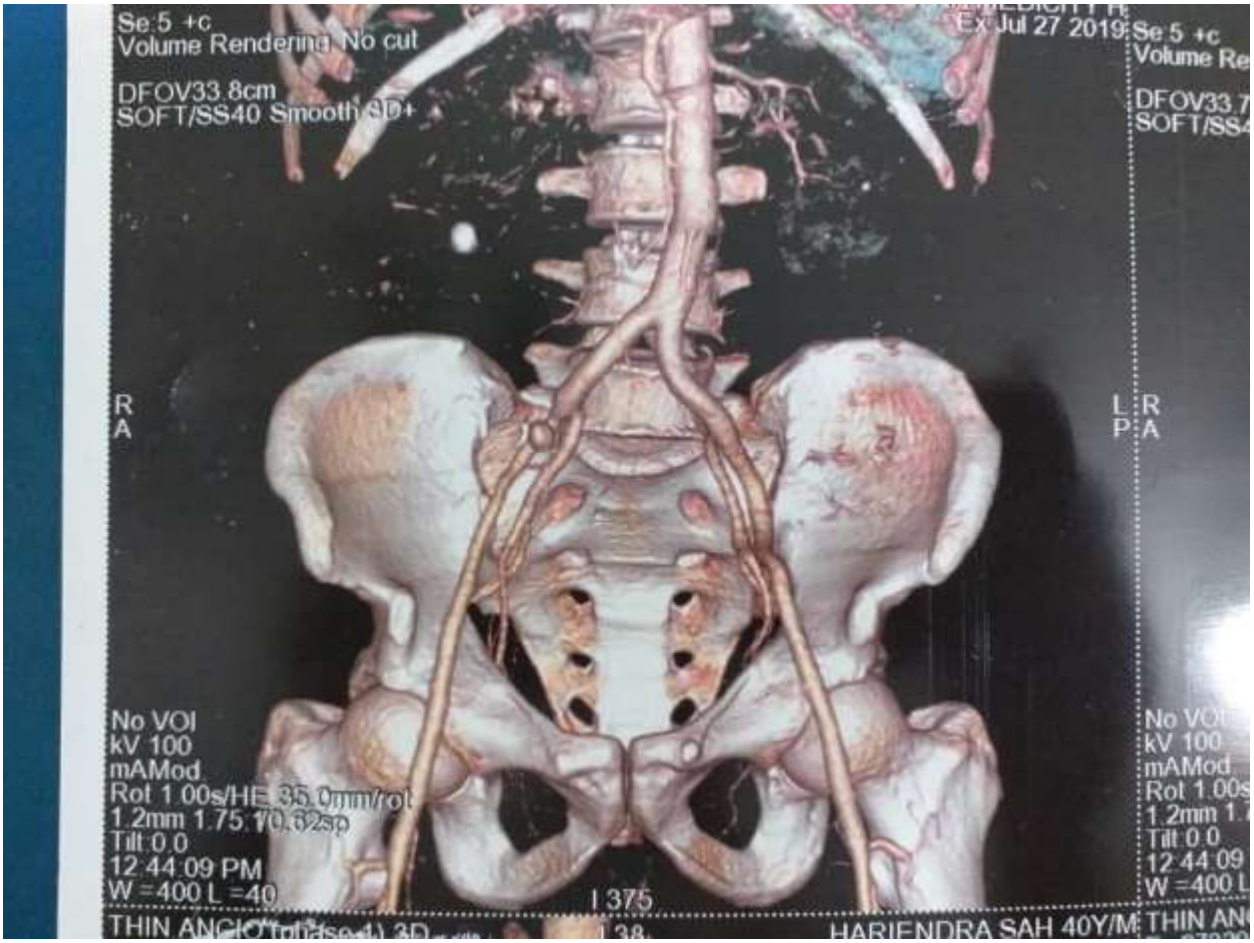

Figure 1: Diagnosis of Pseudo aneurysm of right external iliac artery communicating with right ureter (Possibly fistulous communication)

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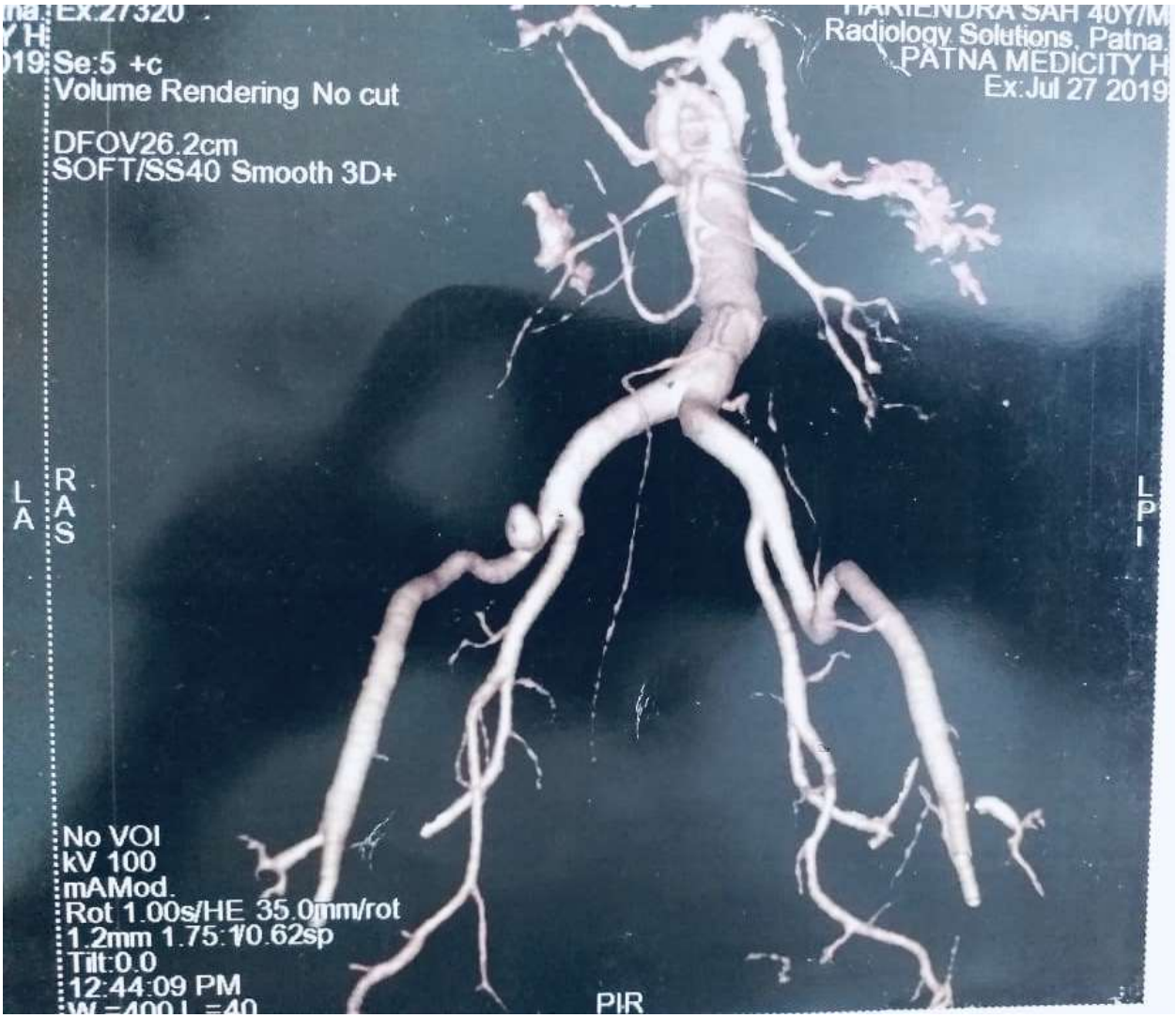

Figure 2: CT angiography of aneurysm arising from right external iliac artery.

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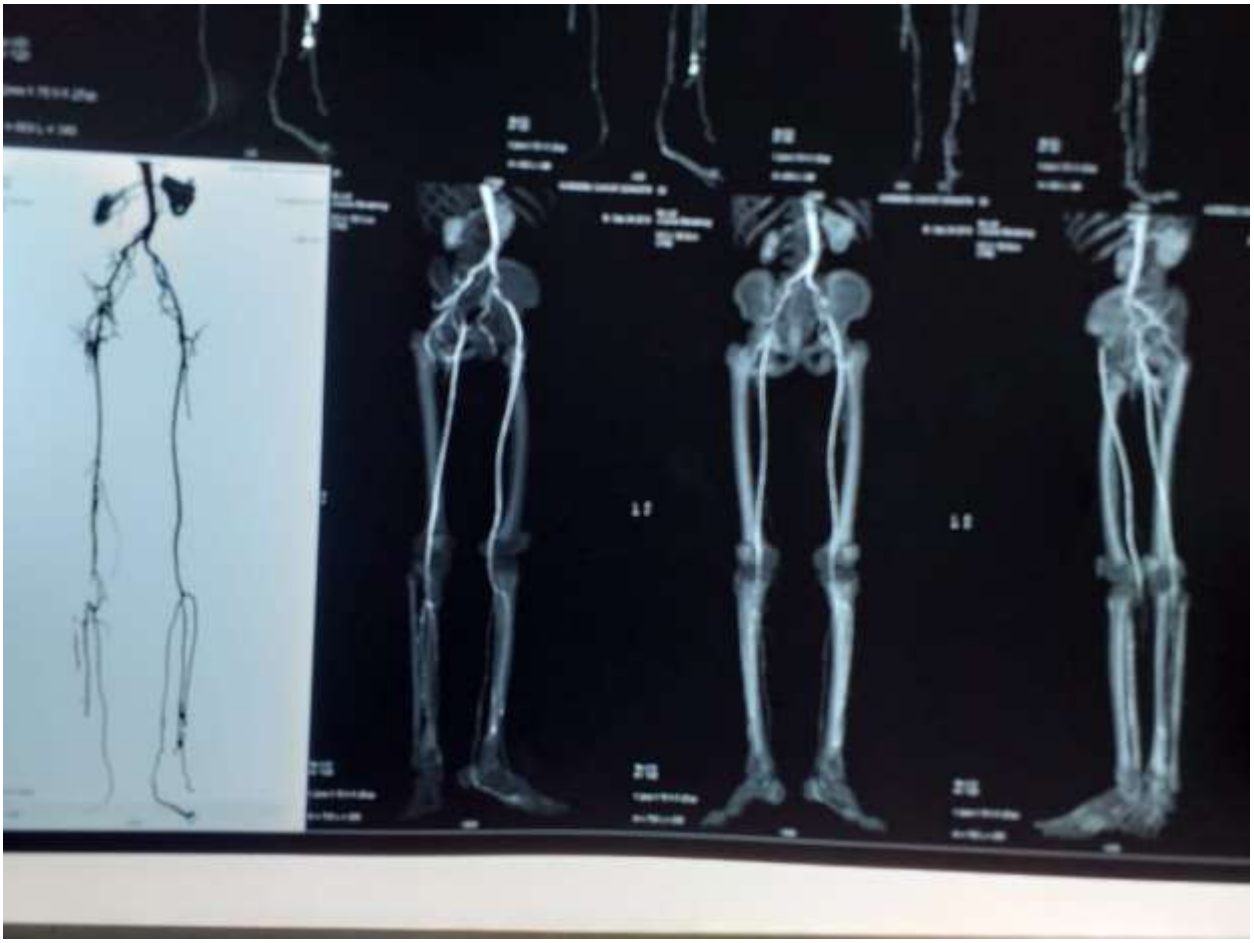

Figure 3: CT angiography of both lower limbs.

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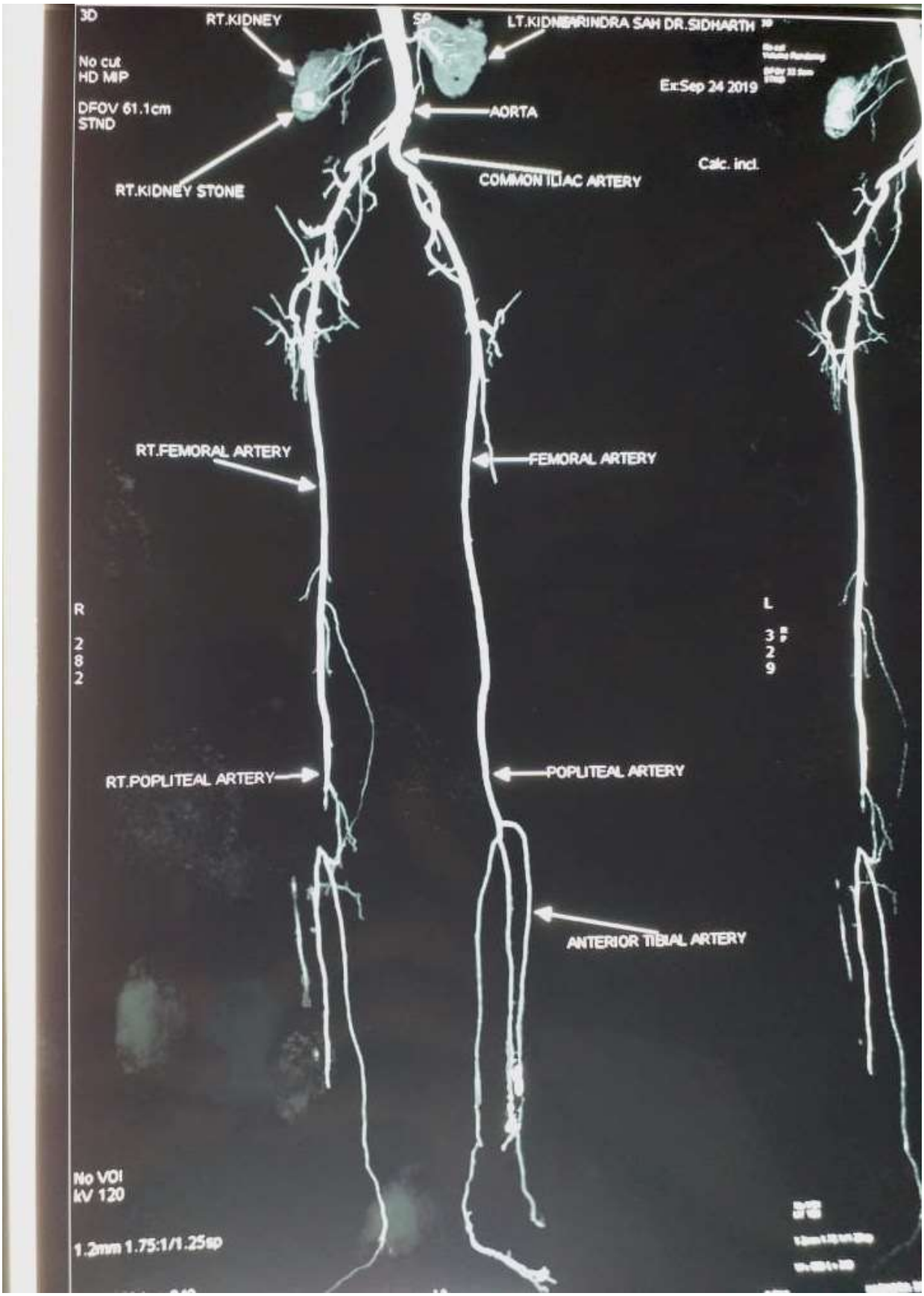

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Figure 4: CT angiography of both lower limbs which showed occlusion at the site of Pseudoneurysm and well-formed collaterals which preserved lower limb circulation.

Clinical presentation.

Uretero-iliac aneurysm fistula (UIAF is generally considered a life-threatening condition and nonintervention 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 present. [14] Severe bleeding generally leads to shock and severe shortness of breath. Other symptoms include dull aching flank pain and 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 a traditionally, well-defined approach for diagnosis of arterioureteral fistulas. However, the procedure itself is very technically challenging because of 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 the pelvis can detect various ureteric and kidney abnormalities. 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 rarely 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. The 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 clots 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 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.

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 decreases to 8% [23]. At present, no consensus has been reached about the best treatment option but Advances 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 hemodynamic control. Patients with a picture of evolving shock may require emergent ureteral or arterial embolization, endovascular stenting, 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 the need for emergent vascular control and extensive fibrosis

in a surgical plane made this approach less appealing in recent years [22]. When open surgery is chosen, then repair of both the artery and ureter is done on a 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, the 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., arteriography, patch closure, interposition graft, bypass), ligation with or without extra-anatomic bypass (if arterioureteral fistulas arise from either common or external iliac artery), and ligation of the internal iliac artery [1]. In 1908, Moskowitz [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, 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 extra-anatomic 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 procedures like nephroureterectomy, nephrostomy tube placement with or without simultaneous stenting, ureteroureterostomy and percutaneous nephrostomy with ureteral ligation [29]

.

The recent addition of endovascular stent grafts 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, endovascular stenting 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 the successful management of arterioureteral fistulas using newly released commercially available polyethylene terephthalate– and polytetrafluoroethylene-covered self-expanding stents [16].

The long-term outcome of endovascular stent treatment for arterioureteral 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, extra-anatomic 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.

As mentioned before, both the ureteral and vascular components of arterioureteral fistulas must be addressed to have a successful outcome, 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 as 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 endograft with evaluation for long-term follow-up are necessary before definitive conclusions can be drawn, but so far, those techniques look promising.

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CONFLICT OF INTEREST.

The author had no conflict of interest.

ETHICAL CONSIDERATIONS.

The study was approved by the Ethics Committee and written informed consent was received from the participant.

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