

Unusual Presentation of Gonadal Vein Aneurysm - Thrombosis and Hydronephrosis: A Rare Case Report

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ABSTRACT

Gonadal vein aneurysm is a very rare anomaly and the aetiology is often unknown. Many factors have been said as contributory such as multiple pregnancies, multiple pelvic surgeries, pelvic tumours or pelvic congestion, but none of them have been strongly or closely associated with the lesion. Here, we present a case of left gonadal vein aneurysm in a female with its management. A 72-year-old, diabetic and hypertensive female presented with complaints of left loin pain. She is multiparous with four normal vaginal deliveries. On evaluation, she was diagnosed to have 4 cm left gonadal vein aneurysmal mass with internal thrombus causing hydronephrosis due to mass compression and perilesional adhesions. She underwent left gonadal vein aneurysm excision.

Many treatment options are available such as endovascular- embolization/sclerotherapy. In the presence of thrombus and dense adhesions, as noted in our case, it is best to do open surgery for safety of the patient and obtain a good outcome.

Keywords: Loin pain, Thrombus, Venous aneurysm

CASE REPORT

A 72-year-old female presented with left loin pain of one month duration. The pain was dull aching, insidious in onset, intermittent and was non radiating. There were no aggravating or relieving factors. There were no other associated complaints. She was diabetic for the past thirty years, hypertensive with coronary artery disease and renal stone disease. She had undergone Extra Corporeal Shock Wave Lithotripsy (ESWL) for left renal calculi, eight years back. She is multiparous with four full term normal deliveries in the past. Physical examination was unremarkable.

On evaluation, blood biochemistry was within normal limits. Erythrocyte sedimentation rate was 39 mm/hour. Abdominal ultrasound revealed a 4 cm size aneurysmal mass, arising from the gonadal vein, near the point of drainage into the left renal vein. There was a thrombus within the aneurysm, with resultant hydronephrosis of the left kidney.

The contrast enhanced CT scan confirmed the findings with a 4 cm left gonadal vein aneurysm with an internal thrombus (red arrow in [Table/Fig-1]), located close to its termination into the left renal vein, with peri-lesional adhesions causing compression on upper ureter leading to hydronephrosis.

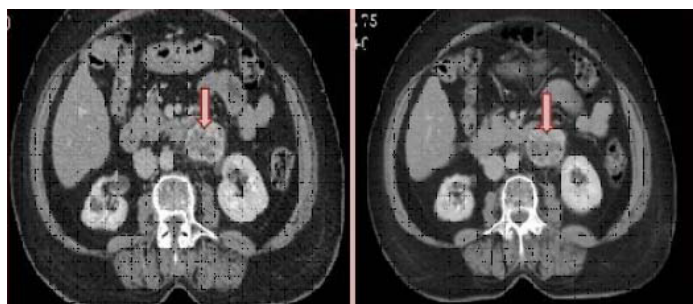
A preoperative diagnosis of gonadal vein aneurysm with resultant thrombosis and hydronephrosis was made. The cause of pain was found to be hydronephrosis secondary to obstruction by the

thrombosed aneurysmal mass. Various treatment modalities like open/laparoscopic excision of the aneurysmal mass, sclerotherapy of the aneurysmal vessel or conservative management of the thrombosed vessel were thought of.

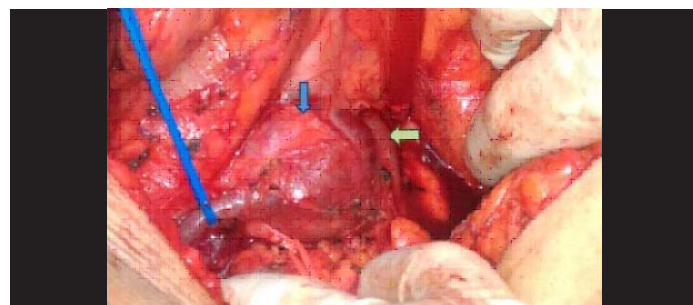
As the thrombosis was identified at the region very close to the renal vein, and as there was an inherent fear of impending embolism, an open surgical excision of the aneurysmal mass was carried out. Patient underwent exploration and excision of left gonadal vein aneurysm. Intraoperatively, a 4 x 3 cm left gonadal vein aneurysm with internal thrombus with dense peri-lesional adhesions was noted. The left upper ureter was found to be compressed by the aneurysm and encased by the adhesions leading to hydronephrosis. The ureter was released from all those adhesions. The lesion was very closely adherent to left renal vein and required sharp dissection [Table/Fig-2]. Postoperative period was uneventful. Patient is doing well on follow up after 10 months with no pain or hydronephrosis.

DISCUSSION

Venous aneurysm is defined as “a solitary area of venous dilatation containing all three layers of the vein wall that communicates with a main venous structure by a single channel and must have no association with an arteriovenous communication or pseudo aneurysm” [1].



[Table/Fig-1]: Contrast enhanced CT scan of abdomen, showing the gonadal vein aneurysm with thrombosis (red arrow).



[Table/Fig-2]: Intraoperative photograph depicting the thrombosed aneurysmal mass (blue arrow), gonadal vein (blue tape), ureter compressed by the mass (red tape) and the renal vein (green arrow) in close proximity.

S. No	Author	Findings/Presentation	Treatment given
1	Kauffmann K et al., [5]	Right ovarian vein aneurysm with pulmonary embolism causing acute respiratory distress	Streptokinase for thrombolysis ; Surgical excision for aneurysm at a later date
2	Kishi Y et al., [6]	Left ovarian aneurysm masquerading as mesenteric cyst abdominal discomfort	Excision of mesenteric cyst.
3	Golder WA et al., [7]	Left ovarian and IVC aneurysm; hemangioliopoma of back muscle; multiple bony abnormalities	No treatment; Conservatively managed.
4	Yoo J et al., [8]	Right ovarian vein aneurysm causing right flank discomfort for few weeks. Diagnosed to have right ovarian aneurysm and para-pelvic cyst.	No treatment
5	Ghersin E et al., [9]	Post liver transplant with ovarian vein aneurysm	Aneurysm excision
6	Present case	Left gonadal vein aneurysm presenting with thrombus and hydronephrosis	Aneurysm excision and thrombectomy

[Table/Fig-3]: Comparison with the previous similar reported studies [5-9].

The venous aneurysms are most often seen to develop from the superficial veins of the head and neck. Such aneurysms are not so common in the abdominal viscera. Visceral aneurysms are commonly seen in portal veins and superior mesenteric veins [2].

Portal venous aneurysms are considered to be the most common visceral venous aneurysms [3]. Aneurysms involving the veins of the deep system may rarely be associated with deep venous thrombosis and pulmonary embolism [4]. Gonadal vein aneurysm is a very uncommon condition and is of unknown aetiology and presents as a vascular lesion with impending risk of pulmonary thromboembolism [5].

The rarity of gonadal vein aneurysm may be due to its indolent nature and asymptomatic behaviour. In our case report, we present a case of left gonadal vein aneurysm that presented to us with persistent bothersome left loin pain and hydronephrosis. Surgical excision of the aneurysmal mass relieved the obstruction and pain. Such aneurysms can be difficult to diagnose and if left untreated, may occasionally be a source of significant morbidity and mortality [4].

After an extensive PubMed search since 1998 till June 2016 with keywords like venous aneurysm, thrombosis, gonadal vein and hydronephrosis, we observed that there were only five such similar cases reported before and ours is the sixth case to be ever reported in the English literature. The very low incidence may partly be due to its vague symptomatology leading to under recognition. [Table/Fig-3] shows the previous five reported cases, their presentation and their management [5-9].

The indications for surgery in gonadal vein aneurysm: large size of lesion, peri-lesional adhesions, compression causing hydronephrosis and pain. Lesions have hardly any external manifestation. The occurrence is quite rare and it represents only 0.9% to 1% of all venous malformations [2]. Of the reported cases, three were operated and aneurysms were excised and patients improved symptomatically. Only two of these had a preoperative diagnosis of gonadal vein aneurysm [5-7] whereas third case reported by Kishi Y et al., was operated as a case of mesenteric cyst causing abdominal discomfort [6]. However, the postoperative histopathology of the resected specimen was consistent with gonadal vein aneurysm. One was a case of gonadal vein aneurysm post liver transplant [6,7]. All the operated cases have been managed by open surgical techniques and have given good outcome. Laparoscopic repair of gonadal vein aneurysm is also a feasible option. Endovascular techniques of sclerotherapy and embolization are viable options [10]. Sclerotherapy is achieved with sodium tetra decylsulphate or sodium monorrrhate both with gel foam as a carrying agent that leads to chemical phlebitis of the lesion.

CONCLUSION

Thrombosis within the aneurysm and hydronephrosis as a result of obstruction are extremely rare. The close proximity of the thrombosed vein to the left renal vein necessitates an early intervention. A high index of clinical suspicion is mandatory to make an appropriate diagnosis and prompt treatment of this potentially dormant but a rarely devastating condition.

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