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Case Report

True Aneurysm of the Uterine Artery in a Young Nulliparous Female: An Extremely Rare Vascular Entity

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We present the first case of a large true uterine artery aneurysm, with a 5-cm diameter, in a 35-year-old nulliparous woman who presented with lower abdominal pain and dyspareunia. She underwent successful ligation and excision of the aneurysm using the Pfannenstiel approach. The diagnostic modalities and treatment option for such a case is discussed herein.

Keywords: uterine, true aneurysm, uterine artery

Introduction

Aneurysms of uterine artery are rare, and within this subset, they are mostly pseudoaneurysms. They are formed as a result of trauma to the uterine vessels during caesarean section.[1)] Pseudoaneurysm patients present with painless, life-threatening postpartum haemorrhages.[1,2)] True aneurysm of uterine artery is an extremely rare entity. Considering the diameter of uterine vessels, such an aneurysm has high incidence of rupture once they reach a diameter of >1 cm.[3)] Furthermore, such aneurysms have been reported in women over 50. In this report, we describe the case of true aneurysm of uterine artery in a young female reaching a diameter of 5 cm, and against all odds, it remained unruptured during the presentation.

Case Report

A 35-year-old nulliparous female, married for 5 years was referred to us by her gynaecologist, who had been treating her for infertility. She complained of lower abdominal dull pain for the past 3 years which worsened with intercourse. She also complained of having painful intercourse for the past 2 years with pain persisting for a few minutes to an hour after intercourse. She reported no history of vaginal bleeding, abdominal or pelvic trauma, and previous abdominal surgery. There was no family history of vasculitis and collagen disease. On examination, she was of an average build (body mass index, 30) with no marfanoid features. She had a normal hormonal profile; however, her serum creatinine was 3.3 mg/dL (normal range, 0.5–1.1 mg/dL). While being investigated for infertility, she underwent a pelvic ultrasound which revealed a pelvic mass just to the left of her uterus. Active blood flow in the mass was noted on colour flow Doppler. Pelvic magnetic resonance imaging (MRI) revealed a heterogeneous mass to the left of the uterus, but it was completely distinct from it (**Fig. 1**). A computerised tomography (CT) angiogram confirmed the diagnosis of a true aneurysm arising from the left uterine artery, measuring approximately 5 cm in all three dimensions (**Fig. 2**).

She underwent explorative laparotomy, and the aneurysm sac was excised after taking proximal and distal vascular controls. The patient tolerated the procedure well and had an uneventful postoperative recovery. The histology of the resected specimen showed pathological process in all layers of the aneurysm wall. There was reduction in the vascular smooth muscle cells, mononuclear lymphocytes and macrophages infiltration along with fibrosis in both the tunica media and adventitia.

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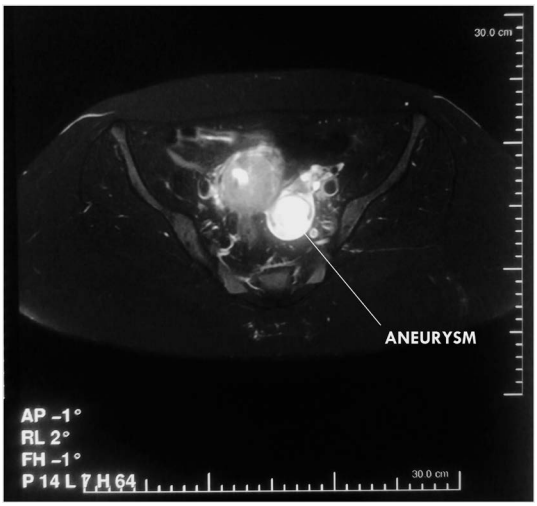

Fig. 1 Magnetic resonance imaging (MRI) of the pelvis showing a large mass on the left displacing the uterus to the right and anteriorly.

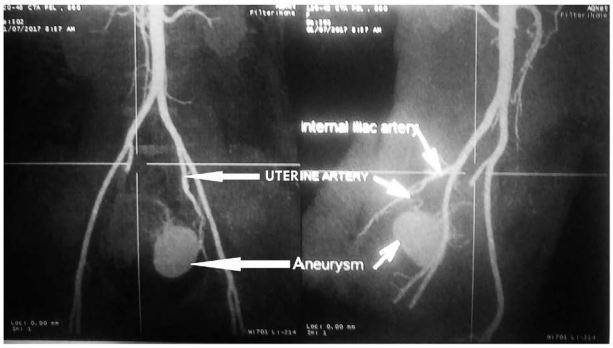

Fig. 2 Computerised tomography (CT) angiogram showing an aneurysm of the left uterine artery.

Discussion

Aneurysms of the uterine artery are a rare entity. Commonly, these are pseudoaneurysms which are formed as a result of damage to uterine vessels following caesarean section. The incidence of uterine artery pseudoaneurysm in child-bearing females is estimated at 0.2%–0.3%.[4] Pseudoaneurysm rupture and subsequent postpartum haemorrhage have been reported in the literature.[2,5] A case of rare true aneurysm was found in a 77-year-old patient; originally misdiagnosed as pelvic sarcoma, it was later detected during laparotomy.[6] Another case of a ruptured true aneurysm measuring 1 cm has been reported in a 69-year-old female presenting with retroperitoneal haematoma.[3] There is no report in the medical literature of a true aneurysm of this size (5 cm) in a nulliparous young female. We present this as the first such case wherein a patient had a true aneurysm presenting with lower abdominal pain and dyspareunia. Furthermore, it was of an unusually large size of 5 cm and was unruptured.

Uterine artery aneurysm can be diagnosed by pelvic ultrasound which shows a hypoechoic mass; however, it can be misdiagnosed for a uterine fibroid because of the complexity of pelvic anatomy and proximity of such aneurysm to the uterus. A colour flow Doppler may reveal blood flow in sac, but it can also be misdiagnosed for a uterine fibroid with red degeneration. A CT angiogram is the gold standard for this type of investigation. It is helpful not only in diagnosis but also in assessing the morphology of the aneurysmal sac. Moreover, it has the added advantage that if anatomy is suitable and adequate facilities and expertise are available, treatment with embolisation may be performed.

Endovascular treatment with angiographic arterial embolisation is the preferred treatment because it is non-invasive, safe and effective.[7] However, its disadvantage includes failure in completely cutting off blood supply from the uterine side, possibly resulting in recurrence. Surgical exploration is reserved for the cases wherein endovascular expertise is unavailable or has failed. Patients with poor renal reserves, such as our case, are often not good candidates for endovascular treatment, considering the amount of contrast needed to complete the procedure. Further, open exploration with simple ligation of uterine or internal iliac artery is not a good option because blood flow from the uterine side will not allow complete exclusion from circulation. In such cases, the aneurysm may be excised if it can be easily shelled out or if all inflow channels ligated from within the open sac can be cut off from the blood supply. However, open exploration, requiring laparotomy, is more invasive and increases the risk of damage to the nearby structures such as the ureter and rectum. In addition, pelvic dissection may result in postoperative adhesions which may affect fertility in women of child-bearing age.

There is a higher risk of uterine ischaemia after uterine artery occlusion (UAO). Lichtinger et al. measured uterine pH following UAO and concluded that it postoperatively returned to a normal value within 6 h in 80% of patients.[8] Other studies also concluded fast revascularisation from collaterals, negating the theory of clinically significant uterine ischaemia.[9,10] The data to predict the pregnancy rates following UAO is insufficient. Chen et al. reported pregnancy rates of 41.6% in sexually active females who underwent UAO for fibroids; however, there is a relatively high rate of early miscarriage (41.2%).[11] Similarly, Chang et al. concluded that fertility rate following UAO was not compromised, but they did note a higher rate of spontaneous abortion. Patient counselling regarding these risks is recommended.[12]

Conclusion

To the best of our knowledge, a large, unruptured, true uterine artery aneurysm has not been previously reported in the literature. In this case, the patient was treated with ligation and excision. Although endovascular options are much safer in such a case, but conventional surgical excision is recommended for these cases where endovascular techniques are unavailable or there are relative contraindications to use such techniques.

Disclosure Statement

All authors have no conflict of interest.

Author Contributions

Study conception: RU
Data collection: MR
Analysis: MJ
Investigation: RU, MJ
Writing: all authors
Critical review and revision: RU, MJ
Final approval of the article: all authors

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