Twisting around an axis: A case report of uterine torsion

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Abstract

Introduction: A woman presenting with sudden, severe pelvic pain should prompt an evaluation for torsion, most commonly, ovarian torsion. Uterine torsion is rare, especially in a non-gravid uterus.

Case: A post-menopausal woman with known history of uterine leiomyomata presented with sudden, acute pain that was non-amenable to medication. Imaging demonstrated a significant increase in uterine size but was otherwise normal. Primary diagnoses included a degenerative fibroid, leiomyosarcoma, or pelvic thrombosis. Surgical intervention revealed a levo-rotated uterus. Final pathology demonstrated a leiomyosarcoma.

Conclusion: Pelvic organ torsion most often presents as sudden pain that is unrelieved by medication. Diagnosis is sometimes made with imaging and Doppler studies. Surgical intervention is often required. In a patient with a rapidly enlarging fibroid uterus with acute pain, one should consider a uterine torsion.

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with findings significant for an enlarged and lobulated uterus measuring 19.8 × 15.0 × 16.6 cm with multiple uterine fibroids which obscured the underlying endometrium. A small amount of ascites was present in the upper quadrants.

Due to the patient’s multiple co-morbidities, the decision was made to proceed with medical stabilization prior to surgical intervention with continued close monitoring. Her hemoglobin level was optimized prior to surgery with two units of packed red blood cells. The patient was then scheduled for an exploratory laparotomy with hysterectomy and bilateral salpingo-oophorectomy. Due to increasing pain, the surgery was moved forward by a day.

5. Therapeutic Intervention

Upon entry into the abdomen, one liter of blood was evacuated. The uterus was described as bulky, edematous, with multiple masses and visualization was initially limited due to sheer size and bleeding. However, upon packing of the bowel and evacuation of the hemoperitoneum, the uterus was noted to have levo-rotated and torsed upon itself at the level of the junction between the cervix and uterine corpus (Fig. 1). Initial frozen pathology was reported as a ruptured, necrotic fibroid uterus appeared as a round, soft mass weighing 1540 g and measuring 16.0 × 15.0 × 10.0 cm. A thin rim of pink serosa was surrounding the mass. The serially sectioned specimen revealed soft, tan, fleshy, whorled tissue. Histologic sections of the uterus contained a smooth muscle tumor composed of abundant ordinary spindle smooth muscle elements. An epithelioid tumor component was present both as dispersed present amidst the smooth muscle fascicles as well as in expansile nodular areas of near pure epithelioid growth. Both the ordinary spindleled and epithelioid components marked with desmin, caldesmon and smooth muscle actin. The tumor contained foci of necrosis, not of a hyaline type, but of an individual cell nature typical of sarcoma. The spindled component was not cytologically atypical, but the epithelioid component, which occupied more than 50% of the neoplasm, was notably atypical. The mitotic count was 4/10 per high-power field, with some aberrant forms noted. Together, the atypicality, epithelioid differentiation, and necrosis were judged sufficient to establish a diagnosis of leiomyosarcoma with epithelioid features. Given the size and final histological findings, a stage IB sarcoma was diagnosed. The patient was referred to a gynecologic oncologist for further management. The patient has since not required any further radiological or chemotherapeutic intervention.

To the authors’ knowledge, this is the first case report of a leiomyosarcoma presenting as a uterine torsion. Written consent was obtained for the publication of this case report.

6. Follow-up and Outcome

A final pathologic diagnosis was delayed to obtain expert consultation. Grossly, the fibroid uterus appeared as a round, soft mass weighing 1540 g and measuring 16.0 × 15.0 × 10.0 cm. A thin rim of pink serosa was surrounding the mass. The serially sectioned specimen revealed soft, tan, fleshy, whorled tissue. Histologic sections of the uterus contained a smooth muscle tumor composed of abundant ordinary spindle smooth muscle elements. An epithelioid tumor component was present both as dispersed present amidst the smooth muscle fascicles as well as in expansile nodular areas of near pure epithelioid growth. Both the ordinary spindleled and epithelioid components marked with desmin, caldesmon and smooth muscle actin. The tumor contained foci of necrosis, not of a hyaline type, but of an individual cell nature typical of sarcoma. The spindled component was not cytologically atypical, but the epithelioid component, which occupied more than 50% of the neoplasm, was notably atypical. The mitotic count was 4/10 per high-power field, with some aberrant forms noted. Together, the atypicality, epithelioid differentiation, and necrosis were judged sufficient to establish a diagnosis of leiomyosarcoma with epithelioid features. Given the size and final histological findings, a stage IB sarcoma was diagnosed.

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Post-operatively, the patient’s course was complicated by an ileus but with eventual return of bowel function. The patient was discharged on post-operative day five.

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7. Discussion

Uterine torsion is defined as the rotation of the uterus more than 45 degrees on its long axis, and most commonly occurs at the junction between the cervix and the uterine corpus (Fig. 1). Dextro-rotation occurs in two-thirds of cases, while levo-rotation is seen in the remaining third [4–6]. The exact mechanism for the occurrence remains unclear, although possible explanation includes weakness of the parametrial tissues following menopause [3,7]. Risk factors for this condition include a large uterine size (such as pregnancy) or a large mass (for example leiomyoma). Piot et al. described a case series in which the most common causes in the gravid uterus were: fibroids in 32%, uterine anomalies (including bicornuate uterus) in 15%, pelvic adhesions in 7%, ovarian cysts or other adnexal mass in 7%, and abnormal fetal presentation or fetal abnormality in 5% and 3%, respectively [8]. The presentation in a non-gravid uterus is extremely rare and diagnosis is most commonly made using imaging, such as ultrasonography or MRI [3,7]. Imaging findings on ultrasound may include: transposition of placental site compared with previous scan, transposition of known uterine leiomyoma compared with previous scan, and Doppler recognition of ovarian vessels anterior to the uterus [9]. In most cases, however, symptoms are non-specific and hence diagnosis is typically not made before surgical intervention [3,4,7,10,11].

Surgical intervention was required for diagnosis of uterine torsion in our case since imaging had failed to demonstrate any overt evidence. Given her post-menopausal state, and rapidly enlarging uterine size, the differential diagnosis included a necrotizing fibroid and pelvic malignancy, but not uterine torsion. This is the first reported case of uterine torsion, to the authors’ knowledge, where the causative process was a leiomyosarcoma. The patient's lack of obvious imaging findings, vague symptoms, and multiple co-morbidities may have clouded the pre-operative diagnosis. The significant increase in uterine size prompted...
a higher suspicion of severe disease. Only during surgery was the diagnosis made but was invaluable in the therapeutic intervention and management of the patient.

**Contributors**

Sophia Halassy performed the literature review and drafted the manuscript.

David Clarke provided supervision and revised the manuscript.

**Conflict of Interest**

The authors declare that they have no conflict of interest regarding the publication of this case report.

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**Patient Consent**

Obtained

**Provenance and Peer Review**

This case report was peer reviewed.

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**References**

[1] K.L. Moores, M.G. Wood, R.P. Foon, A rare obstetric emergency: acute uterine torsion in a 32-week pregnancy, BMJ Case Rep. 2014 (2014).

[2] A. Hashimoto, et al., A case of uterine torsion concurrent with a ruptured ovarian endometrial cyst, Abdom. Radiol. (NY) 41 (9) (2016) 1707–1712.

[3] N. Havaldar, K. Ashok, Torsion of non-gravid uterus with ovarian cyst - an extremely rare case, Pan. Afr. Med. J. 18 (2014) 95.

[4] A. Dua, K. Fishwick, B. Deverashetty, Uterine torsion in pregnancy: a review, Int. J. Obstet. Gynecol. 6 (1) (2005).

[5] J. Corr, Axial torsion of the gravid uterus in two successive pregnancies, Am. J. Obstet. Gynecol. 46 (5) (1943) 4.

[6] P. Collinet, F. Narducci, L. Stien, Torsion of a nongravid uterus: an unexpected complication of an ovarian cyst, Eur. J. Obstet. Gynecol. Reprod. Biol. 98 (2) (2001) 256–257.

[7] D. Sharma, M. Usha, Torsion of a non-gravid uterus: a rare cause of acute abdomen, Int. J. Reprod. Contracept. Obstet. Gynecol. 2 (2) (2013) 3.

[8] D. Piot, M. Gluck, H. Oxorn, Torsion of the gravid uterus, Can. Med. Assoc. J. 109 (10) (1973) 1010–1011.

[9] P. Guié, et al., Uterine torsion with maternal death: our experience and literature review, Clin. Exp. Obstet. Gynecol. 32 (4) (2005) 245–246.

[10] C.H. Hawes, Acute axial torsion of the uterus, Ann. Surg. 102 (1) (1935) 37–40.

[11] S. Qureshi, et al., Torsion of preterm gravid uterus: a case report, Int. J. Case Rep. Images 4 (7) (2013) 4.