



Torsion of Giant (3 Kg) Non - Gravid Myomatous Uterus with Congenital Cervical Agenesis with Absence of Right Side Ovary and Fallopian Tube: Extremely Rare Case

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Case Report

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Abstract

Uterine torsion is defined as rotation of the uterus around its longitudinal axis by more than 45 degrees. Uterine torsion is supposed to be a rare entity with a combination of diagnostic as well as management challenges. The uterine torsion is mostly associated with uterine anomalies and torsion is common in a gravid uterus rather than a non-gravid uterus. Till date, less than fifty such cases have been reported. Here we report a case report of gynaecology uterine torsion over its axis, which was an incidental finding and was managed with great surgical expertise and during the times of COVID pandemic.

Keywords: Non Gravid Uterine Torsion; Mullerian Anomaly; COVID-19; Pandemic; Rare; Adnexal Torsion; Ovarian Torsion; Computed Tomography; Ultrasonography; Genitourinary

Introduction

Uterine torsion encountered in a non-gravid uterus is extremely rare [1,2]. A literature search reveals less than 50 reported cases of uterine torsion in a non-gravid uterus. Uterine torsion is a rare entity. Among the pelvic masses, ovarian torsions are commonly encountered in a clinical practice. Any torsion is associated with ischemic and gangrenous changes presenting as acute abdomen and is characterized by decreased blood flow on doppler studies. However, uterine torsion do not present in the same way and are often diagnosed intra operatively. It is considered as a "once in a lifetime" opportunity for the gynecologist. Hence we present this case for its rarity.

Case Report

A 43 years old, unmarried, nulligravida, diagnosed case of pott's spine operated for thoracic corpectomy with lower limb paralysis with bed sores, referred from neurosurgery department in view of uterine fibroid.

Patient gave history of tubercular meningitis? Tuberculosis cervical spine 22 years ago for which she was admitted at hospital and took treatment for 3 years for MDR-TB. Post-meningitis, patient had bilateral lower limb paralysis for a year, after which she regained bilateral lower limb power. Three years ago, patient again developed weakness in lower limbs and had fever spikes. She was diagnosed of tubercular spondylodiscitis of D8 and D9 and underwent thoracic corpectomy.

Post operatively, patient had persistent abdominal pain for which Computed Tomography was advised 6/9/19. Computed Tomography was suggestive of intramural uterine fibroid with cystic changes measuring 15.5 x 20.3 x 22.5 cm arising from posterior uterine wall.

(12/3/2020) Ultrasound was suggestive of Bilateral polycystic kidney with liver cysts

Tumor markers were sent - CA 125-18.4; AFP 13.4; BHCG

-2.61; LDH -231; CEA - 0.94

Patient lost follow up due to the COVID -19 pandemic and presented to Grant Government Medical College, Mumbai with complaints of pain in abdomen and weakness of lower limbs.

She had attained menopause 2 years back
On clinical examination, general condition was fair, afebrile.

Pulse: 90/min; blood pressure: 120/80 mmhg, SpO₂: 99% at RA, on systemic examination, Cardiovascular and respiratory examination was normal. On per abdomen examination solid mass of 30 weeks of gestation , occupying umbilicus, left hypogastric, left lumbar region, regular margins, mobile, firm in consistency. Superficial skin was normal.

On local examination, -3x2 x3 cm bed sore seen in right greater trochanter region -3 x5 x 0/5 cm bed sore seen bilateral over natal cleft region with heavy granulation seen. On per rectal examination: rectal mucosa free; lower border of pelvic mass felt. Margins were regular

12/2/2021: Covid 19 swabs RT PCR reported negative.
12/2/2021: gene xpert: MTB not detected.

14/2/2021: plastic surgery reference done i/v/o bed sores. Advised wound culture sensitivity and frequent change of positions jellonet dressing.

23/2/2021 computed tomography abdomen + pelvis was suggestive of multiple hepatic and bilateral renal cysts, suggestive of autosomal dominant polycystic kidney disease mild hepatomegaly (18 cm), large hypodense solid lesion of 11x13x14cm arising from the left ovary with torsion of the ovary? dysgerminoma.

Fitness for surgery was obtained from neurosurgery, nephrology and plastic surgery.

Patient underwent: exploratory laparotomy with? mixed mesodermal uterine ovarian mass excision with left salpingectomy with omentectomy under General anesthesia after pre anesthetic fitness.

in situ-large mass of 30cm*25cm*20cm extending from the lower abdomen and reaching upto the xyphisternum superiorly with fullness of flanks with solid consistency, the mass has hemorrhagic surface, dilated vessels seen on surface of mass. Mass was adherent to left anterior abdominal wall, blood supply from vessels extending from left anterior abdominal wall, all the adhesions separated systematically with achieving haemostasis.

Anteriorly, mass was adherent to large intestine, posteriorly was reaching upto iliac vessels using harmonic scalpel and bipolar cautery, all the bleeders were systematically cauterized. Anterior abdominal wall, all the adhesions separated systematically with achieving haemostasis. Fine dissection and done with hemostatic control and mass was delivered out .In situ we have found that The mass has undergone with the axial rotation of 360 degree with 4-5 rotating rings seen at the pedicle. Pedicle of the mass contains major vessels, part of fallopian tube, infundibulopelvic ligament. Uterus was not seen and must have developed in tumor remnants of ovarian tissue. The cervix was absent and the Right sided Fallopian tube and Right sided ovary was absent congenitally.

Ureters were pulled up, ureteric dissection done and ureter pulled down. bilateral ureters traced in their course completely-> intact and not injured. Urinary bladder was pulled up with dilated vessels over the surface.

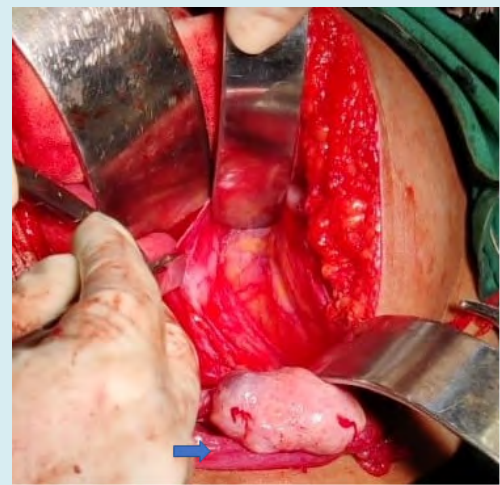


Figure 1: Left side fallopian tube and ovary saved

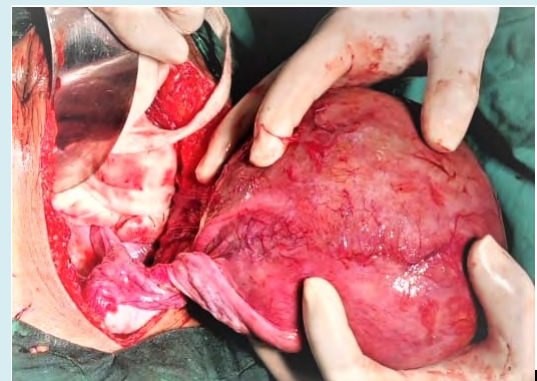


Figure 2: Myomatous Non - Gravid Uterine torsion and Congenital Absence of cervix.

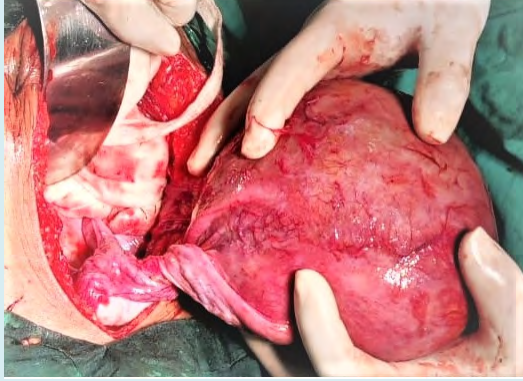


Figure 3: Right side fallopian tube and ovary, congenitally absent.

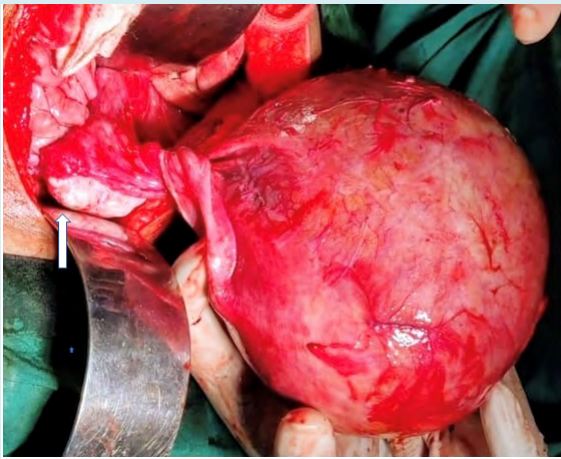


Figure 4: Left side Fallopian tube and Ovary in the pedicle of torsion

Frozen section was sent and was suggestive of leiomyoma with no evidence of malignancy. Patient withstood the procedure well. Post op monitoring was done and was started on higher injectable antibiotics

10/3/2021: Histopathological report suggestive of proliferative phase endometrium with leiomyoma; fallopian tube stump unremarkable and left fallopian tube had normal histology. Patient was discharged later on.

Discussion

Uterine torsion is defined as the rotation of the uterus more than 45 degrees on its long axis, and mostly reported at the junction between the cervix and the uterine corpus [3].

Rotation of the gravid uterus around 45 degrees is physiological and is due to the recto-sigmoid colon on the posteriorlateral aspect of the uterus [4]. Dextro – rotation

is more commonly encountered (around two thirds of the patient) rather than the levo-rotation reported in the rest [5,6,7]. The pathogenesis is attributable to the wakening of the uterine supports and parametrium following the post-menopausal status [8,9].

Torsion is usually secondary to the uterine pathology like uterine leiomyoma, gravid state or the uterine anomalies.

Piot, et al. has reported the most common factor associated with uterine torsion to be leiomyoma [10].

The symptoms with which the patients present include asymptomatic presentation, very rarely pain in abdomen and fetal malpresentation in a gravid uterus. Till date around 200 cases have been reported. The failure to be diagnosed on imaging modality is a major challenge.

Intra operatively, the diagnosis can be confirmed by palpating the two round ligaments. If the symmetry is distorted then the diagnosis of uterine torsion is confirmed. In our case it was complete 360 degrees torsion.

The differential diagnosis of ovarian torsion, broad ligament fibroid should be kept in mind. Broad ligament fibroids of huge size might cause displacement and distortion of anatomy to make the case more difficult [11].

The ovarian torsion with benign masses like teratoma may obscure the peritoneal space and distort the abdominal anatomy mimicking uterine torsion. Clinical diagnosis may be difficult and hence the possibility of the same should be borne in mind [12].

The congenital absence of cervix a very rare disorder with its incidence being only 1 in 80,000 [13]. It is considered as 1B classification of Müllerian anomaly [14,15].

The surgical importance of round ligament is well establishes in such cases as it helps in delineating the pelvis anatomical structures precisely [16]. Uterine torsion encountered in a non-gravid uterus is extremely rare [1,2]. A literature search reveals less than 50 reported cases of uterine torsion in a non-gravid uterus.

The patient was managed with a high surgical safety index and a great surgical expertise. The patient and relatives were satisfied with the management and gave the consent for publication of this case report.

Conclusion

This case is reported for its rarity and its diagnostic and management challenges that were associated. Diagnosing a

malformed uterine torsion is difficult on imaging modalities as well as intra operatively. Its a very rare entity but sometime can become Emergency with torsion and its consequences. The treatment is always surgical and it depend on the degree of torsion and damage occurred. Need of Fertility sparing surgery to be considered in young woman who wanted to preserve uterus for the fertility. if older women then go for the Hysterectomy. Best clinical Acumen to Diagnose and a great surgical expertise required to treat the condition.

Conflict of Interest

The author declares that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Obtained

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