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Case Report

Case report on uterine inversion secondary to endometrial stromal sarcoma: a diagnostic challenge

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ABSTRACT

Nonpuerperal uterine inversion (NPUI) is rarer entity. They are usually preceded by various benign and malignant pathologies, in our case a 60-year-old multiparous woman presented with postmenopausal bleeding and the presence of a vaginal mass over the past four months. Her clinical examination revealed a large necrotic, malodorous growth filling the entire vaginal canal and cervix was not visualized due to the obstructive growth. Bimanual examination posed challenges due to the expansiveness of the mass. Imaging revealed endometrial hyperplasia and biopsy confirmed carcinoma of the endometrium. Examination under anesthesia revealed uterine inversion, correction of anatomy was done by Haultain's procedure and complete staging was performed. Her histopathology revealed endometrial stromal sarcoma. We report this case due to challenging diagnosis of uterine inversion. Also, this case underscores the rarity of NPUI and its potential association with malignancies like endometrial stromal sarcoma.

Keywords: Non-puerperal uterine inversion, Chronic inversion, Malignancy, Endometrial stromal sarcoma

INTRODUCTION

Uterine inversion, classified as puerperal or nonpuerperal, represents a rare gynecological complication with distinct etiologies.[1] Puerperal inversion occurs post-delivery, with an incidence of 1 in 30,000 deliveries.[2] NPUI, a considerably rarer entity, lacks a well-established incidence estimate, demanding a heightened clinical suspicion for accurate diagnosis.[1] This case report details a unique instance of NPUI caused by endometrial stromal sarcoma, emphasizing the intricacies of diagnosis and the surgical correction employed.

CASE REPORT

A 60-year-old multiparous woman presented with postmenopausal bleeding and the presence of a vaginal mass over the past four months. Additionally, she experienced urinary voiding symptoms, culminating in urinary retention. Menopause had occurred 16 years prior, with a history of normal perimenopausal transition, her cycles in reproductive age were regular with average flow. Patient reported no hormone replacement therapy and had never undergone cervical cancer screening. She had no history of any chronic medical illness/any surgery.

With a BMI of 22 kg/m², her systemic examination revealed no noteworthy findings. Her blood pressure was within normal limits. Per speculum examination revealed a 8×10 cm necrotic, malodorous growth filling the entire vaginal canal up to the introitus, which bled upon contact. The cervix was not visualized due to the obstructive growth. Bimanual examination posed challenges due to the expansiveness of the mass, preventing the separate palpation of the cervix and uterus. Rectovaginal examination did not add.

Her blood work was unremarkable apart from mild anemia. Her blood sugar profile was normal. Pelvic ultrasound indicated endometrial hyperplasia, and a CT scan revealed diffuse endometrial thickening measuring 12 mm infiltrating the fundoposterior wall, with the endometrial cavity filled with blood. There were no signs

of ascites or lymphadenopathy. Biopsy confirmed carcinoma of the endometrium, leading to the decision to proceed with an examination under anesthesia.

Examination under anesthesia corroborated the clinical findings. It was decided to proceed with staging laparotomy. Laparotomy revealed an intriguing intraoperative discovery. The uterine fundus was not visualized; instead, a cervical ring was observed with bilateral round ligaments, ovarian ligaments, and fallopian tubes entering inside the ring as shown in Figure 1. A diagnosis of uterine inversion was made intraoperatively. The vaginal mass was comprised of the malignant growth from the uterine fundus leading to uterine inversion. Decision was made to proceed with Haultain's procedure to correct the inversion followed by hysterectomy and additional procedures. An incision was made at the 6 O'clock position on the inversion ring. The large mass made it impossible to correct the inversion, hence the mass was excised vaginally under careful guidance of finger protecting the fundus from above. After the resection of the mass the uterine fundus was pushed cranially from the vaginal end, successfully correcting the uterine inversion. Subsequently, a total abdominal hysterectomy, bilateral salpingoopherectomy, bilateral retroperitoneal lymph node dissection, and infracolic omentectomy were performed. The final histopathological examination revealed high-grade endometrial stromal sarcoma involving over 50% of the myometrium.

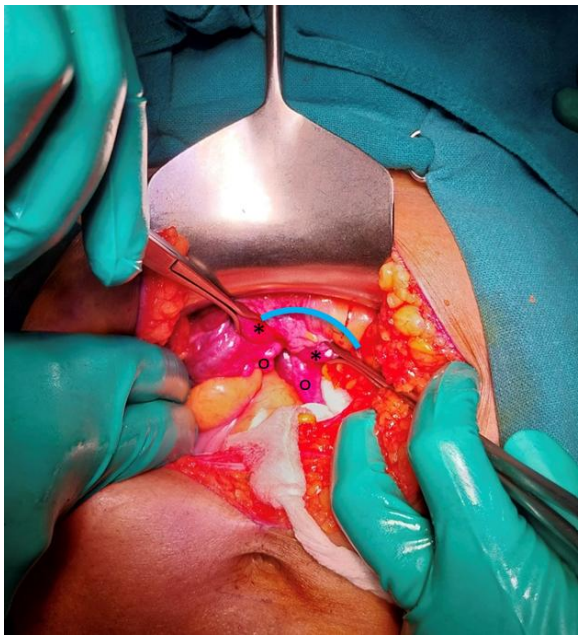

Figure 1: Laparotomy view: round ligaments; fallopian tubes; solid blue line representing anterior border of the cervical ring.

DISCUSSION

Chronic uterine inversion, a rare condition involving the descent of the uterine fundus through the cervix, typically manifests with vaginal bleeding, abdominal pain, foul-smelling discharge, or urinary disturbances. Chronic inversions may result from various antecedents, such as fibroids, endometrial polyps, or malignancies like sarcoma and cervical carcinoma.[1]

Unlike puerperal inversion, the mechanisms triggering chronic inversion remain less understood. Plausible explanations include the sudden emptying of a distended uterine cavity due to an intrauterine tumor, leading to the expulsion of the tumor through the cervix and subsequent inversion. Patient factors like increased intra-abdominal pressure or gynecological procedures may contribute to chronic inversion.[1]

Clinical diagnosis is facilitated by the visualization of a descending mass through the cervix and the inability to palpate the uterine fundus on bimanual examination. Ultrasonography can reveal a longitudinal groove indenting the fundus, aiding in diagnosis.[1]

Non-surgical methods of correction of uterine anatomy include O'Sullivan's hydrostatic technique, use of uterine balloon tamponade device but the efficacy of these methods are not clear.[2,3] Various surgical techniques described in literature for correction of chronic inversion are: Huntington procedure involves grasping the round ligaments and the uterus below the area of inversion and slowly pulling up repeatedly until the uterus is reinverted. Haultain procedure involves incising the posterior wall of the vaginal-cervical ring and carrying up the posterior wall of the uterus until it is reinverted to its normal anatomy. The Kustner procedure approaches the posterior aspect of uterus via cul-de-sac entry vaginally, splitting the posterior wall and reinverting the uterus.[4] Spinelli operation is also a vaginal procedure in which incision is given on the anterior aspect of the cervix and then the uterus is reinverted.[5] Robotic and laparoscopic surgeries are also being done increasingly for this correction.[6] Abdominal cerclage surgery has also been reported to prevent occurrence of recurrent uterine inversion.[7,8] In our case we corrected the uterine anatomy by using the Haultain technique.

CONCLUSION

In this case, the diagnosis of uterine inversion was missed pre-operatively both at clinical evaluation as well as by imaging. This case underscores the rarity of NPUI and its potential association with malignancies like endometrial stromal sarcoma. Timely diagnosis, coupled with a nuanced understanding of surgical correction techniques, is crucial for managing this uncommon yet clinically significant condition. Further research and awareness are warranted to refine diagnostic approaches and optimize treatment strategies for NPUI.

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