

Case Report

Uterine Adenofibroma: An Unusual Cause of Nonpuerperal Uterine Inversion in Postmenopausal Female

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ABSTRACT

Uterine adenofibroma is an extremely rare benign mixed mullerian tumor, most often presenting as vaginal mass with pain and abnormal uterine bleeding in postmenopausal females. Nonpuerperal uterine inversion is also an uncommon entity. We present a rare case of nonpuerperal uterine inversion due to a uterine adenofibroma. A 56-year-old postmenopausal female presented to us with bleeding and discharge per vaginum. Examination showed a polyp. Due to associated comorbidities, polypectomy was chosen as management modality failing which laparotomy was done, and the uterus was found to be inverted through the cervix, hysterectomy was done. Histopathological evaluation showed uterine adenofibroma. This is the first case of inversion reported due to uterine adenofibroma.

KEYWORDS: Nonpuerperal, uterine adenofibroma, uterine inversion

INTRODUCTION

With no incidence documented in literature, nonpuerperal uterine inversion is such an unusual phenomenon that gynecologists can spend their entire career without coming across one.^[1] Adenofibroma of the uterus is a rare benign mixed-mullerian tumor. The majority of cases arising from endometrium, only 10% cases affects cervix.^[2] We herein describe a case of uterine adenofibroma with uterine inversion; their clinical features, diagnosis, and management.

CASE REPORT

A 56-year-old postmenopausal female presented to us with complaints of on and off vaginal bleeding and discharge per vaginum for the past 4 years. She also had severe crampy pain in the back and lower abdomen since the last 1 week. She was a known case of hypertension, type 2 diabetes mellitus (on insulin), and coronary artery disease with history of percutaneous transluminal coronary angioplasty done 3 years back. The patient was pale with hemoglobin of 6.5 gm%. Pelvic examination revealed the vagina to be full of an irregular polypoidal growth of 7 cm, with bleeding and discharge. Cervix could be felt extremely high up. Ultrasound showed a slightly enlarged uterus, with thick endometrium, with a cystic mass in vagina. Polypectomy

had been attempted thrice as earlier the patients' medical condition was not fit to allow general anesthesia. This time, laparotomy was done and the uterus was found to be inverted through the cervix. The tubes and ovaries were completely drawn into the fundus [Figure 1a]. Huntington's procedure was performed to correct the inversion followed by a total abdominal hysterectomy and bilateral salpingo-oophorectomy. Cut section showed the uterus to be full of multiple polypoidal masses as shown in the picture [Figure 1b]. Histopathological examination revealed a biphasic tumor composed of benign glands and stroma (Hematoxylin and Eosin, ×40) [Figure 2a]. The glands were cystically dilated at places and lined by cuboidal to low columnar epithelium. They were separated by oval to spindle-shaped endometrial stromal cells which were immunopositive for CD10 [Figure 2b]. No necrosis, mitosis, or cytological atypia were noted either in the epithelial or stromal component. The endometrium was normal with no evidence of malignancy. The patient had an uneventful postoperative recovery period and was discharged after 4 days. She has been following with us and is well after 6 months of surgery.

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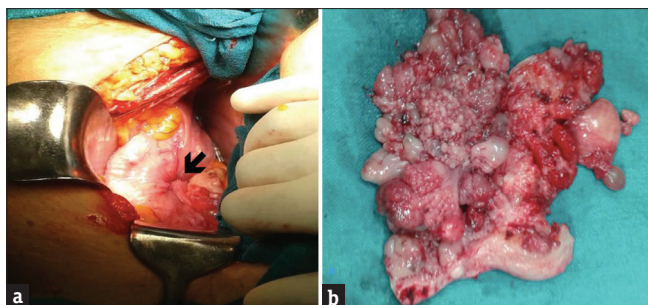


Figure 1: (a) Both tubes and ovaries were completely drawn in through the fundus. The arrow shows the depression at the fundus. (b) Cut section showing the uterus full of a polypoidal mass

DISCUSSION

Uterine inversion has been classified as puerperal (obstetric) or nonpuerperal (gynecological). The incidence of puerperal inversion is around 1 in 3500 delivery, however, nonpuerperal uterine inversion as mentioned is an even rarer event.^[1] Inversion can be either incomplete, when the fundus is inverted but has not passed out of the cervix and complete when the inverted fundus has passed through the cervix.

A review done by Mwinyoglee *et al.* reported that in almost 97% cases cause of nonpuerperal uterine inversion is tumors, 20% of them being malignant one.^[3] As reported in literature, most common cause of gynecological uterine inversion is submucous fibroids, followed by sarcoma and endometrial carcinoma and last idiopathic. Uterine inversion is hypothesized to occur as an attempt of the uterus to expel the tumor through the cervix. Clinical features include vaginal discharge, irregular vaginal bleeding, crampy pain, pelvic discomfort, mass protruding into or through the vagina.

Making a preoperative diagnosis can be very challenging, and many a times, inversion can be misdiagnosed as cervical malignancy due to similar presentation. Inability to palpate the uterine fundus on bimanual examination and inability to visualize the cervix after removal of the vaginal mass has been proposed for the diagnosis of uterine inversion. Magnetic resonance imaging can prove to be a useful imaging modality for preoperative diagnosis of uterine inversion. A “V-shaped” uterine cavity and a thick, inverted fundus in a sagittal section and section through the upper level show layers of the inverted uterus with “bull’s-eye” configuration in T2-weighted images.^[4] On ultrasound, the same can be diagnosed by visualization of an indentation at the fundus on longitudinal section.^[5] However, in our case, ultrasound did not pick up the inversion.

Surgical treatment of inversion for benign diseases such as uterine leiomyoma includes a simple hysterectomy either vaginal or abdominal. For malignant conditions,

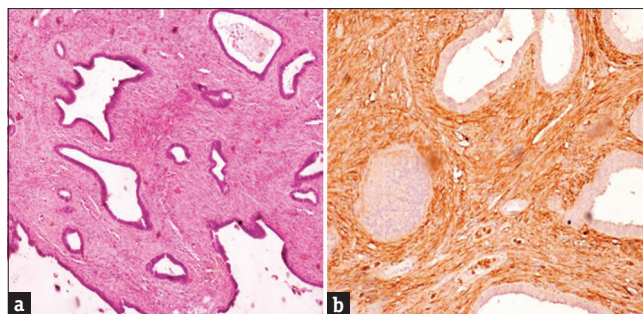


Figure 2: (a) Cystically dilated glands with benign stromal and epithelial components (H and E, ×40). (b) The stromal component was immunopositive for cell differentiation marker 10 (CD10)

abdominal approach should be preferred. The first step in surgery should be to correct the inversion.^[6] Although there are reports of hysterectomy without repositioning the uterus which could be technically challenging.^[7] Vaginal approach includes the Spinelli’s procedure that involves giving a vertical incision on the cervicoisthmic constriction ring anteriorly or Kustner’s procedure where the incision is given posteriorly. The abdominal correction could be attempted by application of Alli’s clamps just beyond the constriction ring of the inversion in an attempt to try and pull out the fundus and the uterine wall gradually (Huntington’s procedure) or by making a vertical incision posteriorly at the constriction ring (Haultain’s procedure). We corrected the inversion in our case by the Huntington’s method before proceeding for hysterectomy.

Adenofibroma of the uterus is a rare benign mixed-mullerian tumor usually occurring in the postmenopausal age group but have also been reported in younger females as well.^[8] About 90% of the adenofibromas arise in the endometrium, rest 10% have been reported to occur in the cervix.^[8] Most often the women present with postmenopausal/irregular bleeding per vaginum or pain in the lower abdomen.^[9] Clinical examination may reveal a normal sized or enlarged uterus or a polypoidal mass in the vagina. Hysterectomy is the treatment of choice for endometrial adenofibroma as it is associated with high rates of recurrence after local removal and due to the chances of carcinoma development in both adenofibroma and the adjacent endometrium.^[10] The detailed histopathological examination is needed to differentiate this entity from a uterine adenocarcinoma. This should be always be considered in the differential diagnosis of females with recurrent polypoidal growths in the vagina and abnormal bleeding.

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Conflicts of interest

There are no conflicts of interest.

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