CASE REPORT

Chronic uterine inversion with malignancy mimicking carcinoma cervix

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SUMMARY

Chronic non-puerperal uterine inversion is a rare event, with only a handful of cases reported in literature. We report a case of a 64-year-old postmenopausal woman who presented with complaints of mass per vaginam and postmenopausal bleeding. On examination, she appeared to have third-degree cervical descent with an irregular growth seen over what appeared to be the cervix, biopsy of which showed keratinising squamous cell carcinoma. Subsequently, an inverted uterus was diagnosed at laparotomy with an irregular growth seen over the inverted portion of the uterine fundus, histopathology of which revealed squamous cell carcinoma of endometrium.

BACKGROUND

There are over 150 cases of chronic non-puerperal uterine inversion, with most reports citing a prolapsed uterine fibroid as the cause. This is the only case in which we found a malignant growth in the inverted fundus of the uterine cavity, leading to the rare diagnosis of uterine inversion with carcinoma endometrium. Clinical diagnosis was difficult due to the distorted anatomy and could be confirmed only at laparotomy during which the inverted uterus could be clearly visualised. Our case is rare in its presentation and pathology and posed a diagnostic and surgical challenge to learn from.

CASE PRESENTATION

A 64-year-old postmenopausal woman presented to the gynaecology outpatient clinic with complaints of mass coming out of vagina since the past 30

Figure 2 T2-weighted MRI showing heterogeneously enhancing mass in the region of the cervix with a dip in the uterine fundus.

years. She first noticed this mass after the birth of her youngest child; however, she did not seek any medical attention for the same as it did not interfere with her daily activities. Since the past 3 months,

Figure 1 Local examination showing what appeared to be complete uterine prolapse with irregular friable growth present over what appeared to be the cervix.

Figure 3 Intraoperative image showing a dimple in the pelvis containing bilateral round and infundibulopelvic ligaments which had inverted into the vagina with presence of a constriction ring over the uterus.

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Figure 4 Gross specimen of the uterus showing a fleshy ulcerative lesion of 7×6×1.5 cm at fundus with healthy-looking cervix.

she also developed bleeding per vaginam and foul-smelling discharge per vaginam which was profuse and blood stained. She was multiparous with history of previous four vaginal deliveries conducted at home by traditional birth attendant. She was a known chronic smoker with history of chronic obstructive lung disease. On examination, she was cachexic with a body mass index of 18.2 kg/m² and her blood pressure and systemic examination were normal. On examination, abdomen was

Figure 5 Immunohistochemistry showing malignant cells stained strongly positive for oestrogen receptor.

soft with no organomegaly and no palpable lymph nodes. On per speculum examination, she appeared to have third-degree cervical descent with no prolapse of bladder or bowel. There was an irregular friable growth of 3×2 cm present over what appeared to be the cervix of which a biopsy was taken ([figure 1](#)). On per vaginal examination, bilateral fornices were shallow and the surrounding vaginal mucosa appeared erythematous. On per rectal examination, rectal mucosa and bilateral parametria appeared free.

INVESTIGATIONS

On pelvic ultrasonography, the uterus was anteverted and low-lying with a 5.5×3.0×3.2 cm collection in the uterine cavity; bilateral adnexa were normal. An MRI was performed which showed a heterogeneously enhancing mass in the region of the cervix which was also filling the endometrial cavity, and there appeared to be a dip in the uterine fundus ([figure 2](#)). A biopsy was taken from the lesion which showed a keratinising squamous cell carcinoma.

TREATMENT

With a provisional diagnosis of carcinoma cervix with uterine prolapse, the patient was planned for surgery. On laparotomy, a 3 cm dimple was noted in the pelvis containing bilateral round and infundibulopelvic ligaments which had inverted into the vagina and constriction ring was visible over the uterus ([figure 3](#)). We proceeded with a posterior incision given over the cervical ring as described in Haultain procedure and reduced the prolapsed uterine fundus through it. This was followed by type I hysterectomy with bilateral salpingo-oophorectomy in the usual manner and the specimen was sent for histopathological examination. On gross examination, the specimen of uterus showed a large ulcerative lesion measuring 7×6×1.5 cm at the uterine fundus, extending up to the serosa ([figure 4](#)). Microscopic examination of slides taken from the lesions showed squamous cell carcinoma which was breaching the uterine serosa and involving the vaginal cuff. On immunohistochemistry, the cells were immunopositive for oestrogen receptor and progesterone receptor which was suggestive of an endometrial origin of the tumour ([figure 5](#)). Bilateral tubes and ovaries were free of tumour. A definitive diagnosis of chronic uterine inversion with squamous cell carcinoma of endometrium stage IIIB was thus established.

OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative recovery and was discharged on postoperative day 6. She received adjuvant postoperative vaginal brachytherapy with three fractions of 21 cg up to 5 mm tissue depth and is presently in disease remission and on routine follow-up.

DISCUSSION

Chronic non-puerperal uterine inversion is infrequently encountered in modern clinical practice, and hence its identification and management can be a challenge. It can be caused by benign (70%) or malignant (20%) uterine tumours.¹ There are only a hundred reported cases in literature, with most reports citing a prolapsed submucosal fibroid or large polyp as the cause.² In most cases, it is thought that the underlying pathology causes weakening of the uterine wall leading to prolapse of the uterine fundus into the uterine cavity. However, in our patient, the cause of uterine inversion is not clear, and we hypothesise that squamous cell carcinoma developed in the uterine endometrium secondary to inversion.

Diagnosis is made by clinical examination and aided by imaging. On vaginal examination, an attempt must be made to palpate the cervix and assess the depth of vaginal fornices in order to differentiate it from prolapse of the uterus. In case of complete inversion, the cervix may be flush with the vagina and may be difficult to identify as seen in our patient. Further, on rectal examination, a cup-shaped dimple may be palpated, suggesting uterine inversion. An MRI or USG may suggest the diagnosis of inversion by showing the typically described U-shaped cavity or target appearance and pseudostripe, respectively.³ However, the oedematous and swollen uterus due to the vascular stasis in chronic non-puerperal cases makes the correct diagnosis by MRI difficult. In our patient, the presence of low-lying uterus on ultrasound and clinical examination findings were highly suggestive of prolapse uterus. Only on reviewing the MRI films post-operatively, the presence of dip at uterine fundus was noted and hence diagnosis of uterine inversion could be made.

The management must be individualised. The first step in the approach to an inverted uterus should be to try to bring the uterus back to its anatomical position and the second step should be hysterectomy. This surgical repositioning can be done abdominally by the technique described by Huntington and Haultain,^{4,5} and vaginally by that of Spinelli.⁶ All surgical correction procedures describe how to approach the bladder and the ureters in the constriction ring. In our patient, we used the Haultain procedure by which the constriction ring was excised posteriorly, the lower part of uterus was grasped with Allis forceps and gradually the procedure was repeated until entire correction was performed.

Endometrial squamous cell carcinoma accounts for less than 1% of all malignancies of the corpus uteri.⁷ Diagnosis of a pure endometrial squamous cell carcinoma is based on Fluhmann criteria (1928) which includes endometrial carcinoma that does not involve the cervical epithelium, there is no co-existent endometrial adenocarcinoma and there is no contiguity between the endometrial cancer and the squamous cervix epithelium.⁸ The aetiopathogenesis remains obscure, but a theory of endometrial squamous metaplasia is generally accepted. In the present patient, we think that chronic exposure of the endometrial columnar epithelium of the inverted uterus to the external environment led to squamous metaplasia followed by dysplasia and malignancy. Given the rarity of this condition, there is no consensus for the best way to manage these patients. Many different treatment options have been reported in the literature: surgical resection with or without adjuvant chemotherapy/radiotherapy.⁹ Survival data of patients affected by PESCC are scarce and controversial. The few available data suggest that these malignancies have a mixed behaviour resembling both endometrial and cervical cancer.⁷ For this reason, women with early-stage disease have a favourable prognosis, whereas in case of locally advanced cancer, survival is generally poor.⁷

To our knowledge, this is the first case in which malignancy developed in the underlying setting of an inverted uterus, hence the presentation of our case and pathological findings posed a diagnostic and therapeutic challenge to learn from.

Patient's perspective

Translated from Hindi

I have four children and no money; all my deliveries were home births and conducted by the 'dai' (traditional birth attendant). For my final childbirth, I went to the fields, sat down and pushed my baby out, the uterus also came out with him and has remained that way since then. I decided to visit the hospital now only because I am having vaginal bleeding at this late age, and my daughter brought me here. I had no problems otherwise, except difficulty during intercourse, but I have been staying away from my husband for the past 10 years.

Learning points

- ▶ Chronic non-puerperal uterine inversion is a rare entity usually caused by benign uterine tumours.
- ▶ The diagnosis may be difficult in postmenopausal women and is often confused with cervical carcinoma.
- ▶ MRI may aid in the diagnosis in difficult cases and preoperative biopsy should be performed to plan route of surgery.

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