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CASE REPORT

A. Sinha · D. Sinha · M. Aslam · M.N. Fitzgibbon

Non-puerperal complete uterine inversion caused by malignant mixed mullerian tumour of the uterus

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Abstract Non-puerperal uterine inversion is an extremely rare entity, which many gynaecologists would never come across in their lifetime. Diagnosis can be difficult. Our patient was a 91-year-old lady who presented with profuse vaginal bleeding. Hysteroscopy was unsuccessful as the cervix was completely replaced by a friable growth. A total abdominal hysterectomy and bilateral salpingo-oophorectomy was planned as a palliative measure to stop bleeding. Uterine inversion was suspected for the first time at laparotomy and confirmed after dissecting the hysterectomy specimen, which revealed both tubes and ovaries lying inside the inverted uterus. Histology showed a malignant mixed mullerian tumour of the uterus.

Keywords Uterine inversion · Non-puerperal uterine inversion · Mixed mullerian tumour

Introduction

Non-puerperal uterine inversion is an extremely rare entity, which the majority of the gynaecologists will never come across in their clinical experience. A high index of clinical suspicion is required in order to make a preoperative diagnosis of this condition. In this article we report our experience in one such case.

Case report

A 91-year-old woman was admitted as an emergency with a 3-day history of vaginal bleeding and difficulty in passing urine. She was generally fit and well prior to that. Bleeding started as light brown offensive discharge, but gradually progressed to heavy fresh bleeding. There was no associated pain or pelvic discomfort. There was nothing relevant in her past medical, surgical or gynaecological history. All her five children were born normally and she attained a natural menopause at 45 years. She was not on any regular medications. On examination, she was an alert, elderly, frail-looking woman without any physical disability. She was normotensive. Cardiovascular examination did not show any abnormality. Her abdomen was soft, non-tender without any evidence of organomegaly or ascites although her bladder was distended. Speculum examination revealed a large, necrotic, haemorrhagic mass filling the vaginal vault with heavy bleeding per vaginum. An indwelling catheter was inserted and 600 ml of urine removed. All her blood results, chest X-ray and ECG were unremarkable, except haemoglobin, which was 8 gm/dl.

An urgent examination under anaesthesia (EUA) showed that the growth had completely replaced the cervix without any identifiable cervical canal. Hysteroscopy was attempted, but was unsuccessful as no cervical opening was found. The growth was very friable and foul-smelling. Bimanually, a normal-sized, freely mobile uterus was felt without any evidence of adenexal mass, infiltration or fistula. A rectal examination was normal. The patient bled profusely during the examination. A biopsy was taken and the vagina was packed to stop bleeding. An ultrasound scan showed a fairly well-defined solid heterogenous mass, possibly uterine, arising out of the pelvis (7.8×9.7×6.7 cm). There were a few small cystic areas within the mass. Neither ovary was seen. There was no free fluid or para-aortic lymph node enlargement. The gall bladder, kidneys and spleen were unremarkable and the liver looked rather bright, but no focal lesions were noted. Histological examination of biopsy showed evidence of a malignant mixed mullerian tumour (carcino-sarcoma) of uterine origin. Systemic antibiotics were given. An attempt to correct her anaemia with repeated blood transfusion was ineffective as she continued to bleed heavily.

A. Sinha (✉) · M. Aslam · M. Fitzgibbon
Obstetrics and Gynaecology, Wordsley Hospital,
West Midlands, UK
e-mail: kanhaiyakrishna@hotmail.com
Tel.: +44-1384-351060

D. Sinha
Obstetrics and Gynaecology, City Hospital,
Birmingham, B18 7QH, UK

Three days after her admission her case was discussed at the multidisciplinary meeting and it was decided to perform total abdominal hysterectomy and bilateral salpingo-oophorectomy as a palliative measure to stop the bleeding. Unfortunately, MRI could not be arranged before the operation. At laparotomy the uterus was neither seen nor felt. A small depression was noted in the centre of the pelvis with broad ligaments fanning around it. Two cord-like structures were also noted to be entering the depression, which on palpation felt like round ligaments. On introducing an index finger into the pit a solid structure was felt, which, with some firm upward pressure from below, was pushed out of the pit. This looked like cervix with friable growth all around. At this point uterine inversion was suspected. The broad ligament was opened after dividing the round ligaments on either sides, the uterovesical pouch was opened, the uterus identified and the bladder was pushed down without difficulty. We started placing clamps as is usually done in a hysterectomy. As each pedicle was cut more and more of the uterus came into sight, we still could not find the adenexae on either side. While the last pedicle was being severed to complete hysterectomy, an extremely friable and necrotic growth arising from the fundus became detached and fell into the vagina. The mass measured about 6–7 cm and was sent separately for histological examination. Dissection of the specimen confirmed the diagnosis of uterine inversion as both ovaries and tubes were lying within the uterus. Inside the uterus adjoining the cervix two yellowish, discrete nodules measuring about 2 cm were noted. The friable growth was mainly from the fundus and some from the cervix.

Histology confirmed a malignant mixed mullerian tumour (carcino-sarcoma) of uterine origin with involvement of cervix. Due to positive peritoneal washings, the disease was staged as FIGO (International Federation of Gynaecology and Obstetrics) Stage III. The tumour extension into the cervix consisted mainly of the epithelial component. There were also multiple psammomatous calcifications. The yellowish nodules were reported to be necrotic tumour nodules. Both ovaries and tubes were clear of disease. The patient made an excellent recovery in the postoperative period. She refused any further treatment (chemotherapy or radiotherapy). Unfortunately she died 3 months postoperatively due to pulmonary embolisation.

Discussion

Non-puerperal uterine inversion is an extremely rare condition and presents as a diagnostic challenge to gynaecologists. They are often caused by leiomyomas, but rarely by sarcomas or carcino-sarcomas. A small proportion may also be idiopathic. Mwinyoglee et al. [1] have described that, of the 77 cases reported in the English-language literature, 75 (97.4%) were produced by tumours and 20% of these tumours were malignant.

Uterine inversion can be complete, incomplete or total. In the incomplete variety the fundus remains within the uterine cavity, whereas in complete uterine inversion the fundus passes through the external os. With total inversion the fundus protrudes through the vulva and may be accompanied by inversion of the vagina [2]. In complete uterine inversion the external cervical os may become constricted and this may cause venous stasis, which may lead to pulmonary embolism, not to mention infection, necrosis and chronic anaemia [3].

The aetiology of uterine inversion includes thinning of the uterine wall at tumour implantation and also gradual relaxation of the musculature at the point of attachment of the pedicle. This is followed by generation of expulsive forces by the uterus in order to empty the cavity, the cervix then dilates and the tumour extrudes into the vagina.

The main symptoms of non-puerperal uterine inversion are vaginal bleeding, foul-smelling vaginal discharge, abdominal pain and urinary disturbances. Skinner and Louden [2] reported a case in a patient who presented with worsening menorrhagia, dysmenorrhoea and dyspareunia due to an atypical leiomyoma. Our case presented with a short history of severe vaginal bleeding and urinary hesitancy. The diagnosis of uterine inversion can be difficult and is often made at the time of laparotomy, as in our case. However, every attempt should be made to make the diagnosis before operation to allow appropriate planning of the operation, thus avoiding complications. For correct diagnosis there are two important criteria: difficulty demonstrating the cervical canal along with a mass in the vagina, and non-palpation of the uterine fundus on bimanual examination. Cases have been described in the literature in which the diagnosis was made before the operation solely on the basis of clinical findings [1, 4].

In our case diagnosis was not suspected even on EUA as the mass was quite mobile and the inverted cervix gave the impression of a small postmenopausal uterus. Even an ultrasound scan was not very helpful in our case. An MRI scan would have been much more valuable in defining uterine morphology. The signs of uterine inversion on T2 MRI scans are a U-shaped uterine cavity and a thickened inverted uterine fundus on a sagittal image and a “bulls-eye” configuration on an axial image. Takano et al. [5] reported a case of uterine inversion due to leiomyosarcoma in which a T1-weighted dynamic MRI enhanced by gadolinium revealed an extruded tumour in the vagina and a high intensity U-shaped uterine cavity. The same authors reported another case in which MRI showed a high-intensity, U-shaped uterine cavity and an inverted uterus, and also recommended it as a very useful diagnostic tool in cases of inverted uterus Fig. 1.

Preoperative intravesical ultrasonography (IVU) should be performed to delineate any associated abnormalities of the urinary tract. Irregular bladder mucosa and a dilated pelvi-calyceal system can be demonstrated due to pressure effect.

After establishing the diagnosis a total abdominal hysterectomy and bilateral salpingo-oophorectomy is usually recommended for surgical staging as well as for the removal of the disease [6]. However, a vaginal route for hysterectomy and bilateral salpingo-oophorectomy has

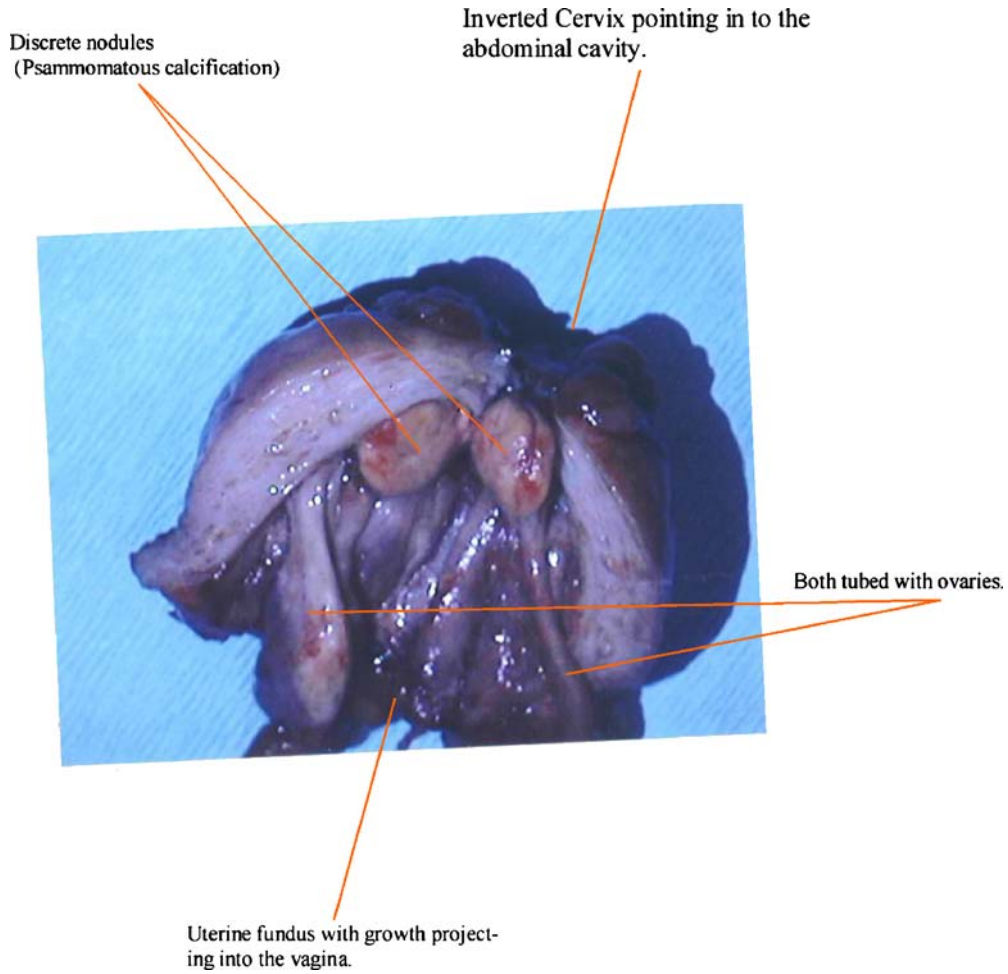

Fig. 1 Figure shows a case of uterine inversion depicting the inverted cervix, discrete nodules and uterine fundus with growth projecting into the vagina

also been used. Laparoscopy can be performed prior to vaginal hysterectomy to establish the diagnosis and the structures within the cup of inversion or to find out associated pelvic pathology. Rattray et al. [4] reported a case in which a combined abdominovaginal approach was taken; the tumour bulk was removed vaginally to enable an easier abdominal hysterectomy.

Our case was quite interesting as the diagnosis was suspected for the first time at laparotomy and confirmed by finding tubes and ovaries inside the dissected specimen. To our knowledge this is the first reported case in which both tubes and ovaries have been found within the inverted mass. There has been a case report showing elongated round ligaments and fallopian tubes within the cup of inversion, but the ovaries were outside. Adjuvant chemotherapy has also been recommended in the management of uterine inversion caused by sarcoma. Hanprasertpong et al. [7] reported an unusual case of chronic non-puerperal inversion caused by malignant mixed mullerian tumour that was successfully treated by surgery and radiotherapy.

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