Gynecology & Obstetrics
ISSN: 2161-0932

Aung and Hill, Gynecol Obstet 2013, 3:4
DOI: 10.4172/2161-0932.1000167

Case Report Open Access

Spontaneous Rupture of the Uterus Associated with Mixed Mullerian Tumours

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Abstract

Spontaneous rupture of the uterus is a rare occurrence. We report a case of uterine rupture associated with a mixed mullerian tumour which presented as a acute abdominal emergency. We further discuss a review of the world literature.

Keywords: Uterus; Pyometria; Mullerian tumour

Introduction

Pyometra is a rare condition predominantly affecting elderly women involving the accumulation of purulent material in the uterine cavity. Although often resulting from benign causes such as leiomyomas, infection and congenital cervical anomalies, it can also be associated with genital malignancy (most commonly cervical) and the subsequent radiation cervicitis resulting from treatment. We present a case of a 61 year old patient who presented acutely and was found to have a ruptured pyometra. Furthermore, histological analysis of specimens obtained at laparotomy revealed a rare utero carcinosarcoma or mixed mullerian tumour of the uterine corpus.

Patient Review

A 61 year old woman Para 2 who has post menopausal for five years. She presented with a three week history of offensive red/brown vaginal discharge and abdominal pain localised to the left iliac fossa. She had a past medical history of ischemic heart disease, left bundle branch block, stage 3 chronic kidney disease, type II diabetes and hypertension.

Initial ultrasound scan showed an anteverted bulky uterus with an endometrial thickness of 6.5mm and no obvious adnexal masses.

Examination revealed a soft and non-tender abdomen and a black fungating lesion over the cervix with offensive vaginal discharge which bled on smear testing. She was put on an urgent list for hysteroscopy and biopsy.

Whilst awaiting the procedure she presented to the emergency department with a three day history of increased abdominal pain, severe bloating, nausea, vomiting and absolute constipation for 7 days.

On arrival, she was apyrexial. Examination revealed a distended abdomen, which was globally tender with guarding and absent bowel sounds. Her erect chest x-ray and abdominal x-ray showed no abnormalities. Apart from a raised white blood cell count of 16.3×10^6, all her blood tests were normal. A CT scan of her abdomen showed a perforated uterus and pyometra.

At laparotomy one litre of free pus was found in the abdominal cavity. The uterus was 18 weeks in size with a fundal perforation. A total abdominal hysterectomy and bilateral salpingo-oophrectomy was performed.

The uterine specimen weighed 1100 g and measured 180×110×70 mm. The lumen was occluded by a degenerate tumour mass measuring 180×110 mm which occupied the entire body of the uterus and infiltrated through the serosal surface at the fundus. The cervix was dilated around the mass which protruded into the endocervical canal. Both ovaries were macroscopically normal.

Further detailed examination of the uterus confirmed a diffusely invasive malignant tumour obliterating the uterine cavity and infiltrating through the muscular wall. The tumour contained complex atypical glands, some showing squamous metaplasia. There were wide areas of coagulative necrosis noted amounting to more than 50% of the tumour mass. No heterologous elements were seen. The tumour had breached the serosal surface macroscopically and lymphovascular invasion was seen to be present. The histological features were consistent with uterine carcinosarcoma with a predominant leiomyosarcoma component.

The patient was admitted to ITU overnight but subsequently made an uneventful post-operative recovery.

Discussion

Pyometra is defined as the accumulation of purulent material in the uterine cavity. Although rare in the younger population (0.01-0.5% in gynaecological patients), its incidence is as high as 13.6% in elderly women [1]. It occurs as a result of impaired natural drainage of the cervix, most commonly secondary to malignant disease of the genital tract and subsequent radiation. Benign tumours, puerperal infection and congenital cervical anomalies have also been implicated. Spontaneous rupture of the uterus in conjunction with pyometra is an extremely rare event with only 30 cases reported within the English literature to our knowledge [2].

Clinically patients may complain of purulent vaginal discharge, post-menopausal bleeding and lower abdominal pain however there are several reports of the condition presenting as an acute abdomen with nausea and vomiting [3-6], all of which were seen in our patient. In the majority of patients a ruptured pyometra is diagnosed intra-operatively although in a few cases, computed tomography and magnetic resonance imaging is useful in diagnosis. With the exception of one patient immediate laparotomy has been the mainstay of treatment in all reported cases with peritoneal lavage or drainage performed usually with hysterectomy [6].

Cervical carcinoma is the most common malignancy to cause

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Received August 24, 2013; **Accepted** September 07, 2013; **Published** September 12, 2013

Citation: Aung C, Hill NCW (2013) Spontaneous Rupture of the Uterus Associated with Mixed Mullerian Tumours. Gynecol Obstet 3: 167. doi:10.4172/2161-0932.1000167

a ruptured pyometra. Histology from the specimens obtained from our patient revealed changes consistent with uterine carcinosarcoma otherwise known as a mixed mullerian tumour of the uterine corpus. A review of literature has found only two cases of endometrial cancer associated with a ruptured pyometra [7,8].

Mixed mullerian tumours refer to the wide spectrum of rare neoplasms comprising of both epithelial and mesodermal components. Either one or both of these cell types may show changes ranging from mild atypical to frank malignancy. Although the wide variation in histological features often leads to confusion in classification, malignant mixed mesodermal tumours usually refers to those containing heterologous elements i.e. skeletal, bone and cartilage tissue whereas carcinosarcoma refers to tumours with a homologous mixture of carcinomatous and sarcomatous elements derived from tissues normally found within the Mullerian system [9].

Due to the variation in classification of such tumours, it has been difficult to determine their true incidence however studies suggest that they account for less than 5% of all uterine malignancies. Diagnosis of these tumours can also prove difficult. Cytological evaluation is negative in up to 45% of cases and histological diagnosis from endometrial curettings may only be of help in up to 50-70% of cases due to the small amount of tissue obtained and to the presence of necrosis and inflammation of the tumour tissue [9]. Ultimately hysteroscopy and biopsy under visual guidance appears to be the most accurate method of obtaining an initial diagnosis [9].

Characteristically these tumours are associated with a poor prognosis as they tend to metastasise hematogenously with many patients found to have extra-uterine disease at surgery despite clinically appearing to have disease confined to the uterus [10]. Initial tumour stage is currently the only reliable prognostic factor with depth of myometrial invasion and lymph-vascular space invasion being significant predictors of extra-uterine disease [10]. At present definitive management for mixed mullerian tumours has not been established although due to its aggressive nature, treatment frequently involves the combination of surgery with radiotherapy and/or chemotherapy.

Conclusion

We present a rare case of a spontaneously ruptured pyometra in combination with an unusual endometrial carcinoma. We believe that this is the first case of its kind and also adds to the existing case series of spontaneously ruptured pyometra to highlight the importance of suspecting this diagnosis in elderly female patients presenting with an acute abdomen.

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