Adenoma Malignum Presenting With Profound Hyponatremia

Kelsey E. Mills, MD, FRCSC,[1] Paul Shuen, MD, FRCSC[2] Lynne Zolis, MD, FRCSC[2]

[1]Department of Obstetrics and Gynaecology, University of Toronto, Toronto ON

[2]Department of Obstetrics and Gynaecology, North York General Hospital, Toronto ON

Abstract

Background: Adenoma malignum of the cervix (also referred to as minimal deviation adenocarcinoma) is a rare malignancy. Although previous reports have described adenoma malignum presenting with mucinous vaginal discharge, no reports to our knowledge have described a presentation with profound hyponatremia due to fluid losses.

Case: We present a case of adenoma malignum in a 52-year-old woman who presented with substantial watery vaginal discharge and profound hyponatremia.

Conclusion: Despite being a rare cervical tumour, adenoma malignum should be considered in the differential diagnosis of watery vaginal discharge. This tumour can present with severe electrolyte disturbances.

Résumé

Contexte : L'adénome malin du col utérin (aussi connu sous le nom d'adénocarcinome à déviation minime) constitue une tumeur maligne rare. Bien que des signalements précédents aient indiqué que l'adénome malin s'accompagnait d'un écoulement vaginal mucineux, nous n'avons trouvé aucun signalement décrivant la présence concomitante d'une profonde hyponatrémie attribuable à des pertes liquidiennes.

Cas : Nous présentons un cas d'adénome malin chez une femme de 52 ans qui connaissait un écoulement vaginal aqueux substantiel et une profonde hyponatrémie.

Conclusion : Bien qu'il s'agisse d'une tumeur cervicale rare, l'adénome malin devrait être pris en considération dans le diagnostic différentiel de l'écoulement vaginal aqueux. Cette tumeur peut s'accompagner de graves perturbations de l'équilibre électrolytique.

J Obstet Gynaecol Can 2015;37(7):624–627

Key Words: Adenoma malignum, minimal deviation adenocarcinoma, cervical cancer, electrolyte imbalance

Competing Interests: None declared.

Received on July 29, 2014

Accepted on August 21, 2014

INTRODUCTION

Adenoma malignum of the cervix, also described as minimal deviation cervical adenocarcinoma, is a relatively rare form of cervical adenocarcinoma. Studies have reported an incidence of approximately 1% of all cervical adenocarcinomas.[1–3] This tumour has been described in several case series, and in these cases the most common symptoms were profuse mucinous vaginal discharge and vaginal bleeding.[2–6] This tumour has also been associated with Peutz-Jeghers syndrome (hereditary intestinal polyposis).[7–9]

To our knowledge, adenoma malignum presenting with severe electrolyte disturbances has not been previously reported. We describe here the case of a woman with adenoma malignum who presented with profound hyponatremia and hyperkalemia, and we describe the subsequent surgical management.

THE CASE

A 52-year-old woman of Korean ethnicity, gravida 3 para 3, presented to the emergency department at our hospital with a history of nausea and vomiting for several days, without diarrhea or fever. She was believed to have gastroenteritis and was admitted to the internal medicine service because of dehydration.

At the time of admission, she described having a watery vaginal discharge that had been present over the preceding two to three years, and that had worsened over the past few months. She had observed some blood in the discharge. Her final menstrual period had been more than two years before admission. She had a history of mild asthma controlled with inhaled salbutamol. She had previously undergone three Caesarean sections via midline laparotomy

for unknown indications. She stated that she had never had a pelvic examination or a Pap smear.

She described some recent loss of appetite and generalized fatigue, but no significant weight loss. The patient was ambulatory, but occasionally felt dizzy. She felt generally unwell. After admission, it was determined that the patient had urinary retention, and a Foley catheter was placed. Her serum sodium was low (125 mmol/L). She also underwent transvaginal pelvic ultrasound assessment that revealed an anteverted uterus measuring 14 × 3.4 × 6.6 cm. The endometrial diameter was 5 mm at the fundus. The ovaries appeared normal. A lower uterine segment mass measuring 12.4 × 9.8 × 13.8 cm and containing solid and cystic components was noted; this was felt most likely to be a degenerating fibroid.

The gynaecology service was consulted, and during a speculum examination it was estimated that several hundred millilitres of watery mucinous fluid drained from the vagina. The pelvic examination revealed a large vaginal mass consisting of copious gelatinous material. The cervix could not be clearly seen. A methylene blue dye tampon test (to rule out a vesico–vaginal fistula) was negative.

The patient underwent an abdominal and pelvic MRI with gadolinium for better characterization of the vaginal mass. MR images identified a multiloculated, predominantly cystic central lesion measuring 8.9 × 8.6 × 10 cm, located within the cervix and extending into the lower uterine segment. No inguinal or iliac lymphadenopathy was seen. The differential diagnosis from the MRI included giant nabothian cysts, cervical polyps, adenoma malignum, and mucinous carcinoma of the cervix (Figure 1).

The patient was seen in consultation by a gynaecologic oncologist, and the decision was made to proceed with surgical management. The patient returned to the emergency department at our hospital approximately one week after her discharge from the internal medicine service, and several weeks before the date of her planned surgery. She presented with nausea, vomiting, headache, and malaise. She was found to be markedly hyponatremic, with a sodium level of 107 mmol/L. At that time, her serum potassium was elevated at 5.7 mmol/L, and serum creatinine was 109 mmol/L (from a baseline of 60 mmol/L). Her electrocardiogram was normal. She was re-admitted and was seen in consultation by the nephrology service. The hyponatremia and hypovolemia were presumed to be due to the loss of total body water from her continuing copious vaginal discharge.

The patient's hyponatremia was slowly corrected by the nephrology service with IV fluids and free water restriction. Because of concern about her serum electrolyte disturbances, the decision was made to proceed with surgery immediately to remove the mass. She underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy, left ureterolysis, and lysis of adhesions.

At surgery, the mass was initially not visible in the pelvis. After the vesico–uterine peritoneum was mobilized and the uterine artery pedicles had been suture ligated, the mass came into view as traction was applied to the uterus (Figure 2). The left ureter was completely mobilized from the level of the common iliac artery to the ureteric tunnel in order to proceed safely with the surgery. The rectovaginal space was developed and the vaginal mass was slowly drawn up into the pelvis. An anterior colpotomy was performed and the entire specimen of uterus, cervix, vaginal mass, and both adnexa was transected from the vaginal vault (Figure 3). The remainder of the procedure was unremarkable. The patient's postoperative course was unremarkable, and her serum electrolytes remained within the normal range.

The final pathologic diagnosis of the surgical specimen was a mucinous cervical neoplasm with features consistent with minimal deviation adenocarcinoma (adenoma malignum). No lymphovascular space invasion was noted. There was no involvement of exocervical or paracervical soft tissue margins. This case was reviewed by the gynaecologic oncology and pathology departments at a tertiary hospital, and it was concluded that no adjunctive treatment was required. Postoperatively, the patient has remained well.

DISCUSSION

The management of patients with adenoma malignum can be challenging because it is a rare tumour and because it is considered well differentiated; consequently, it may be difficult to differentiate it from benign endocervical glands.[3,10] In addition, unlike more common squamous carcinomas or adenocarcinomas of the cervix, this tumour is often not identified by routine cervical cytology screening because of its endophytic rather than exophytic growth pattern.[3,9] One report described cytology findings consistent with inflammation in patients who had confirmed diagnoses of adenoma malignum.[11]

Presenting with electrolyte disturbances due to this tumour is clearly unusual. A previous report also described an unusual presentation of this tumour, with copious fluid leakage that was presumed to be due to urinary incontinence but which was in fact leakage of mucoid fluid from the tumour.[10] In that case, a 45-year-old woman had presented with an 11-year history of presumed urinary leakage, and