



NON-PUERPERAL UTERINE INVERSION COMPLICATING THE TWISTING OF A PROLAPSED FUNDAL SUBMUCOUS FIBROID: A CASE REPORT IN YAOUNDÉ, CAMEROON

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ABSTRACT

Uterine inversion (UI) is a rare complication of the post-partum. Occurrence of UI out of the puerperal context is rarer and very few gynaecologists will be lucky to encounter and manage a case during their careers. To the best of our knowledge, only one case has been reported in Cameroon so far. We herein report the case of a 45 year old Gravida 9 Para 4.0.5.4 woman admitted in our emergency unit with a stage 3 non-puerperal uterine inversion (NPUI) complicated with hemorrhagic shock. The NPUI followed the twisting of a prolapsed fundal submucous fibroid. She was adequately resuscitated (including blood transfusion). Then, she underwent total abdominal hysterectomy with conservation of ovaries, after reduction of the inversion through posterior vaginal colpohysterotomy. Post-operative course was uneventful. Histology of the specimen revealed no malignancy. Risk factors, etiopathogeny, signs and symptoms and management of NPUI are reviewed.

INTRODUCTION

Uterine Inversion (UI) refers to descent of the uterine fundus to or through the cervix, so that the uterus is literally turned inside out [1]. Depending on the period of onset, UI is either puerperal (obstetrical) or non puerperal (gynaecologic) [2]. Uterine inversion is classified into four stages: stage1: the inverted fundus remains in the uterine cavity; stage 2: complete inversion of the fundus through the cervix; stage 3: the inverted fundus protrudes through vulva and stage 4: inversion of the uterus and vaginal wall through the vulva [1, 2]. Some authors simply classified UI as incomplete (when the inverted fundus doesn't cross the cervix) or complete (when the inverted fundus

protrudes beyond the cervix) [3]. UI is acute when it is diagnosed within 24 hours of onset; It is sub-acute when diagnosed between the first and the 28th day after onset; it is chronic when diagnosis is made more than 28 days after onset [4]. Puerperal UI accounts for 85.8% of cases and is the consequence of inadequate management of the third stage of labour while Non Puerperal Uterine Inversion (NPUI) is rarer and more difficult to diagnosed and treat [1,2]. We herein report a case of NPUI caused by the twisting of a prolapsed fundal submucous fibroid in a 45 year old Gravida 9 Para 4.0.5.4 woman.



PATIENT AND OBSERVATION

M.S. 45 year old Gravida 9 Para 4.0.5.4 was admitted in our emergency unit for a sudden protrusion of a mass through the vulva during micturition 3 hours earlier. This was associated with profuse bleeding, intense pelvic pain and pronounced asthenia. History revealed that she underwent twisting of a prolapsed fundal fibroid (15 centimetres of diameter) 26 days prior to admission. The stump of the pedicle was sutured but the procedure was complicated by massive haemorrhage that prompted transfusion of 1000 millilitres (ml) of whole blood. [Figure 1]. She was discharged two days later and postoperative course was marked by light haemato-purulent vaginal discharge from day 6 till admission into our unit. This was managed by self-medication with metronidazole (1500 milligrammes/day) and amoxicillin-clavulanic acid (2 grammes/day). Histology of the specimen was not available. Due to lack of financial means and clear instructions on danger signs, she did not go back to her treating doctor. On physical exam her general condition was good. Vital parameters were: blood pressure of 70/55 millimeters of mercury, pulse rate of 112 pulsations/minute, respiratory rate of 26 cycles/minute and

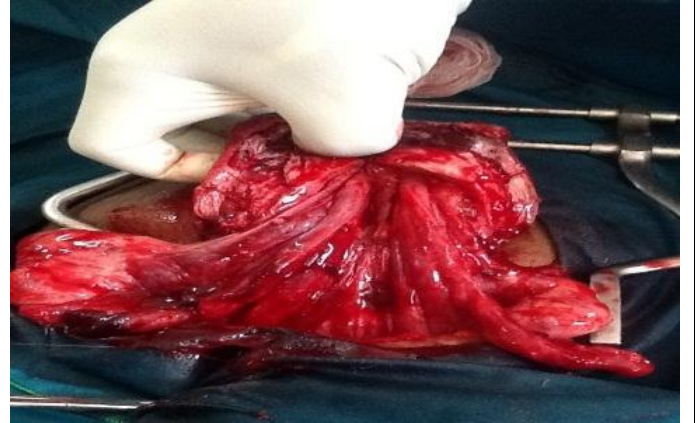
temperature of 37.8 degrees Celsius. Conjunctivae were very pale. On inspection, there was a 10 centimetres reddish mass protruding out of the vulva with several suture threads on its inferior pole. That mass was covered with necrotico-purulent secretions (Figure 1). On digital exploration, that mass was firm, tender and in continuity with vaginal walls. The vagina was shortened and cervical rim was not palpated. Internal genital organs were not palpated through the rectum.

The working diagnosis was NPUI complicated by hemorrhagic shock. The workup revealed normocytic normochromic anemia with a haemoglobin level of 7.4 grammes/litre and a normal clotting profile. After resuscitation (fluids, blood transfusion, oxygen and painkillers), surgery was done with antibiotherapy (ofloxacin and metronidazole). The first part of the procedure was through vaginal route and consisted of a median colpohysterotomy followed by reduction of the UI (Figure 2). The second part was via laparotomy and consisted of total abdominal hysterectomy with conservation of ovaries. Postoperative course was uneventful and the patient was discharged seven days later. Histology of the specimen revealed no malignancy.

Fig 1. View of the inverted uterus protruding out of vulva with necrotico-purulent zones and surgical threads



Fig 2. Abdominal view of the pelvis after reduction of the inverted uterus via posterior colpohysterotomy



DISCUSSION

To the best of our knowledge, only one case of NPUI has been reported in Cameroon so far [5]. Indeed, NPUI is a very rare pathology that very few gynaecologists will encounter and manage during their careers [1]. Given that only few cases have been reported worldwide, the incidence of NPUI is unknown [1, 2].

In our case, the NPUI was caused by the twisting of a prolapsed fundal submucous fibroid. It has been reported that 71.6 – 85% of NPUI are due to submucous fibroids [1, 2]. In one out of five cases, the intra-uterine tumour causing NPUI is malignant (mainly sarcomas)[2, 6]. Thus, it is recommended to obtain frozen section histology of the inverted fundus/mass before reduction to better manage the patient in case of malignancy. We did not carry out a frozen section for three reasons: the patient

was in shock, the inverted uterus was not macroscopically suspect of malignancy and frozen section histology was not available. The presence of a submucous fibroid (or any other mass) is not sufficient to cause NPUI. It must be fundal, pedunculated and big enough to dilate the cervix by distending the uterine cavity [7]. Our patient had a pedunculated and prolapsed submucous fibroid of 15 cm of diameter. The twisting certainly caused/worsened the NPUI by suddenly emptying the uterine cavity and pulling the fundus downwards. This corroborates the theory stating that sudden emptying of a distended uterine cavity is the mechanism of onset of UI [6].

Our patient was therefore at high risk of developing NPUI following the procedure. Thus her treating doctor should have given her clear (and written)

instructions on danger signs with early postoperative appointment. The extreme rareness of NPUI can explain why this was not done. Reporting such cases will raise awareness on NPUI among practitioners in our milieu. On her side, instead of going back to the doctor for proper management of her abnormal vaginal discharge the patient underwent auto-medication for more than three weeks. This attitude is frequent among patients in our environment and can be explained by poverty, inappropriate postoperative counselling and beliefs.

NPUI usually presents with vaginal bleeding, pelvic pain, and urinary symptoms. Our patient had metrorrhagia for three weeks with sudden worsening during painful protrusion of the uterus out of the vulva. It was an easily diagnosed stage 3 NPUI. The diagnosis of NPUI is difficult at early stages and may require pelvic ultrasonography, computerised tomography and magnetic resonance imaging [1, 2].

Given the patient's age and with her informed consent we carried out a radical procedure. Reduction via laparotomy (following Huntington's or Haultain's techniques) is indicated for NPUI at early stages with early diagnosis [8, 9]. We first carried out a reduction through vaginal route (as per Kustner's technique): median posterior colpohysterotomy, reduction and repositioning of the uterus in the abdominal cavity and colpography [10]. The second technique of reduction via vaginal route (described by Spinelli) encompasses anterior colpohysterotomy and dissection of the vesico-uterine fold [10]. The latter has a high risk of injury to the bladder and the ureters because NPUI modifies the normal anatomy. For the same reason, vaginal hysterectomy is risky in case of NPUI [10]. After reduction following Kustner's technique we did a total abdominal hysterectomy with conservation of ovaries. Outcome was favourable in our case because of rapid management by an experienced gynaecologist surgeon.

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It is worth noting that management of NPUI at early stages can be done by laparoscopically assisted vaginal hysterectomy. This approach reduces the risk of bleeding (laparoscopic devascularisation of the uterus) and the risk of injury to the urinary tract. Moreover, coelioscopy confirms the diagnosis and assesses the degree of ischemia of the uterus [11]. However this approach is difficult to implement in resource-constrained countries like Cameroon.

CONCLUSION

This case illustrates that NPUI (despite its rareness) occurs in a specific population (big intra-uterine tumours). A good knowledge of its etiopathogeny can be used to predict its onset. Successful surgical management of NPUI is delicate and requires not only experience but also specific skills from the gynaecologic surgeon.

AUTHOR'S CONTRIBUTIONS

Professor Nana, Drs Tompeen and Eko, managed the case and wrote the manuscript. Dr Esiene is the anesthetist/ intensive care specialist who managed the case and she wrote the manuscript. Drs Fouogue, Fouelifack, Essiben and Fouedjio followed up the patient and wrote the manuscript. Professors Nana and Mbu supervised the whole team.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interests for this manuscript.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.



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