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Acute Non-Puerperal Uterine Inversion About A Case

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Abstract **Case Report**

We report a case of uterine leiomyosarcoma initially diagnosed on pathological examination of a vaginally performed interannexal total hysterectomy specimen for a giant 10 cm/8 cm fibroid delivered through the cervix prolapsed out of the vulva and causing acute neglected uterine inversion in a 30-year-old nulliparous patient with a history of a 4x3x2 cm submucosal myoma on pelvic ultrasound.

Keywords : Neglected acute non-puerperal uterine inversion; uterine leiomyosarcoma; total vaginal hysterectomy; submucosal myoma; giant fibroid delivered through the cervix; uterine cervix.

INTRODUCTION

Acute non-puerperal uterine inversion is a rare clinical situation. Leiomyosarcoma is a possible etiology. Given the urgency of the situation in most cases, especially hypovolemic shock, vaginal total hysterectomy is the safest and most reliable surgical technique.

Uterine leiomyosarcoma is a rare malignant conjunctival tumor with a poor prognosis, representing between 2 and 6% of malignant tumors of the uterine body [1, 2]. It is the most frequent type of uterine sarcoma, developing from the myometrium and most often presenting as a banal myoma or in necrobiosis. Anatomopathological diagnosis is often made on the surgical specimen. The gold standard of treatment is surgery, while the role of adjuvant therapies remains debated [1, 3, 4].

CASE REPORT

A 30-year-old nulliparous patient with a history of submucosal myoma measuring 4x3x2 cm, discovered on pelvic ultrasound one year ago, presented to the gynaecological emergency department with a 2-day history of a mass outside the vulva, pelvic pain and genital haemorrhage.

General examination revealed a pale facies, arterial hypotension 90/50 mmHg, heart rate 150 bpm and dizziness. Gynaecological examination revealed a whitish, hard, bumpy, irregular, pedunculated mass, externalized outside the vulva, measuring 10/8 cm, independent of the vaginal walls and connected to the left side of the cervix prolapsed outside the vulva by a thick pedicle, clinically suggestive of a giant uterine fibroid delivered through the cervix prolapsed outside the vulva. Examination revealed incipient necrosis of the prolapsed cervix. Attempts to reduce and reposition the mass were unsuccessful.

An emergency laboratory workup showed a hemoglobin of 7 g/dl. The patient was transfused with 4 packed red blood cells, and an indwelling urinary catheter was inserted.

Suprapubic ultrasound revealed no uterus or ovaries, but an intravaginal mass. The diagnosis retained after clinical examination and pelvic ultrasound was that of acute neglected uterine inversion secondary to a giant submucosal fibroid delivered through the cervix prolapsed outside the vulva, given the patient's history.

The patient was admitted to hospital and her hemodynamic status was restored. The decision to perform a total inter-annexal vaginal hysterectomy was taken after consultation and consent from the patient.

The patient was transfused during the operation, which began with exeresis of the giant fibroid, reduction and repositioning attempts were again unsuccessful, and the hysterectomy began with an anterior colpotomy with

careful vesico-uterine detachment, followed by an annular incision of the posterior lip of the uterine cervix and posterior detachment of the douglas cul de sac, then ligation-section of the right and left uterosacral ligaments, then ligation-section of the right and left uterine arteries, then opening of the vesico-uterine peritoneum and douglas, then ligation-section of the right and left round ligaments, then ligation-section of the right and left utero-ovarian pedicles, and finally a total inter-annexal hysterectomy and closure of the vaginal slice. The operation was carried out without any notable incident. Surgical specimens were sent for anatomopathological study.

Post-operative monitoring showed an improvement in the patient's condition, with hb=8.6g/dl two days after surgery. Given the positive evolution, the patient was discharged from hospital on the fifth day after the operation.

Pathology of the surgical specimens showed a leiomyosarcoma, and the patient underwent further work-up concluding in stage 1 leiomyosarcoma. After discussion of the patient's case, chemotherapy and pelvic radiotherapy were ruled out, and it was decided to see the patient every 3 months for 3 years, then every 6 months for 3 years, then once a year for up to 10 years.

Figure 1: Uterine leiomyosarcoma presenting as a giant 10/8 cm uterine fibroma delivered through the cervix which is also prolapsed out of the vulva

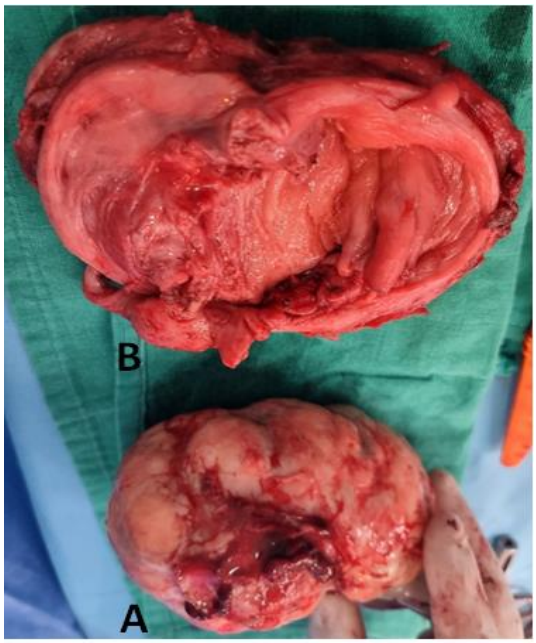

Figure 2: Leiomyosarcoma presenting as a uterine fibroid delivered through the cervix (A), inverted uterus excised (B)

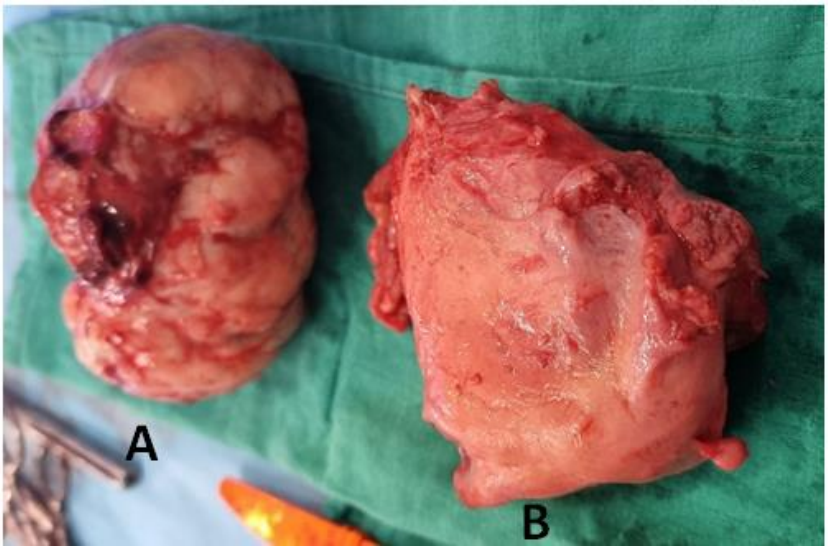

Figure 3: Surgical specimens from vaginal myomectomy (A) and total hysterectomy (B) sent for anatomopathological study. The uterus is reshaped after excision

DISCUSSION

Uterine inversion is defined as the inverting of the uterus into a finger or "uterine invagination". It is a rare pathology, more frequent in obstetrics than in gynecology. Uterine inversion is serious, and can be life-threatening due to the hypovolemic shock it causes [5]. In the majority of cases, this involved post-menopausal women or women over 45 years of age [6].

Several factors are implicated in the pathophysiology of non-puerperal uterine inversion : the presence of a uterine tumor ; localized preferentially on the uterine fundus; on a thin uterine wall; with a small tumor pedicle ; rapid tumor growth ; and cervical dilatation by distension of the uterine cavity [7-10].

The etiology found in 70 to 85% of cases of gynaecological uterine inversion, depending on the author, is submucosal myoma [11]. In 15 to 30% of cases, malignant tumors are involved, with uterine sarcomas (leiomyosarcoma, embryonal rhabdomyosarcoma, endometrial stromal sarcoma) at the forefront.

Depending on severity, there are four degrees of inversion [11]: first degree : the uterine fundus is depressed into a "cul de fiole" or cupula ; second degree: the inverted uterus passes through the external orifice of the cervix; third degree: the uterine body becomes intravaginal and may become completely externalized; fourth degree or total inversion: the vaginal walls participate in the inversion.

Two other classifications have been proposed for non-puerperal uterine inversion : partial or total inversion [12] and acute or chronic inversion according to the onset and evolution of symptoms [13, 14].

Acute non-puerperal uterine inversion is often dramatic. It is characterized by increased pain intensity and heavy menorrhagia [15]. Chronic non-puerperal uterine inversion is characterized by pelvic discomfort, vaginal discharge, minimal menorrhagia and severe anemia [14, 16, 17].

Management involves prior medical reanimation with restoration of blood volume and broad-spectrum antibiotic therapy, followed by surgical management. Hysterectomy is virtually indispensable in cases of 3rd and 4th degree uterine inversion [18].

The laparoscopic-vaginal approach already described by the Auber *et al*., team [19] seems to be a good alternative for confirming the diagnosis, assessing the degree of ischemia of the adnexa and vagina, and devascularizing the uterus laparoscopically by controlling the uterine pedicle from its origin.

In the literature, uterine artery embolization is indicated in chronic non-puerperal uterine inversion, generally of the 2nd and 3rd degree, and in acute puerperal inversion reducible to conservative treatment. Preoperative embolization in acute non-puerperal uterine inversion may be useful to limit blood loss.

Uterine leiomyosarcoma is a rare conjunctival malignancy arising from the mesenchymal elements of the myometrium [20]. The relative frequency is 1.3% of all uterine cancers [21] and corresponds to 40-50% of uterine sarcomas. There is Classical uterine leiomyosarcoma and the epithelioid and myxoid variants.

There are 2 leiomyosarcomas per 1,000 uterine fibroids [21, 22]. They are characterized by great anatomopathological heterogeneity. Their prognosis is poor, with a five-year survival rate of around 30% [1, 23]. They must be diagnosed early, as patient survival correlates with tumour stage [1]. Recurrence rates for

leiomyosarcomas vary from 35 to 70%, depending on the author. Most often affecting the pelvis, they occur within two years of diagnosis.

Treatment is essentially surgical, and must be complete from the outset [24, 25]. The first stage of the operation involves peritoneal cytology and exploration of the abdomen. Most authors perform a non-conservative hysterectomy, although some have shown that ovarian conservation does not affect survival. Complementary procedures depend on the degree of exploration : visceral excision depending on extension, and pelvic curage if adenopathies are palpated.

Radiotherapy reduces the incidence of pelvic recurrence, but does not improve overall survival [24, 26, 27]: its place should be discussed on a case-by-case basis. Post-operatively, radiotherapy may be proposed when exeresis is incomplete due to disease extension, and in the absence of surgical revision. When surgery is contraindicated or refused by the patient, exclusive radiotherapy may also be discussed.

Chemotherapy may be offered before surgery if the tumour is deemed unresectable from the outset, or after surgery if tumour resection was not optimal, or in the case of distant metastases. The main protocols include cis-platinum, adriamycin and ifosfamide, and more recently gemcitabine and docetaxel [26]. When a leiomyosarcoma is discovered histologically on a hysterectomy specimen, no adjuvant treatment is required, and there is no need to reintervene for lymph node staging, as leiomyosarcomas are not very lymphophilic. However, it is preferable to carry out an extension work-up to ensure that there are no pulmonary or abdominal metastases (chest X-ray and abdomino-pelvic scanner). Depending on the histological risk of uterine invasion, postoperative radiotherapy may be proposed.

In stage 1, the tumor is located only in the uterus. Stage 1 is divided into stages 1A and 1B. In stage 1A, the tumor measures 5 centimeters or less. In stage 1B, the tumor measures more than 5 centimeters.

In our patient, leiomyosarcoma presented as a giant fibroma delivered through the cervix prolapsed out of the vulva with acute uterine inversion neglected. Total vaginal hysterectomy is the technique of choice and represents the radical treatment given that the extension workup concludes stage 1.

CONCLUSION

Acute non-puerperal uterine inversion is a rare gynecological complication. Leiomyosarcoma is a possible cause, but it is a rare cancer with a poor prognosis. It is rarely diagnosed preoperatively, most often presenting as a benign myoma or necrobiosis. The main prognostic factor is mitotic activity. Treatment is dominated by surgery. Radiotherapy can only reduce local recurrence without altering survival, and chemotherapy has not proved effective.

When the tumor is confined to the uterus (stage 1), as described in our case and presenting as a giant fibroid delivered through the cervix prolapsed out of the vulva and causing uterine inversion, treatment consists of total vaginal hysterectomy.

REFERENCES

1. Salazar, O. M., Bonfiglio, T. A., Patten, S. F., Keller, B. E., Feldstein, M., Dunne, M. E., & Rudolph, J. (1978). Uterine sarcomas. Natural history, treatment and prognosis. *Cancer*, *42*(3), 1152-1160.
2. Livi, L., Paiar, F., Shah, N., Blake, P., Villanucci, A., Amunni, G., ... & Harmer, C. (2003). Uterine sarcoma: twenty-seven years of experience. *International Journal of Radiation Oncology* Biology* Physics*, *57*(5), 1366-1373.
3. Acharya, S., Hensley, M. L., Montag, A. C., & Fleming, G. F. (2005). Rare uterine cancers. *The lancet oncology*, *6*(12), 961-971.
4. Livi, L., Andreopoulou, E., Shah, N., Paiar, F., Blake, P., Judson, I., & Harmer, C. (2004). Treatment of uterine sarcoma at the Royal Marsden Hospital from 1974 to 1998. *Clinical Oncology*, *16*(4), 261-268.
5. Beringer, R. M., & Patteril, M. (2004). Puerperal uterine inversion and shock. *British Journal of Anaesthesia*, *92*(3), 439-441.
6. Lupovitch, A., England, E. R., & Chen, R. (2005). Non-puerperal uterine inversion in association with uterine sarcoma: case report in a 26-year-old and review of the literature. *Gynecologic oncology*, *97*(3), 938-941.
7. Kouamé, A., Koffi, S. V., Adjoby, R., Diomandé, F. A., Effoh, D., Oussou, C., & Kouakou, F. (2015). Non-puerperal uterine inversion in a young woman: a case report. *Journal of the West African College of Surgeons*, *5*(3), 78-83.
8. Song, Y. J., Yang, J., Yun, H. S., Lee, S. K., Kim, H. G., Lee, D. H., ... & Na, Y. J. (2016). Non-puerperal uterine inversion presented with hypovolemic shock. *Journal of Menopausal Medicine*, *22*(3), 184-187.
9. Rathod, S., Samal, S. K., Pallavee, P., & Ghose, S. (2014). Nonpuerperal uterine inversion in a young woman : a case report. *J Clin Diagn Res*, *8*(9), OD01-2.
10. Casanova, J., Huang, K. G., Adlan, A. S., & Artazcoz, S. (2014). Uterine inversion caused by submucosal leiomyoma. *J Gynecol Surg*, *29*(6), 294-296.
11. Lupovitch, A., England, E. R., & Chen, R. (2005). Non-puerperal uterine inversion in association with uterine sarcoma: case report in a 26-year-old and review of the literature. *Gynecologic oncology*, *97*(3), 938-941.

12. Takano, K., Ichikawa, Y., Tsunoda, H., & Nishida, M. (2001). Uterine inversion caused by uterine sarcoma: a case report. *Japanese Journal of Clinical Oncology*, *31*(1), 39-42.
13. Moulding, F., & Hawnaur, J. M. (2004). MRI of non-puerperal uterine inversion due to endometrial carcinoma. *Clinical radiology*, *59*(6), 534-537.
14. Zhang, X., Sun, L., Chen, X., & Hua, K. (2015). Uterus preserving reposition of non-puerperal uterine inversion under laparoscope: a case report and literature review. *Gynecologic and obstetric investigation*, *79*(3), 206-209.
15. Aarts, J. W., Nieboer, T. E., Johnson, N., Tavender, E., Garry, R., Mol, B. W. J., & Kluivers, K. B. (2015). Surgical approach to hysterectomy for benign gynaecological disease. *Cochrane database of systematic reviews*, (8), CD003677.
16. Tuckett, J. D., Yeung, A., Timmons, G., & Hughes, T. (2010). Non-puerperal uterine inversion secondary to uterine sarcoma and ascites demonstrated on CT and MRI. *European Journal of Radiology Extra*, *75*(3), e119-e123.
17. Fofie, C. O., & Baffoe, P. (2010). Non-puerperal uterine inversion: a case report. *Ghana medical journal*, *44*(2), 97-99.
18. Pelissier-Komorek, A., Lucereau-Barbier, M., Diab, J., Gavillon, N., & Graesslin, O. (2013). Acute non-puerperal uterine inversion the third degree. *Gynecologie, Obstetrique & Fertilite*, *41*(2), 130-132.
19. Auber, M., Darwish, B., Lefebure, A., Ness, J., & Roman, H. (2011). Management of nonpuerperal uterine inversion using a combined laparoscopic and vaginal approach. *American journal of obstetrics and gynecology*, *204*(6), e7-e9.
20. Van Dinh, T., & Woodruff, J. D. (1982). Leiomyosarcoma of the uterus. *American Journal of Obstetrics and Gynecology*, *144*(7), 817-823.
21. Schwartz, L. B., Diamond, M. P., & Schwartz, P. E. (1993). Leiomyosarcomas: clinical presentation. *American journal of obstetrics and gynecology*, *168*(1), 180-183.
22. Leibsohn, S., d'Ablaing, G., Mishell Jr, D. R., & Schlaerth, J. B. (1990). Leiomyosarcoma in a series of hysterectomies performed for presumed uterine leiomyomas. *American journal of obstetrics and gynecology*, *162*(4), 968-976.
23. Nordal, R. R., & Thoresen, S. Ø. (1997). Uterine sarcomas in Norway 1956–1992: incidence, survival and mortality. *European Journal of Cancer*, *33*(6), 907-911.
24. Socolov, R. V., Pricop, F. Z., & Stratan, D. (1992). The malignancy of uterine sarcoma compared with suspected leiomyomas : an anatomo-clinical study of 16 cases. *Rev Fr Gynecol Obstet*, *87*(10), 457-460.
25. Gerst, P. H., Levy, J., Swaminathan, K., Kshettry, V., & Albu, E. (1993). Metastatic leiomyosarcoma of the uterus: unusual presentation of a case with late endobronchial and small bowel metastases. *Gynecologic oncology*, *49*(2), 271-275.
26. Echt, G., Jepson, J., Steel, J., Langholz, B., Luxton, G., Hernandez, W., ... & Petrovich, Z. (1990). Treatment of uterine sarcomas. *Cancer*, *66*(1), 35-39.
27. Major, F. J., Blessing, J. A., Silverberg, S. G., Morrow, C. P., Creasman, W. T., Currie, J. L., ... & Brady, M. F. (1993). Prognostic factors in early-stage uterine sarcoma: a Gynecologic Oncology Group Study. *Cancer*, *71*(S4), 1702-1709.