Nonpuerperal uterine inversion caused by an adenosarcoma: A case report

Inversion utérine non puerpérale causée par un adénosarcome: Cas clinique

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Abstract

Introduction: Eighty-five per cent of uterine inversions are puerperal. Non-puerperal uterine inversion is usually caused by tumours that exert a traction force on the fundus of the uterus. This causes the uterus to be partially or completely inverted. It is commonly related to benign tumours like submucosal leiomyomas. Nevertheless, malignancies are an infrequent association.

Case presentation: We report a case of a 35-year-old female patient, medically and surgically free, gravida0 para0, complaining of menometrorrhagia associated with pelvic pain for 2 years. A suprapubic ultrasound scan showed an enlarged, globular uterus with a heterogeneous, undefined mass of 49 mm in size. MRI scan showed the appearance of a U-shaped uterine cavity and a thickened inverted uterine fundus with an endometrial infiltrating mass of 25 mm. Intraoperative exploration showed uterine inversion involving the ovaries; the fallopian tubes and the round ligaments and a necrotic intracavitary mass. The malignancy of the tumor was confirmed through anatomopathological examination as Adenosarcoma.

Conclusions: Uterine inversion is rare outside the puerperal period, and malignant etiology must not be overlooked. Therefore, comprehensive care with meticulous etiological investigation is crucial.

Key words: Uterine Inversion, Nonpuerperal ,Adenosarcoma ,Case report.

Résumé

Arrière-plan: Quatre-vingt-cinq pour cent des inversions utérines sont puerpérales. L'inversion utérine non puerpérale est généralement précipitée par des tumeurs exerçant une force de traction sur le fond de l'utérus, provoquant le retournement partiel ou complet de l'utérus. Elle est le plus souvent associée à des tumeurs bénignes comme les léiomyomes sous-muqueux. Cependant, les tumeurs malignes sont une association rare.

Présentation du cas: Nous rapportons le cas de Mme MS âgée de 35 ans, sans antécédents pathologiques notables, G0P0, qui consultait pour une ménométrorragie associée à des douleurs pelviennes depuis 2 ans. Une échographie sus-pubienne a montré un utérus globuleux avec une masse mal limitée hétérogène de 49 mm. A l'IRM pelvienne, le corps utérin était prolabé réalisant l'aspect en « doigt de gant » et le siège d'un épaississement endométrial solido-kystique de 25 mm.

L'exploration per-opératoire a montré une inversion utérine emportant les deux annexes et les ligaments ronds et un processus intra cavitaire nécrosé friable. L'examen anatomo-pathologique a confirmé la malignité : Adénosarcome.

Conclusions: L'inversion utérine se produit rarement en dehors de la période puerpérale. La crainte est de passer à côté d'une étiologie maligne. D'où l'importance d'une prise en charge globale avec une enquête étiologique minutieuse.

Mot clés: Inversion utérine, non puerpérale, adénosarcome, rapport de cas

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Introduction

Eighty-five percent of uterine inversions are puerperal. Nonpuerperal uterine inversion is typically caused by tumors that pull on the fundus of the uterus, resulting in partial or complete inversion. Benign tumors such as submucosal leiomyomas are most commonly associated with this condition. Although rare, malignancies may also be associated with uterine inversion. The diagnosis of uterine inversion can be difficult on the basis of clinical examination alone. Imaging may be required to confirm and elucidate the diagnosis.

Observation

Mrs. MS 35 years old, medically and surgically free, gravida0 para0, complaining of menometrorrhagia associated with pelvic pain for 2 years. On examination, the patient was hemodynamically stable, afebrile, with pale conjunctiva and an enlarged uterus on abdominal palpation. A gynaecological examination revealed an intact hymen.

Due to the intact hymen, transvaginal ultrasound was not possible. A suprapubic ultrasound showed an enlarged globular uterus with a heterogeneous undefined mass of 49 mm. A malignant tumour was suspected, and a pelvic magnetic resonance imaging (MRI) scan was performed.

It showed the appearance of a U-shaped uterine cavity and a thickened, inverted uterine fundus on a sagittal image and a "bull's-eye" configuration on an axial image. The uterus was the site of an endometrial infiltrating mass of 25mm, with a high T2 signal, hyperintense on diffusion-weighted sequence with a low apparent diffusion coefficient (ADC), heterogeneously enhanced after gadolinium injection. A few spots of spontaneous high T1 signal were present related to hemorrhagic areas. There was no intraperitoneal effusion. The conclusion was a uterine inversion on an infiltrating endometrial mass. (Figure 1+2)

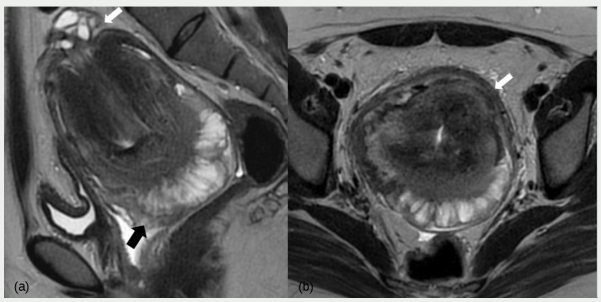

Figure1. T2 weighted MRI images

a: sagittal T2 weighted image showing inverted uterus fundus, with « U shape » uterus appearance , an endometrial infiltrating masse, poorly defined, with T2 hyperintense signal (black arrow), and an ovary above de the cervix (white arrow) .

b: axial T2 weighted image showing a « bull's eye » configuration of the middle part of uterus, with from the center outwards the uterine inverted corpus, the cervix and the invaginated round ligaments (white arrow).

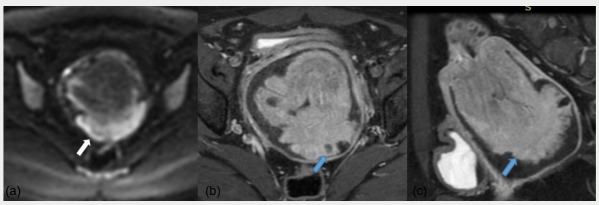

Figure2. Axial Diffusion weighted image (a), axial (b) and sagittal (c) fat-suppressed TSE T1 weighted images after gadolinium showing infiltrating diffusion hyperintense endometrial mass (white arrow), heterogeneously enhanced after gadolinium injection (blue arrow).

Exploratory laparotomy revealed uterine inversion involving the ovaries; the fallopian tubes and the round ligaments (Figure3).

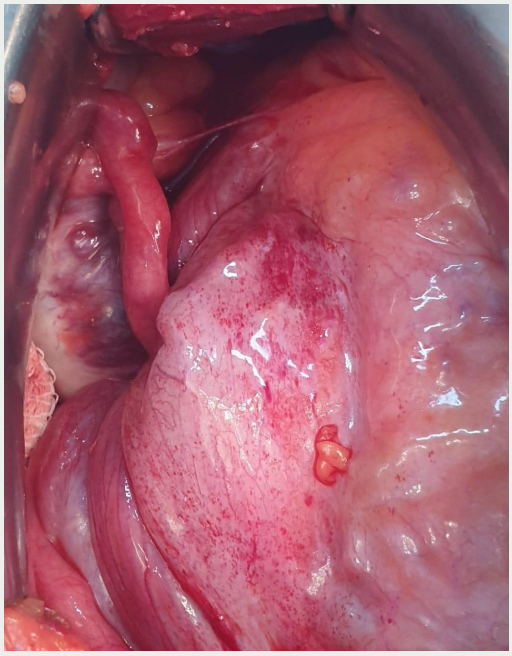

Figure 3. intraoperative exploration showing uterine inversion involving the ovaries; the fallopian tubes and the round ligaments.

A vertical hysterotomy of the posterior surface of the uterus allowed excision of a friable necrotic intracavitary mass. This was completed by surgical release of the constriction ring and manual reduction of the uterine inversion and then hysterorrhaphy.

The surgical specimen was sent for frozen section anatomo-pathological examination showing undifferentiated tumor proliferation. The anatomo-pathological examination confirmed the malignancy: Adenosarcoma.

The patient recovered well from the operation without complications and was referred to an oncologist for further treatment.

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Discussion

Nonpuerperal uterine inversion is an extremely rare complication. These are sporadic cases.

The clinical presentation of non-puerperal uterine inversion includes a feeling of vaginal fullness; pelvic pain associated or not with menometrorrhagia.

Other signs may be associated such as vaginal discharge and urinary signs such as urinary burning, pollakiuria, dysuria or even urinary retention in complete inversion.
Several degrees are described depending on the location of the uterine fundus: (1)
- 1st degree: The fundus of the uterus is located in the endometrial cavity.
- 2nd degree: The fundus of the uterus exceeds the os of the cervix.
- 3rd degree: The fundus of the uterus exceeds the opening of the vagina.
- 4th degree: The uterus and vagina are upside down.

The diagnosis can be difficult in the incomplete forms (1st and 2nd degree) where the diagnosis is generally made by imaging.
On the other hand, the diagnosis is easy in the complete forms (3rd and 4th degree) by the visualization of a mass exteriorized by the vagina generally bleeding on contact with absence of the uterine fundus on vaginal examination associated with abdominal palpation.
If the tubal ostium is seen, it is conclusive to uterine inversion. But if the mass is infected, the ostium may not be easily visible. Ultrasonography is the first line examination, due to its simplicity and accessibility. It can help both the diagnosis of nonpuerperal uterine inversion and the diagnosis of its etiology.
The Ultrasonographic sign in uterine inversion is the « target sign » in transversal section, with the hyperechoic fundus surrounded by a rim of fluid within the space between the inverted fundus and the vaginal or cervical wall (2). In longitudinal plane, we observe the uterus deformed into a "Y" shape, with depression of the uterine fundus in incomplete (or partial) inversion (3), and a deformation in "U" shape in complete inversion (4). Ultrasound can also show if there is a uterine mass with its characteristics (limits, echostructure, and vascularization with color Doppler...). However, it has its limitations, it is indeed an operator-dependent technique. In addition, the presence of a large or infiltrating uterine mass may make analysis of uterine morphology and structure difficult.
MRI is the best imaging modality to diagnose uterine inversion. It allows not only the diagnostic confirmation and the type of inversion, but it also allows to characterize the underlying mass. MRI shows as our presented case a U-shaped uterine cavity on the sagittal and coronal images. This appearance is due to invagination of the uterine fundus into the cervix or vagina, with loss of the normal convex appearance of the top of uterus. On axial images, a « bull's eye » sign is observed (5). Some authors suggested that the best sign to confirm uterine inversion on MRI is to visualize the round ligaments bulging centrally out the top of the uterus. MRI also characterizes the causal mass, and studies with high performance the adjacent structures providing an exhaustif locoregional extension assessment in case of malignancy.
Submucosal myoma is the etiology found in 70 to 85% of cases according to the authors (6). In 15 to 30% of cases, malignant tumours are involved, particularly sarcomas. (6). The incidence of uterine sarcomas represents 3 to 7% of all malignant tumors of the uterus, and it occurs mainly in women over 50 years old (7).Uterine sarcomas include leiomyosarcoma , endometrial stromal sarcoma , adenosarcoma , fibrosarcoma, and undifferentiated uterine sarcoma (8). Uterine adenosarcoma is the rarest subtype, accounting for only 5% of all uterine sarcomas (9) and occur in women of all ages (15 to 90), with an average age of 58(10). Sarcomas give as symptoms vaginal bleeding, lower abdominal pain, abdominal distention, and may rarely be complicated by uterine inversion.
The management of early-stage uterine sarcomas consists of total abdominal hysterectomy and bilateral salpingo -oophorectomy. Since the risk of lymph node and omental metastases is negligible, the morbidity of bilateral pelvic lymphadenectomy and omentectomy is best avoided. Additionally, as adenosarcomas are indolent and low-grade malignancies, cytotoxic chemotherapy is unlikely to be beneficial and there are no studies supporting the role of radiotherapy (8).

Conclusion

Uterine inversion is a rare occurrence outside of the postpartum period. A comprehensive examination with careful etiological investigation is of utmost importance to avoid overlooking a malignant cause.

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