

Benign Cystic Mesothelioma of the Uterus: Myth or Reality?

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Abstract

Case Report

Mesothelioma is a very rare tumor of mesodermal origin that covers the surface of body organs such as the peritoneum, pleura and pericardium. This tumour in the majority of cases presents in its malignant form where it represents some characteristics: increased cellularity, cytological atypia, papillary formations, even invasion. We report the case of Mrs AB, 37 years old, blood group A positive with no particular pathological history, nulli gesture nulli pare, consulted for pelvic pain and abdominal bloating. The clinical examination showed a distended bloated abdomen with a 20 centimetre pelvic mass with regular contours and painlessness. Abdominal and pelvic ultrasound showed a globular uterus increased in size by 200 millimetres with a multilocular cystic image intra myometrial (corporal anterior and fundial) of 188*79 millimetres. Intraoperatively, a globular uterus was found to be enlarged with a 15 centimetre parovascular cystic mass, dependent on the anterior aspect of the uterus, with a rock water fluid content, without any other associated abnormalities. The postoperative course was simple. To conclude, Benign cystic mesothelioma of the uterus is a tumour of diagnostic value. This tumour is spleen. The preoperative evaluation allows the diagnosis to be suspected, especially by pelvic magnetic resonance imaging.

Keywords: Mesothelioma, pelvic mass, benign cystic mesothelioma, uterine mass, ultrasound, magnetic resonance imaging.

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INTRODUCTION

Mesothelioma is a very rare tumor of mesodermal origin that covers the surface of body organs such as the peritoneum, pleura and pericardium.

This tumour in the majority of cases presents in its malignant form where it represents some characteristics: increased cellularity, cytological atypia, papillary formations, even invasion. In contrast, benign mesothelioma is much more rare.

Benign mesothelioma often has no exposure to asbestos and may present as a pain or lump. Benign mesothelioma is a relatively rare tumour of the female genital tract. It is most often asymptomatic and discovered incidentally.

This tumour is most commonly found in the fallopian tube and uterus, and more rarely in the ovary and para-ovarian tissue. It is also known to be the most common benign tumour of the tube [1].

Generally, benign mesothelioma appears as a small, single, solid nodule and its frequency is underestimated because it is often mistaken for a leiomyoma or adenomyoma [2].

The diagnosis is often made by histological discovery. The cystic form of benign uterine mesothelioma is much rarer and was first described in 1987 [3].

Based on our observation and a review of the literature, we will discuss (the clinic, histo pathological aspects, treatment and prognosis) of benign cystic mesothelioma of the uterus.

CASE REPORT

Mrs AB, 37 years old, blood group A positive with no particular pathological history, nulli gesture nulli pare, consulted for pelvic pain and abdominal bloating.

Pelvic pain and abdominal bloating of rapidly progressive onset and worsening 2 months ago.

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The clinical examination showed a distended bloated abdomen with a 20 centimetre pelvic mass with regular contours and painlessness. Abdominal and pelvic ultrasound showed a globular uterus increased in size by 200 millimetres with a multilocular cystic image intra myometrial (corporal anterior and fundial) of 188*79 millimetres.

Two ovaries in follicular view.
No hepatic, biliary, pancreatic or urinary anomaly (figure 1).

In front of this cystic mass, several differential diagnoses can be evoked: the most frequent being ovarian cyst and other diagnoses must also be evoked such as cystic degeneration of a uterine fibroid uterine fibroid, adenomyoma, congenital uterine cyst Naboth's egg or a hydatid cyst of the uterus.

An abdomino-pelvic CT scan was performed showing the presence of rounded pelvic cystic formations, one opposite the uterine fundus measuring 150*50 millimetres, the second is of corporal intra-mural seat measuring 28*28 millimetres, the ovarian origin of the two cystic images is probable.

A pelvic magnetic resonance imaging was carried out showing an enlarged uterus with a pure cystic formation with hypo T1 signal, hyper T2 signal without enhancement after injection of gadolinium, with an intra

mural seat in contact with the endometrium without deforming it. It measures 150*56*53 millimetres and is composed of thin regular partitions with a T1 and T2 hypo signal which are homogeneously enhanced and identical to that of the adjacent myometrium after injection of gadolinium, without any tissue nodule being visualised. This cystic mass with intra-myometrial partitions is suggestive of a benign cystic mesothelioma of the uterus, with the two ovaries in place of normal size and follicular.

In view of this persistent pelvic cystic mass, surgical exploration was indicated.

Intraoperatively, a globular uterus was found to be enlarged with a 15 centimetre parovascular cystic mass, dependent on the anterior aspect of the uterus, with a rock water fluid content, without any other associated abnormalities. The procedure performed was a resection of the cystic mass.

Histological analysis showed smooth muscle tissue bordered by a bed of endotheliiform cells reminiscent of mesothelioma. On Immunohistochemistry, the lining cells show intense staining with calretinin and cytokeratin, allowing the diagnosis of benign cystic mesothelioma of the uterus or an intra intra myometrial mesothelial cyst.

The postoperative course was simple.

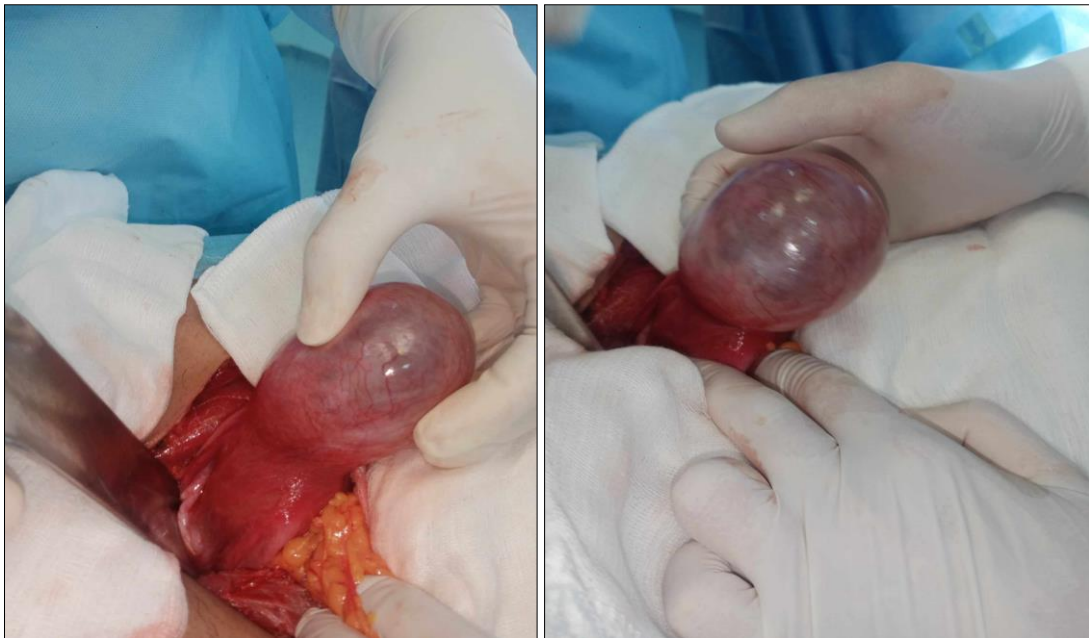


Figure 1: Aspect macroscopique de la masse utérine

DISCUSSION

Mesotheliomas are very rare neoplasms, representing a proliferative neoplasm composed of epithelial and mesenchymal cells, which converge on various organ surfaces in the body.

Mesotheliomas can be divided into benign and malignant types, and the benign ones are much rarer [4]. The distinction between benign and malignant mesothelioma is always histological [5].

Cytological atypia, high cell density, significant mitotic activity, invasion of the underlying stroma and necrosis were always associated with malignant mesothelioma. Immunohistochemical analysis is also increasingly useful in guiding the diagnosis.

The molecules associated with the malignant type are: (EMA, p53, glut-1 and IMP-3), while the molecules associated with the benign type are: (Desmin, Calretinin, and D2-40) [6-7].

A review of the literature has been carried out and only 14 cases of benign mesothelioma have been reported so far [8-12].

Benign mesothelioma of the uterus was first described in 1942 [13]. The old name of adenomatoid tumour was due to the presence of pseudoglandular structures suggestive of an adenoma [14].

The incidence of benign mesothelioma of the uterus in hysterectomy traps is 0.37% in the series by Huang *et al.*, [15], and 1.2% according to Mainguené [16]. The average age of onset is 41 years, with extremes ranging from 20 to 85 years [15].

In our case the patient was 37 years old. Most benign mesotheliomas of the uterus are subserous or located in the outer layers of the uterus. The posterior aspect of the uterus, especially near the horns or fundus, is a preferred location [17].

In our case, the mesothelioma had a non-classical location: subserosal, on the anterior surface of the uterus. Benign mesothelioma of the uterus most often presents as a single nodule. The macroscopic appearance is similar to that of leiomyoma. It is a firm, well-circumscribed mass, but less distinct than a leiomyoma, white yellow in colour on section, more or less fasciculated. The average diameter is close to 2 cm [17].

Several histological forms have been found. Quigley and Hart [18], described four forms: adenoid, angiomatoid (the most frequent), solid and cystic. A combination of two or more components is possible.

The cystic form of benign uterine mesothelioma is the most common [3-20], and is localised subserosally or intramurally. Two of the six cases of cystic mesothelioma of the uterus reported in the literature have multilocular tumours with a solid component [3-19].

The course of benign uterine mesothelioma is always favourable, with no risk of recurrence or malignant transformation. However, it can progress to a progressive increase in size with tumours exceeding 10 centimetres. In our case the tumour measures 15 centimetres in length.

In the rare cases where the diagnosis of benign mesothelioma is made pre-operatively, limited surgical excision is indicated.

Hysterectomy may be performed because of other associated uterine lesions (adenomyosis and/or leiomyoma) [14-17]. In our case, resection of the mass seems to be sufficient due to the absence of risk of recurrence and malignant transformation.

CONCLUSION

Benign cystic mesothelioma of the uterus is a tumour of diagnostic value. This tumour is spleen. The preoperative evaluation allows the diagnosis to be suspected, especially by pelvic magnetic resonance imaging.

Its mesothelial origin is now well established.

The evolution of benign uterine mesothelioma is always favourable, with no risk of recurrence or malignant transformation. The gold standard for the treatment of benign cystic mesothelioma of the uterus is limited surgical resection.

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