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Uterine Smooth Muscle Tumors with Uncertain Malignant Potential (STUMP): About a Case.

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Abstract

Introduction and Importance: Smooth muscle tumors of uncertain malignant potential (STUMP) are smooth muscle tumors whose morphological characteristics do not allow them to be formally classified as benign or malignant. This differentiation is based on histological criteria. Some tumors may present unusual anatomo-pathological aspects, leading to problems of differential diagnosis, notably with leiomyosarcoma.

Case Presentation: We report here the case of a 45-year-old Moroccan woman with no particular pathological history who consulted us for pelvic pain associated with abdominal enlargement, the anatomopathological result of which was in favor of a smooth muscle tumor of uncertain malignancy potential STUMP, its surgical management and evolution, in the gynecological-mammary surgery department of CHU Ibn Rochd in Casablanca.

Clinical Discussion: STUMPs are very rare tumours; their incidence is not well documented, varying around 0.01% of all women undergoing gynaecological surgery for a presumed preoperative diagnosis of uterine myoma. Leiomyosarcomas are defined by the presence of spindle-shaped cells with moderate to severe nuclear atypia, more than ten mitoses in ten fields at 40 X magnification, presence of tumour necrosis. Two of these criteria are necessary for a diagnosis of malignancy. We speak of STUMP when one of the malignancy criteria is present and the second is difficult to assess. The clinical presentation of STUMP does not differ from that of other MSDs such as leiomyoma or leiomyosarcoma. There are no precise data on the metastatic nature of PUMTS. The therapeutic approach differs from one school to another, ranging from simple myomectomy to total hysterectomy or even bilateral adnexectomy. The prognosis for STUMP is better than for leiomyosarcoma. Given the uncertain nature and evolution of these tumours, patients should receive close, long-term clinical and radiological follow-up.

Conclusion: STUMPs are rare tumours, usually of unexpected histological diagnosis after gynaecological surgery for preoperative diagnosis of uterine myoma. The final diagnosis is histological. Their therapeutic management poses a particular problem when the woman wishes to preserve her fertility. The prognosis for STUMPs remains intermediate between that of leiomyosarcomas and leiomyomas.

Keywords: Smooth Muscle Tumors, Uncertain Malignancy Potential, Histological Diagnosis, Gynecological Surgery, Case Report.

Abbreviations: STUMP: Smooth Muscle Tumors of Uncertain Malignant Potential, IHC: Immunohistochemistry

Introduction

Uterine smooth muscle tumors are divided into two categories: benign (leiomyoma) and malignant (leiomyosarcoma). This differentiation is based on histological criteria such as the presence of tumour cell necrosis, cytological atypia and tumour cell mitotic activity **[1]**. Certain tumors may present unusual anatomo-pathological aspects, leading to problems of differential diagnosis, notably with leiomyosarcoma. In 2003, the WHO classified these tumors as smooth muscle tumors of uncertain malignant potential (STUMP). We report the case of a 45-year-old female patient with no previous pathological history, who presented with pelvic pain associated with an increase in abdominal volume. The anatomopathological result was in favour of a STUMP smooth muscle tumour of uncertain malignancy potential, in the gynaecological-mammary surgery department of the Ibn Rochd University Hospital in Casablanca. We ensure that the work was reported in accordance with SCARE 2020 criteria **[2]**.

Patient and observant

Patient information: a 45-year-old Moroccan woman with no particular pathological history, single and nulligravida, always genitally active with regular cycles. The patient was referred to us from a peripheral hospital, accompanied by an ambulance driver, for chronic pelvic pain associated with an increase in abdominal volume evolving for 1 year without associated urinary or digestive signs.

Clinical examination findings: Clinical examination on admission revealed a conscious, normotensive, normocardic, eupneic and apyretic patient. Abdominal examination revealed a voluminous abdomino-pelvic mass extending to the umbilicus. A gynaecological examination (speculum + vaginal examination) was not performed, as the patient was a virgin. Rectal examination revealed a large abdomino-pelvic mass.

Diagnostic approach: Pelvic ultrasound revealed a voluminous left latero-uterine tissue mass rising to the level of the abdominal cavity, roughly oval, lobulated in outline, discreetly vascularized on color Doppler, measuring approximately 15x9 cm. The mass appeared to be pushing back the uterus, which remained normal in size, regular and homogeneous in outline, with both ovaries' unseen and no peritoneal effusion. An abdomino-pelvic CT scan was ordered, which revealed an intra-cavity tissue mass lateralized to the left (spur sign with the uterus), heterogeneously enhanced after injection of contrast medium, rounded, fairly well limited, with patches of necrosis, measuring 165x127x80mm. The mass displaced the uterus and bladder to the right. Blade of peritoneal effusion in FIG. The patient underwent a pre-operative work-up and a pre-anaesthetic visit and was scheduled for exploratory laparotomy after patient consent.

Surgical treatment: The patient was operated on two weeks after presentation to the department, by an experienced surgeon. Under general anaesthesia, after betadine scrubbing, bladder catheterization and placement of the operating field, the incision used was median subumbilical, given the size of the mass on exploration: 16 cm subserous myoma. The two adnexa and the rest of the abdomino-pelvic cavity were unremarkable. The patient underwent myomectomy.

Final diagnosis: The pathological findings were as follows: a nodular formation weighing 786 grams and measuring 15 x 9 x 7.5 cm. On section, it was firm in consistency, with a fasciculated white appearance and the presence of haemorrhagic remodelling. There were no necrotic changes or calcifications. Histological examination of the various specimens showed a fasciculated tumour proliferation, dissociated by hyaline fibrous remodelling. Smooth muscle cells with spindle-shaped nuclei display moderate to severe cytonuclear atypia. Cytoplasm is abundant and poorly limited. Mitotic count showed 9 mitoses/10 CFG. No foci of necrosis were seen. The histological appearance was suggestive of a smooth muscle tumor of uncertain malignant potential (STUMP).

Follow-up and evolution: Immediate postoperative follow-up was straightforward. The case was discussed at a multidisciplinary consultation meeting attended by

oncologists, radiotherapists and radiologists, and the decision was made to carry out clinical and ultrasound monitoring every 3 months. The patient underwent 4 unremarkable clinical and ultrasonographic examinations, with a TAP CT scan showing no secondary localizations.

Discussion

Morphologically, there is a spectrum of uterine smooth muscle tumours with predictable clinical outcomes and conventionally well-defined histological criteria; at either end of this spectrum are leiomyomas and leiomyosarcomas. In between are several variants with unusual histological features, clinical course and prognosis. The World Health Organization has called them smooth muscle tumors of uncertain malignancy potential **[3]**. Unlike uterine leiomyoma, the most common neoplasm of the uterus, STUMPs are very rare tumours; their incidence is not well documented, varying around 0.01% of all women undergoing gynaecological surgery for a presumed preoperative diagnosis of uterine myoma **[4]**. Histologically **[5]**, three criteria are used to classify smooth muscle tumours as benign or malignant. These criteria include nuclear atypia, mitosis index and the presence of tumour necrosis. Thus, leiomyosarcomas are defined by the presence of spindle-shaped cells with moderate to severe nuclear atypia, more than ten mitoses in ten fields at 40 X magnification, presence of tumor necrosis. Two of these criteria are necessary for a diagnosis of malignancy. The term STUMP is used when one of the malignancy criteria is present and the second is difficult to assess. The following cases are thus classified as STUMP: a spindle-cell smooth muscle tumor with moderate to severe nuclear atypia and a borderline mitotic index of between eight and nine mitoses, a spindle-cell smooth muscle tumor with moderate to severe nuclear atypia and necrosis whose tumoral or ischemic nature is difficult to assess, spindle cell smooth muscle tumor with more than ten mitoses and necrosis of a tumoral or ischemic nature that is difficult to assess true tumoral necrosis in a common leiomyoma **[6]**. In our case, the anatomopathological study revealed smooth muscle cells with spindle-shaped nuclei showing moderate to severe cytonuclear atypia, with an estimated mitotic index of 9 mitoses/10 fields at high magnification (CFG), classifying it as STUMP.

To improve the diagnosis of STUMP, in addition to morphology, there are also reports on the role of immunohistochemistry (IHC). Several markers, such as Ki-67, p16, p53, pHH3, Bcl-2, Caveolin-1 or the AT-rich 1 α interactive domain, have been tested **[7,8]**. There are some preliminary reports, but the series are still too small. Some of these markers may also be discussed as prognostic variables: Bcl-2 expression is more frequent in leiomyomas than in STUMP or leiomyosarcoma; thus, if Bcl-2 is expressed in STUMP or malignant tumors, it may have positive prognostic significance **[7,9,10]**. Interestingly, Caveolin-1 expression increases dramatically from benign conditions such as leiomyoma to STUMPs and, more significantly, in malignant tumors such as leiomyosarcoma **[11]**.

Suspecting the diagnosis clinically or radiologically remains difficult, as STUMPs have no specific features compared to benign leiomyoma. The clinical presentation of STUMP does not differ from other MSDs such as leiomyoma or leiomyosarcoma: pelvic pain, pelvic mass, abnormal uterine bleeding, compression of adjacent organs and secondary anemia, infertility, dysmenorrhea and abnormal vaginal discharge may all be present **[12,13]**, making symptoms non-specific in all these histologically and clinically different conditions. In our case, the main complaint that led to this patient's diagnosis was pelvic pain associated with a rapidly growing abdomino-pelvic mass, detected by ultrasound and abdomino-pelvic CT. In the search for metastases, the thoraco-abdomino-pelvic stage should be systematically investigated, as cases of metastases, notably lung metastases, following STUMP have been reported in the literature **[14,15]**. There are no precise data on the metastatic nature of STUMP. In our patient, to date, only one TAP CT scan has been performed, showing no secondary localization **[14]**. This uncertainty in the anatomical-pathological diagnosis frequently leads to therapeutic dilemmas, particularly when the diagnosis is made on the basis of myomectomy specimens from women wishing to preserve their fertility. Therapeutic approaches differ from one school to another, ranging from simple myomectomy to total hysterectomy or even bilateral adnexectomy. In our case, myomectomy was feasible.

The prognosis of STUMPs is better than that of leiomyosarcomas. Various studies have found a significantly lower recurrence rate than in leiomyosarcoma. We found no data in the literature for different recurrence rates, depending on the surgical approach; laparoscopic or open. Nevertheless, some data suggest that there may be an increased risk of relapse following tumor morcellation **[16,17]**. Overall survival at 5 years is estimated at 92-100% of patients **[5]**. In the event of recurrence, the prognosis remains relatively good, as surgery remains a curative option and there is only one case report with a lethal outcome **[18]**.

There have also been reports of metastasis, notably to the lungs, probably in conjunction with the preferential hematological spread of leiomyosarcomas **[19]**.

Given the uncertain nature and evolution of these tumours, patients should receive close, long-term clinical and radiological follow-up **[20,21]**. Our patient underwent a clinical examination and pelvic ultrasound every 3 months, and a TAP CT scan every year, which revealed no abnormalities.

Conclusion

STUMPs are rare tumours, usually of unexpected histological diagnosis after gynaecological surgery for preoperative diagnosis of uterine myoma. Preoperative clinical discrimination of leiomyoma or leiomyosarcoma is often impossible, and the final diagnosis is histological. STUMP has a certain potential for relapse, irrespective of the surgical procedure used, but the only procedure to avoid is morcellation, as there are data on its negative prognostic influence. The therapeutic management of STUMP poses a particular problem when the woman wishes to preserve her fertility. The prognosis of STUMPs remains intermediate between that of leiomyosarcomas and leiomyomas.

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Ethical Approval: The authors institute provided ethical approval for this case study.

Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request

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