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Uterine smooth muscle tumors of uncertain malignant potential (STUMP): management, follow up and prognosis

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Abstract

Smooth muscle tumor of uncertain malignant potential (STUMP) encompasses a broad group of uterine neoplasms that do not meet the current histologic criteria for a diagnosis of either benign or a malignant tumor. We made a retrospective review of patients´s folders with the histological diagnosis of STUMP in our department so that to describe the clinical and imagery features and assess the prognosis after surgery. We recorded 4 cases from 2016 to 2019. Patients were between 31 and 55 years old. The mean age of patients was 42.7 years and most of patients presented abdomino pelvic pain and uterine abnormal bleeding evolving at least more than 3 months. The clinical exam was not particular. Three patients benefited of pelvic ultrasound which described most of the time the aspect of uterine myoma. The MRI was performed for 3 patients also and showed an endometrial process infiltrating the myometrial wall for 2 patients. All of them underwent total hysterectomy and the histology described the aspect compatible with STUMP sometimes associated with uterine myomas or polyp. The first two years follow up did not find any recurrence and the prognostic seems to be good.

Introduction

Smooth muscle tumor of uncertain malignant potential (STUMP) encompasses a broad group of uterine neoplasms that do not meet the current histologic criteria for a diagnosis of either benign or a malignant tumor. It is thought that STUMP may represent a transition tumor between leiomyoma to leiomyosarcoma or possibly undiagnosed low-grade leiomyosarcoma [1]. It represent a poorly defined subcategory of uterine smooth muscle tumors (SMTs). Uterine SMTs, which have a broad spectrum ranging from leiomyosarcomas (LMSs) to leiomyomas (LMs), can be distinguished based on histopathological features including the degree of cytologic atypia, mitotic count activity (mitotic index per 10 high-power fields (MIs/10 HPFs), and presence of tumor cell necrosis [2]. Among women undergoing hysterectomy or myomectomy for a presumed diagnosis of leiomyoma, 0.01% receive a diagnosis of STUMP [3]. The true prevalence of STUMP is difficult to determine due to the rarity of the disease and the inconsistency in diagnostic criteria [4]. Treatment approaches and follow-up of these tumors have been still controversial, particularly in the reproductive age patients with fertility desire, due to the non-aggressive behavior and prolonged overall survival (OS) rate comparing to leiomyosarcomas [5]. The goal of this study is to describe the management and to assess the prognosis of patients diagnosed with STUMP in our department

Methods

We made a retrospective review of patients´s folders with the histological diagnosis of STUMP in the department of gynecology and obstetrics II of the Hassan II teaching hospital of Fez in Morocco between 2016 and 2019. Pre-operative, post-operative and the follow-up datas were collected from folders and introduced in the excel 2013 software. Preoperative datas were: the age in year; complains of patients; the length of the symptomatology in month; the hormonal status, the obstetrical history; the clinical exam finding and the imagery; post-operative datas were represented by the histology and the follow-up was based on the research of recurrence or metastasis by clinical exams and Imagery.

Results

A total of 4 folders with the diagnosis of STUMP have been recorded and analyzed. Patients were between 31 and 55 years old with a mean age of 42.7 years. Patient's description, imagery and surgery procedure and the histology are resumed in Table 1, Table 2, Table 3. All the patients benefited of post-operative follow up for at least 24 months. The first two years follow up did not find any recurrence.

Discussion

STUMPs represent a group of rare and heterogeneous neoplasms from both a histological and a clinical point of view [6,7]. The incidence of STUMPs is not well known [7]. According to Picerno TM *et al*. 0.01% of leiomyoma are diagnosed as STUMP [8]. It also represents 1/3 of uterine sarcomas and 1.3% of uterine cancers [1]. Due to the rarity of these tumors, existing literature on the topic remains scarse and therefore consensus regarding diagnosis, malignant potential, treatment of choice and follow-up has not yet been reached [6,9]. There is no clinical specific symptomatology. STUMPs often presents with symptoms consistent with a benign leiomyoma. The symptomatology include a combination of abnormal uterine bleeding, pelvic mass, or symptoms due to secondary compression and anemia [10]. Pelvic pain and pelvic pressure sensation or combination of them are also described [7]. Sometimes the symptomatology can be represented by the recurrence of myoma as it was the case of one of our patients. The symptomatology of our patients was dominated by abdomino-pelvic pain associated with uterine abnormal bleeding. The risk factors and biological events that lead to STUMP remain poorly understood and thus subsequent clinical behavior difficult to predict [6]. However, we notice in our present study that most of the patients (3 on 4) did not experience any pregnancy at the time of the diagnosis. The age of onset of this disease is similar to that of leiomyoma or leiomyosarcoma, and little is known regarding the specific risk factors that predispose to a diagnosis of STUMP [11].

Due to the rarity of stumps, there is no demographic data to consolidate the hypothesis based on the age of the occurrence. But it has been reported by Guntupalli SR *et al*. [6] that the mean age is 45 years and most of patients is in premenopausal period. In a retrospective study of 6 patients made by Bacanakgil *et al*. [7], the mean age of the patients was 42 years old. Which is similar to what we noticed in our study. STUMPs are likely to occur most of the time in premenopausal period. Most of our patients was in premenopausal period with regular monthly bleeding and only one of them was in post-menopausal period. That was the case in the study made by Bacanakgil *et al*. [7] were they also found out of 6 patients only 1 in post-menopausal period. The clinical examination finding remains poor and doesn´t lead to a diagnosis of STUMPs. Preoperative STUMPs diagnosis with imaging modalities is not easy and there´s no specific image related to STUMPS. However, in front of any abdomino pelvic mass it´s mandatory to perform in first line the pelvic ultrasound. In case the result don´t lead to a possible diagnosis we perform a pelvic RMI which result is better than ultrasound. In a study made by Lara Hughes *et al*. [10], MRI has been used to differentiate between benign and malignant tumors of the uterus utilizing increased signal intensity. They concluded that little evidence exists to distinguish STUMP from leiomyoma [10]. According to some authors, RMI should be used in cases with a high suspicion of a sarcoma [12]. Bonneau *et al*. [13] reported that preoperative STUMP diagnosis or differentiation from leiomyoma or leiomyosarcoma with imaging modalities is not easy. In their study, they compared sonography and MRI findings of 85 leiomyoma and 23 malignant mesenchymal tumors (MMT) and STUMP cases, and reported that presence of single tumor, absence of acoustic shadowing and presence of free fluid are associated with MMT/STUMP. No standard protocols for the management of patients with suspected STUMP have as of yet been approved. Due to the paucity of literature available, a consensus regarding management has not been reached and clinicians are required to model further management and follow-up on limited, largely observational data. It has been suggested that the treatment of choice is a hysterectomy [10]. According to Shapiro A *et al*. [14]. In the event of STUMP diagnosis in myomectomy specimens, considering the proved possibility of recurrence, hysterectomy represents the gold standard for those women who have completed their childbearing and successful

pregnancies following fertility sparing surgery is possible however these patients should be adequately informed of the risk of recurrence and a strict follow-up program through clinical and imaging techniques is mandatory.

As the suspected diagnosis of our patients were uterine myoma, we performed for 3 of them inter adnexial total hysterectomy as they were in premenopausal period and total hysterectomy plus bilateral adnexectomy for the one in post-menopausal period. The final diagnosis were given by the histology. Histologically, three criteria can be used to classify smooth muscle tumours as benign or malignant tumours. These criteria include nuclear atypies, mitosis index and the presence of tumor cell necrosis [15]. The current criteria for the histopathologic classification of smooth muscle tumors are based on the Stanford Criteria, and they are diagnosed by an assessment for abundant mitosis (10 per 10 HPFs), cellular atypia and presence of areas of coagulative tumor cell necrosis [3,4]. Two of these criteria are necessary to withhold the diagnosis of malignancy. STUMP is evocated when one of the malignancy criteria is present and the second is difficult to assess. Thus, the following cases are grouped into the stump category: a smooth fusiform cell muscle tumour with moderate to severe nuclear atypies and a limit mitotic index between eight and nine mitoses, a smooth cell muscle tumour fusiforms with moderate to severe nuclear atypies and necrosis whose tumor or ischemic nature is difficult to assess, a smooth fusiform cell muscle tumor with more than ten mitosis and a necrosis whose tumor or ischemic nature is difficult to assess a true tumor necrosis in a common leiomyoma [16]. STUMPs do not fulfill the diagnosis of leiomyosarcoma. Uterine smooth muscle tumors that show some worrisome histologic features (i.e. necrosis, nuclear atypia, or mitoses), but do not meet all diagnostic criteria for leiomyosarcoma, fall into the category of atypical smooth muscle tumors (STUMP). This diagnosis, however, should be used sparingly and every effort should be made to classify a smooth muscle tumor into a specific category when possible [17]. It´s been reported that the standard follow-up post-hysterectomy was six monthly for 5 years followed by yearly follow-up for a further 5 years with symptom review at each appointment and yearly imaging by MRI for evidence of recurrence or metastasis [10]. Follow-up visits should consist of a history, general and pelvic examination, and imaging studies annually including chest radiography, pelvic ultrasound, MRI and/or PET-CT, to detect recurrences [18]. All our patients benefited of post-operative monitoring for 24 months until now by clinical exam each 3 months and imagery based on TAP - TDM each 6 months for the first year. And a six monthly clinical exam and a year TAP- TDM for the second year. For the time being, none of them showed recurrence or metastasis. The prognosis of STUMP is better than that of leiomyosarcomas. Reported recurrence rate for STUMP is 7-27% [1,18]. A significantly reduced recurrence rate compared to leiomyosarcoma as well as 5-year survival ranging from 92 to 100% of patients is noticed [16] and there is no difference between the rate of patient who underwent hysterectomy and those who underwent myomectomy [6].

Conclusion

STUMPs represent a broad group of uterine neoplasms that do not meet the current histologic criteria for a diagnosis of either benign or a malignant tumor. The preoperative diagnosis is still not possible. According to our observation the prognosis remains good. Further study with a large sample is needed to assess a real prognosis of STUMP which will allow the edition of guidelines concerning its management.

What is known about this topic

- *Smooth muscle tumor of uncertain malignant potential (STUMP) encompasses a broad group of uterine neoplasms that do not meet the current histologic criteria for a diagnosis of either benign or a malignant tumor;*
- *The age of onset of this disease is similar to that of leiomyoma or leiomyosarcoma, and*

little is known regarding the specific risk factors that predispose to a diagnosis of STUMP;
- *No standard protocols for the management of patients with suspected STUMP have as of yet been approved.*

What this study adds

- *We observed in our study that STUMP occurs between 31 and 55 years old and the mean age of patients was 42.7 years and most of patients presented abdomino pelvic pain and uterine abnormal bleeding evolving at least more than 3 months;*
- *The prognosis is good and there is no recurrence the first two years after surgery.*

Competing interests

The authors declare no competing interests.

Authors' contributions

All the authors have read and agreed to the final manuscript.

Tables

Table 1: patient's description

Table 2: imagery and surgical procedure

Table 3: histology

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Table 1: patients description				
	Patient 1	**Patient 2**	**Patient 3**	**Patient 4**
Age (year)	44	41	31	55
Complain	Abdominal pelvic pain and abnormal uterine bleeding	Intermenstrual abnormal uterine Bleeding	Abdominal pelvic pain, abnormal uterine bleeding and recurrence of myoma	Abdominal pelvic pain and abnormal uterine bleeding
Length of symptomatology	05 months	06 months	06 months	24 months
Hormonal status	Pre-menopausal	Pre-menopausal	Pre-menopausal	Post-menopausal
Obstetrical history (Number of pregnancy)	0	0	0	6
Clinical exam	Hypogastric mass	Abdominal pelvic mass	Pelvic unlimited mass	Pelvic and Cervical mass

Table 2: imagery and surgical procedure				
	Patient 1	**Patient 2**	**Patient 3**	**Patient 4**
Pelvic ultrasound	Uterine myoma	Uterine myoma	N/A	Intramural vascularized image of 6cm
Pelvic MRI	N/A	Polymyomatous uterus with large intramural lesion	endometrial tumor process invading the myometer	endometrial process infiltrating the myometer, cervix and parameters
Surgical procedure	Inter adnexial total hysterectomy	Inter adnexial total hysterectomy	Inter adnexial total hysterectomy	Total hysterectomy with bilateral adnexectomy

Table 3: histology				
	Patient 1	**Patient 2**	**Patient 3**	**Patient 4**
Macroscopy	Friable myometrial heterogeneous mass of 6x8x9cm, and 5cm myoma	Interstitial and sub mucous mass of 10 x 9.5x5 cm	Sub mucous mass of 11x5x5cm and 02 Masses under bladder of 11x7x5cm and 4.5x3cm of white and homogeneous	N/A
Microscopy	Oval-shaped epitheliod cells with moderate cytonuclear atypies to eosinophilic cytoplasm and mitotic count less than 10/10CFG	Elongated tumor cells with minimal to moderate focal atypies and mitotic index at 08/10 CFG and area of tumor necrosis	Surface ulcerated fuso-cell proliferation with minimal cellular atypies and presence of congestive beams and wide range of necrosis with a mitotic index at 04/10	Fuso-cellular tumor proliferation arranged in intertwined bundles. High cellularity with frank cyto-nuclear nuclear atypies showing cells with elongated oval nuclei, irregular contours and dense chromatin, with an eosinophilic cytoplasm. The mitotic count was 9 mitoses / 10 CFG. No area of tumor necrosis. Endometrial samples found an atypical complex hyperplasia area.
Final diagnosis	STUMP -leiomyoma	STUMP	STUMP - redesigned leiomyoma - polyp	STUMP endometrial complex hyperplasia