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COEXISTENCE OF UTERINE SMOOTH MUSCLE TUMORS OF UNCERTAIN MALIGNANT POTENTIAL (STUMP) AND SUBSEROSAL LEIOMYOMA: A RARE CAUSE OF POSTMENOPAUSAL BLEEDING

MALİGN POTANSİYELİ BELİRSİZ UTERUS DÜZ KAS TÜMÖRÜ (STUMP) VE SUBSEROZAL LEİOMYOMA BİRLİKTELİĞİ: POSTMENOPOZAL KANAMANIN NADİR BİR NEDENİ

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ÖZET

Malign Potansiyeli Belirsiz Uterus Düz Kas Tümörlerinin klinik davranışı ve risk faktörleri hakkındaki bilgiler son derece sınırlıdır. Malign Potansiyeli Belirsiz Uterus Düz Kas Tümörü olan hastaların çoğu üreme çağındadır. Postmenopozal kanama menopozdan sonra hastaların yaklaşık %10'unda ortaya çıkabilir. Uterin leiomyosarkomlar postmenopozal kanama nedeni olabilse de Malign Potansiyeli Belirsiz Uterus Düz Kas Tümörleri sık görülen bir postmenopozal kanama nedeni değildir. Biz bu raporda Malign Potansiyeli Belirsiz Uterus Düz Kas Tümörü ve multiple bening subserozal leiomyomlara sahip, postmenopozal kanama şikayetiyle başvuran 51 yaşında bir olgu sunulmuştur.

Anahtar Kelimeler: Leiomyom; Malign Potansiyeli Belirsiz Uterus Düz Kas Tümörleri; Postmenopozal.

ABSTRACT

Information about the clinical behaviour and risk factors of Uterine Smooth Muscle Tumors of Uncertain Malignant Potential (STUMPs) is extremely limited. Most of the patients with STUMP are at reproductive age. Postmenopausal bleeding (PMB) can occur about 10% of patients after menopause. An uterine leiomyosarcoma may be the cause of PMB in some cases but STUMPs are not a common cause of PMB. We reported here a 51-year-old woman presenting with PMB having STUMP and multiple bening subserosal leiomyomas.

Key Words: Leiomyoma; Postmenopausal; Uterine Smooth Muscle Tumors of Uncertain Malignant Potential.

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INTRODUCTION

Smooth Muscle Tumors (SMTs) constitute a group of histologic, genetic, and clinical heterogeneous tumorsother than leiomyoma (LM)that including mitotically active leiomyoma, cellular leiomyoma, atypical leiomyoma, uncertain malignant potential (STUMP), and leiomyosarcoma (LMS) (1). The most widespread classification system for uterine smooth muscle tumours is that proposed by Bell et al. (2). LMs are at the benign end of the of smooth muscle cell tumour spectrum, LMSs are located at the malign end. STUMPs are classified as tumours that do not meet the definition of any uterine smooth muscle tumour category. Information about the clinical behaviour and risk factors of STUMPs is extremely limited. The average age of the patients at the diagnosis was found about 43 (3). Most of the patients with STUMP are at reproductive age. Postmenopausal bleeding (PMB) can occur about 10% of patients after menopause (4). An uterine LMS may be the cause of postmenopausal bleeding (PMB) in some cases (5). On the other hand; STUMPs are not a common cause of PMB. We reported here a 51-year-old woman presenting with PMB having STUMP and multiple bening subserosal LMs.

CASE REPORT

A 51-year-old multiparous woman was referred to our clinic withPMB and uterine mass. She was in postmenopausal period for 7 years; and received medical treatment for diabetes mellitus and hypertension. She had undergone a right salpingo-oophorectomy due to an ectopic pregnancy 31 years ago. Family history contained no remarkable pathological features and no additional malignancywas found in the patient. Transvaginal sonography examination revealed an enlarged uterus and a 60 × 54 mm cystic masswith irregular borders and septations inside was observed in the myometrium; the endometrium could not be clearly evaluated.Multiple subserosal LMs were also detected during sonographic examination. The left ovary appeared normal. In the magnetic resonance imaging (MRI), the hyper intensity on T2 AG showed thick septations contained in the uterine fundus; in the minimal hypo intensity of the T1 AG view, a thick-walled 70 × 58 × 60 mm cystic mass lesion was observed. Multiple subserosal LMs were also seen in sagittal MR images (Figure 1).

No pathological findings were present in the endometrial biopsy results or the cervico-vaginal smear. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed. Diagnosis of STUMP case was made on the basis of the presence of diffuse

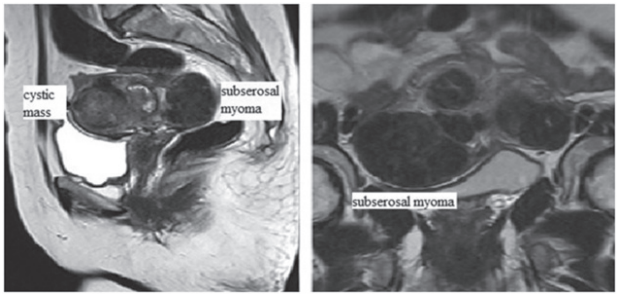

Figure 1 • The cystic mass and subserosal LMs in the MRI.

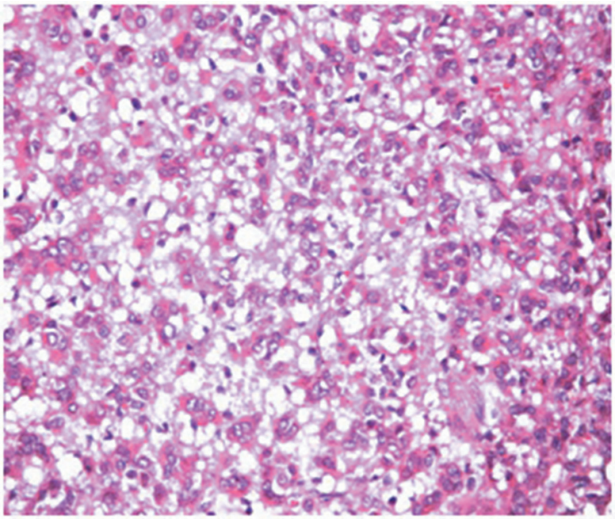

Figure 2 • Diffuse atypia, absence ofnecrosis and increased mitotic activity.

atypia and the absence ofnecrosis andincreasedmitotic activity observed as a result of the postoperative histopathological examination (Figure 2).

The postoperative follow-up was normal and the patient was discharged at second day. No pathological findings were detected in the physical examination and diagnostic imaging modalitiesat two months postoperatively.

DISCUSSION

There is little knowledge of the course of STUMPs and due to the lack of correlation between clinical results and pathological classification.The clinical management is still controversial. According to Bell classification, LMS exhibit at least two of the characteristic signs, which include widespread atypia, tumour cell necrosis and a mitotic count of 3-10 / 10 HPF under microscopy. LMs are defined as smooth-appearing tumours which are atypical, exhibit no tumour cell necrosis and have a mitotic count of ≤ 4 / 10 HPF. Leiomyoma variants

include atypical or simplasticleiomyomas, observed to exhibit mitotic activity and atypia and having mitoses fewer than 10/10 HPF without any cell necrosis. Tumours meeting some but not all of the criteria for malignancy are classified as STUMPs (2).

The women with STUMP are usually under 45 years old (3). Our patient was a 51 year old postmenopausal Caucasian women in this report. Also Guntupalli et al. did not find race/ethnicity to differ significantly between patients with STUMP who did and did not develop a recurrence. Recurrence of STUMP was seen nearly 7% of patients in that study (3). STUMP may recur as LMS indicates that STUMP, unlike mitotically active leiomyoma, should not be strictly considered a benign variant of LM (3). Our patient will be followed up in terms of recurrence or metastasis too.

Abnormalities of the endometrium are usually the reason of PMB. The frequency of the main causes of postmenopausal bleeding was found endometrial hyperplasia (22.3%), endometrial atrophy (21.3%), nondiagnostic (19.9%), endometrial carcinoma (9.5%), cervical carcinoma (6.8%), cervical polyps (4.5%), proliferative endometrium (3.2%), and endometrial sarcoma (3.5%) in a study (6). Similar to other studies, Wong SF et al. reported that pathologies found were similar in women with PMB to early reports, but incidences of submucous fibroids and endometrial polyps were found to be higher (13.4%) (7).Our patient was admitted to hospital with PMB. Transvaginal sonography examination and MRI revealed a enlarged uterus and cystic mass with irregular borders in the myometrium and the endometrium could not be clearly evaluated. Multiple subserosal LMs were also detected during sonographic examination. But diagnosis of STUMP could not be identified by imaging modalities in our patient.

Tanaka et al. claimed that no STUMPs had been identified exactly by imaging (9). They reported that STUMPs showed no hyperintense areas on either T1WI or T2WI, but resembled ordinary LMs (10). Immunohistochemical studies with P16, P21 and Ki 67 would be useful in distinguishing STUMP from other smooth muscle tumours and in demonstrating the malignancy potential of STUMP patients (9-10). Compared to STUMP, Ki 67 expression in LMS is significantly high (9). Atkins et al. reported that in which the type of necrosis is uncertain (coagulative tumor cell vs. hyalinized), the addition of p16 may aid in discerning a subset of STUMP that should be classified as LMS (11). In general, similar to our patient, the the precise histopathological diagnosis of STUMP is made at postoperative period.

Myomectomy, total abdominal hysterectomy and total abdominal hysterectomy salpingooophorectomy may be the primary surgical approaches for STUMP (3). No patient received chemotherapy or radiation therapy after the initial diagnosis of uterine STUMP in a large study (3). Clinical observation and an expectant approach are recommended. Recurrence rates were similar for women who underwent myomectomy and those who underwent hysterectomy (3). In cases of performed myomectomy, it is not clear at the time whether or not a hysterectomy would be necessary because the diagnosis is usually made postoperatively. The patient be informed and her fertility status be taken into consideration in the decision making.

Recurrence of the disease is more likely to occur in the same site and resection is possible (12, 13). Vural et al. reported that the recurrence rate was 15 % in patients with uterine STUMP and recurrences can be in the form of STUMP or LMS (13). Strongly immunoreactive for p16 and p53, supporting earlier observations that these markers may be helpful in the prediction of the behavior of STUMPs (9). Patients diagnosed with STUMPs should receive long-term surveillance (9).

Patients diagnosed with STUMP should be closely monitored and due to the recurrence potential, a gynaecological oncologist should be consulted. It should be kept in mind that STUMP can be a possible reason of PMB. Also STUMP may coexist with multiple beningsubserosal LMs. The limited number of cases, the uncertainty of clinical results and recurrence ratiosimpedes the determination of appropriate follow-up criteria and preparation of a follow-up treatment plan. So, there is a need for more extensive and comprehensive studies about STUMPs.

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