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Case Report

Benign in radiology bombshell in cytology

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ABSTRACT

Smooth muscle tumor of uncertain malignant potential is a surprise post-operative histopathological finding after myomectomy or hysterectomy. These are aligned between benign and malignant which shares but does not fulfil the complete diagnostic criteria of leiomyosarcoma. Preoperative diagnosis by clinical or radiological examination is uncertain and the diagnosis is only by histopathology. Among women undergoing myomectomy or hysterectomy for a presumed diagnosis of leiomyoma, 0.01% receive a diagnosis of STUMP. Once diagnosed as STUMP, the gold standard treatment option is surgical removal of uterus. As there are no strict accurate protocols for the management of STUMP, the treatment choices should be based on the age of the patient, status and desire to preserve fertility, location of the tumor, metastases, recurrences and pathologic types. Here with we are presenting a case of nulligravida admitted for abnormal uterine bleeding with severe anaemia diagnosed as STUMP on histopathological examination, managed by myomectomy followed by regular follow up for the sake of fertility status. Post operative regular clinical and radiological follow up is mandatory for early pick up of recurrence or metastases.

1. Inroduction

STUMP (Smooth muscle tumor of uncertain malignant potential) is an extremely rare smooth muscle tumor that posses both benign and malignant features. It is diagnosed by the histopathological examination and the criteria like cytologic atypia, mitotic index and tumor cell necrosis differentiates leiomyoma from leiomyosarcoma. Presenting features of stump are consistent with uterine myomas[1] such as abnormal uterine bleeding, pain abdomen and pressure symptoms, which differs depending on the size of the uterine mass.[2] It is very difficult to distinguish STUMP from myoma based on radiological examination and confirmation is certain under microscope by pathological examination. WHO has classified STUMP as smooth muscle tumor between benign and malignant criteria.[3] Among the women undergoing Myomectomy or Hysterectomy for a presumed diagnosis of leiomyoma,0.01% receive a diagnosis of STUMP.[4] To preserve fertility myomectomy is considered in limited cases.[5] STUMP has some characteristics of leiomyosarcoma but does not meet the full diagnostic criteria. The STUMP was firstly used in literature by Kempson in 1973.[6] Treatment options, approach and follow up of the tumors have been still controversial, particularly in patients with fertility desire. Recurrence rate ranges from 8.7 to 11% and the possible recurrence is often delayed some years after the initial tumor.[7–9] The clinical presentation and surgical modalities have not been proven enough to distinguish STUMPs from neoplastic entities. The Stanford criteria of STUMP diagnosis include atleast two of the following:

1. Diffuse to moderate atypia
2. Mitotic count of atleast 10 mitotic figures/10 HPF

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3. Tumor cell necrosis[2]

Here we report a case of 31 year old Nulli gravida with a symptomatic huge myoma with severe secondary anaemia diagnosed as a STUMP after the histopathological examination of myomectomy specimen.

2. Case Study

A 31-year old, nulli gravida, married since a year presented to the gynaecological outpatient department with the complaints of general weakness, fatigue and giddiness. She also had a history of polymenorrhoea and heavy menstrual bleeding in the past one year. She had no other systemic diseases and no other surgical history. Her general examination revealed severe pallor. On physical examination of abdomen, there was a warm, hard, solid mass of about 18-20 weeks size with limited mobility. No particularities were noted in speculum examination. A large uterine mass was felt on bimanual examination. With all these history and clinical examination, the patient was provisionally diagnosed as a case of AUB/mass abdomen arising from the pelvis with anaemia. To support the clinical abdominal findings, CT abdomen was done and it revealed a bulky uterus with heterogeneous soft tissue attenuating mass lesion in uterus splaying the endometrial cavity measuring approximately 12.7 cm ×10.1 cm. Multiple hypodense areas noted within (?UTERINE FIBROID) Figure 1. Her haematological tests were normal except Severe anaemia of Hb – 1.9 gm% with all these findings, We came to a definitive diagnosis of AUB/ huge myoma uterus with severe secondary anaemia. After explaining the treatment options of anaemia correction by blood transfusions, myomectomy or hysterectomy (if myomectomy fails and if there is increased haemorrhage), an informed and written consent has been obtained. Anaemia correction done with four packed cell transfusion preoperatively. Proceeded with a plan of open myomectomy with biopsy. This was performed through a supra-pubic sub-umbilical transverse incision. We encountered a 20 weeks sized uterus with an intramural myoma of about 15 × 10 cm arising from the anterior wall of uterus. The myoma enucleated and the bed was sutured followed by muscular and serosal layers. The other parts of uterus and adnexa visualized and was found free from any significant abnormalities. There was no fluid collections and other significant findings suggestive of tumor or malignant deposits throughout the abdomen. Peritoneal wash given and after perfect haemostasis, abdomen was closed in layers. The surgery was difficult enough to require three more units of post-operative blood transfusion due to blood loss during the time of myomectomy. The surgery was uneventful and the specimen was sent for histopathological examination. The gross myomectomy specimen was a single irregular, grey white, soft tissue mass measuring 11×9×6 cms. When we were happy about the surgery which ended successfully with myomectomy, preserving the fertility of the nulligravida and not proceeding to hysterectomy, we were about to expose ourselves to a bombshell of histo-pathological report of the myomectomy specimen on day two of surgery as section showing a tumor composed of interlacing bundles of spindle shaped cells with spindle shaped nuclei, few giant cells and atypical cells with focal areas of cystic degeneration which is Suggestive of Smooth Muscle Tumor of Uncertain Malignant Potential (STUMP) Figure 2. The tumor showed no coagulative tumor cell necrosis and low mitotic index of less than 10/HPF. However, the cytologic atypia is only moderately reported. Further immuno-histochemistry revealed positive and patchy expression of p16, scattered positivity of p53 in few pleomorphic tumor cells and positive for Ki67 in many cells with a MIB index of 5-7% Figure 3. Overall risk of recurrence is < 10%. Expert oncologist opinion obtained, the patient along with the relatives were explained and counselled about the nature, progression of the disease, the risk of recurrence and the fertility options available. The patient was advised a clinical and radiological review every 6 months and yearly MRI (if needed) to rule out the recurrence and metastases. Further a single shot of Leuprolide 11.25 mg given subcutaneous on day three of surgery and the patient was discharged without any adverse event or complication.

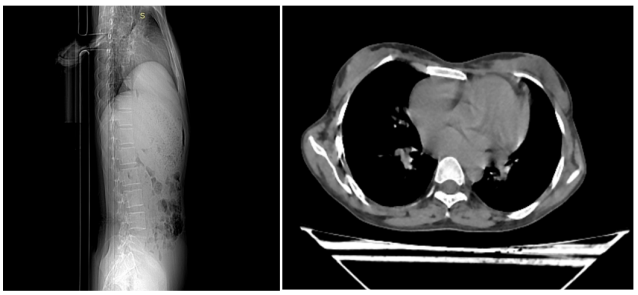

Fig. 1: CT abdomen showing a bulky uterus with heterogeneous soft tissue attenuating mass lesion in uterus splaying the endometrial cavity measuring approximately 12.7 cm ×10.1 cm. Multiple hypo dense areas noted within (Uterine Fibroid)

3. Discussion

STUMP is often a surprise finding after a myomectomy or hysterectomy. In our case, benign leiomyoma was the preoperative diagnosis because of age, size of the uterine mass, clinical features and radiological finding. The diagnosis confirmed after histo-pathological examination. Diagnosis of STUMP is difficult and it is very challenging to distinguish it from benign leiomyoma preoperatively with imaging.[10] STUMP is histologically characterized as a slowly growing and late recurrence borderline tumor.[2] Recurrence rate ranges from 6.9 to 27%.[2,5,10] Bell et al. reported three subdivisions of stump based on histology.

Table 1:

S.No	Sub types	Cytologic atypia	Mitotic index	CTCN
1.	SMT-LMP	Mild - moderate	< 10 MF/10 HPF	+
2.	ALL-LRR	Diffuse moderate - severe	< 10 MF/10 HPF	_
3.	AL-LE	Severe	< 20 MF/10 HPF	_

(**SMT-LMP**: Smooth muscle tumor of low malignant potential/ **ALL-LRR**: Atypical leiomyoma, low risk of recurrence/ **AL-LE**: Atypical leiomyoma with limited experience)

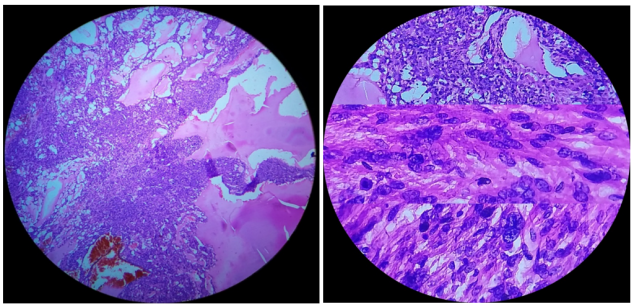

Fig. 2: Histopathological section showing a tumor composed of interlacing bundles of spindle shaped cells with spindle shaped nuclei. Few giant cells and atypical cells with focal areas of cystic degeneration (No coagulative necrosis, Mitotic index <10, moderate cellular atypia)

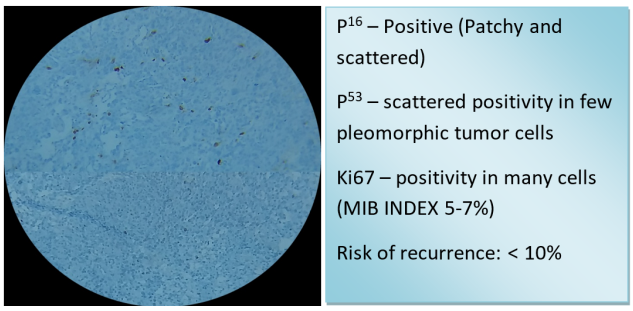

Fig. 3: Immunohistochemical study

STUMP can transform into low grade or high grade smooth muscle tumor and metastasize to other organs after years even in absence of recurrence risk factors such as presence of CTCN or diffuse cytologic atypia. STUMP represents one-third of uterine sarcomas and 1.3% of uterine cancers.[11] Our case was admitted for the abnormal uterine bleeding with severe anaemia, clinical examination revealed a warm, hard, irregular mass of about 18-20 weeks size arising midway to umbilicus. In the report by Guntupalli SR et al., that the mean age for STUMP is 45, our case was 31 years and which is slightly different from what is reported in studies. Treatment choices for STUMP after diagnosis is based on patient counselling and the desire to preserve fertility as there are no specific approved protocols. Hysterectomy prefers to be the gold standard in the event of STUMP diagnosed postoperatively in myomectomy specimens, considering the proved possibilities of recurrence for those who have completed family. For patients with fertility desire, fertility sparing surgery and adequate advice regarding the risk of recurrence and strict follow up is mandatory In our case, as patient is nulligravida, with a desire to have a child, we have not proceeded to hysterectomy, but advised her a regular clinical and radiological follow up.[12] Uterine smooth muscle tumors that show the some worrisome histologic features (CTCN, Cellular atypia and mitotic index) but do not fulfil the diagnosis of leiomyosarcoma are called as STUMPs. Presently, general studies have found a significantly reduced recurrence rate and a 5-year survival rate ranging from 92-100% compared to leiomyosarcoma. And there is no difference between the rate after myomectomy and hysterectomy.[10,13] STUMPs either recur as STUMPs[14] or leiomyosarcomas.[14–16] The treatment of choice for the recurrence is surgical excision followed by adjuvant therapy like pelvic irradiation, chemotherapy (Doxorubicin & Cisplatin), medroxy progesterone and GnRH analogue.[14–18] Even though the efficacy of adjuvant therapy is not proven, it is accepted and uneventful course was noted even in absence of such treatment.[19] In accordance with study by Atkins et al. and Ip et al. the immune histochemical assay for the over expression of p16 and p53 on histologic sample may be useful to identify the risk of recurrence.[9,20] Studies have shown 52% rate of successful pregnancy rates after myomectomies. Even though we don't have any well authorized protocols for follow up, studies suggest that, these patients need to have a follow up every six months for first 5 years and annually for next 5 years. During every visit, they should undergo clinical full body examination, laboratory investigation, chest X-ray, pelvic sonography/MRI and PET CT depending on the histologic features, may be necessary in fertility preserving patients to ensure timely management of potential recurrence.

4. Conclusion

As mentioned in literature, the STUMPs are smooth muscle tumors that show some worrisome features but does not fulfil all the criteria of leiomyosarcoma. These are tumors classified in between benign and malignant. There is no possibility of preoperative diagnosis. The diagnosis is certain by histopathological examination after surgical removal of either myoma or uterus. Patients with STUMP

have symptoms that are commonly found in patient with Abnormal uterine bleeding, pelvic pressure symptoms, symptoms of anaemia, mass abdomen and pelvic pain. The gynaecologists should keep in mind the possibility of uterine STUMPs whenever they come across a giant myoma uterus. The therapeautic schemes should be decided by keeping in mind the following – patient's age, fertility status and desire, pathologic features, recurrences and location of the tumor. Whatever may be the histologic types or location, the standard treatment option is surgical removal followed by post-operative follow up for early diagnosis of recurrence or metastases. We managed this case by myomectomy since we have diagnosed STUMP post-operatively and in view of fertility desire, we have advised a regular follow up followed by hysterectomy after completing her family, if all things are going well without any signs and symptoms of recurrence or metastases.

5. Conflicts of Interest

The authors declare that they have no conflict of interests.

6. Author's Contribution

The final paper has been seen and approved by all authors. The authors accept full responsibility for the design and conduct of the study, had access to the data, controlled the decision to publish.

7. Disclosure

The authors also report the absence of any significant financial support in any organization. The paper had not been published elsewhere previously.

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