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Benign Spindle cell carcinoma in a hysterectomized women : Case Report

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ABSTRACT: We present a case report of Benign spindle cell carcinoma in a 47 year old with a past history of hysterectomy in view of fibroid uterus in 2014. After 7 years , she presented with abdominal discomfort since 1 month.

KEYWORDS: Wandering fibroids, true cut biopsy, benign spindle cell carcinoma

I. INTRODUCTION

Benign metastasizing leiomyoma is a rare, benign, spindle cell neoplasm usually diagnosed in middle-aged women who have a history of uterine leiomyoma. The most common site of metastases is the lung although other sites such as the heart, bones, and lymph nodes have been reported. Although these tumors are metastatic deposits from the uterus, they have no malignant features morphologically or clinically and most have an indolent course. We present a young female with Benign spindle cell neoplasm attached to the gastro-intestinal tract.

II. CASE REPORT

A 47 year old women presented with complaints of lower abdominal discomfort since 1 month. She has undergone total abdominal hysterectomy with bilateral salpingectomy in 2014 in view of fibroid uterus. On general examination , her weight was 73kg and height of 163cm with a BMI of 27.4 kg/m^2.

On inspection , no abnormality noted and hysterectomy scar was present . On palpation , no mass was palpable. On Per speculum examination , vault was healthy. On per vaginal examination, hard mass was felt at the vault , which was fixed. On per rectal examination, same mass felt and rectal mucosa was free.

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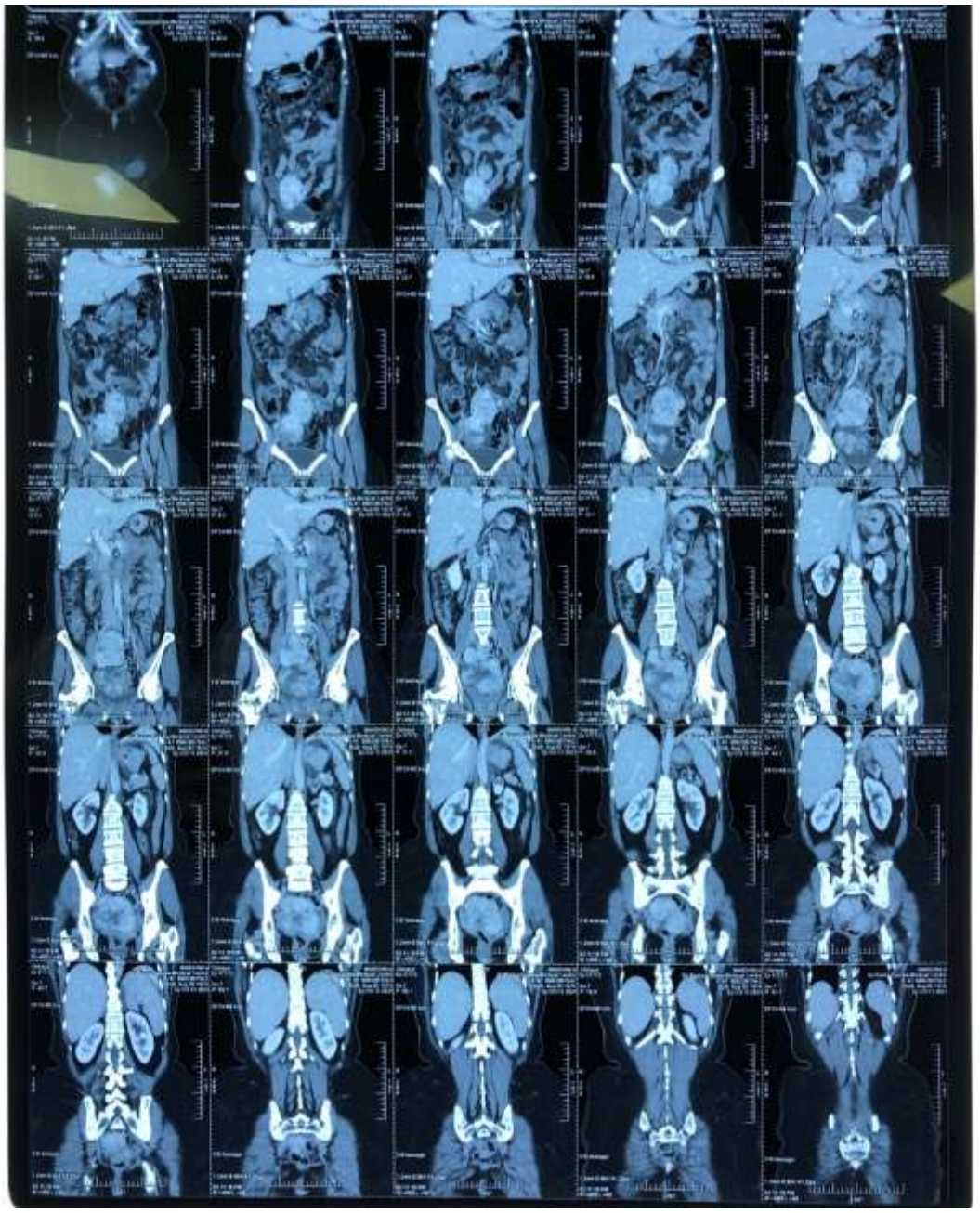
Her hemogram was normal and peripheral blood smear did not show any abnormal cells. Chest X ray was normal. Mammogram showed BIRADS-2. Pap smear was negative for intra-epithelial lesions. TVS pelvis and USG abdomen suggested the possibility of vaginal vault mass lesion. CECT abdomen showed multiple well defined heterogeneously enhancing lesions of varying sizes in the lower abdomen and pelvis , largest of size 6.3x4.4x5.9cm and also few lesions are seen to indent and displace the dome and posterior wall of the bladder.



Tumor markers revealed elevated CA125-59.11IU/L.CEA was normal : 0.65. Ultra sound guided true cut biopsy of the pelvic mass was done and showed benign spindle cell neoplasm. Based on the biopsy reports , a diagnosis of benign spindle cell neoplasm was made.

Staging Laparotomy with bilateral oophorectomy with bilateral urethroscopy with Double J stenting done. A 5x5cm mass over the mesentery of distal ileum , a 1x1 cm nodule over proximal ileal serosa seen. Bilateral ovaries

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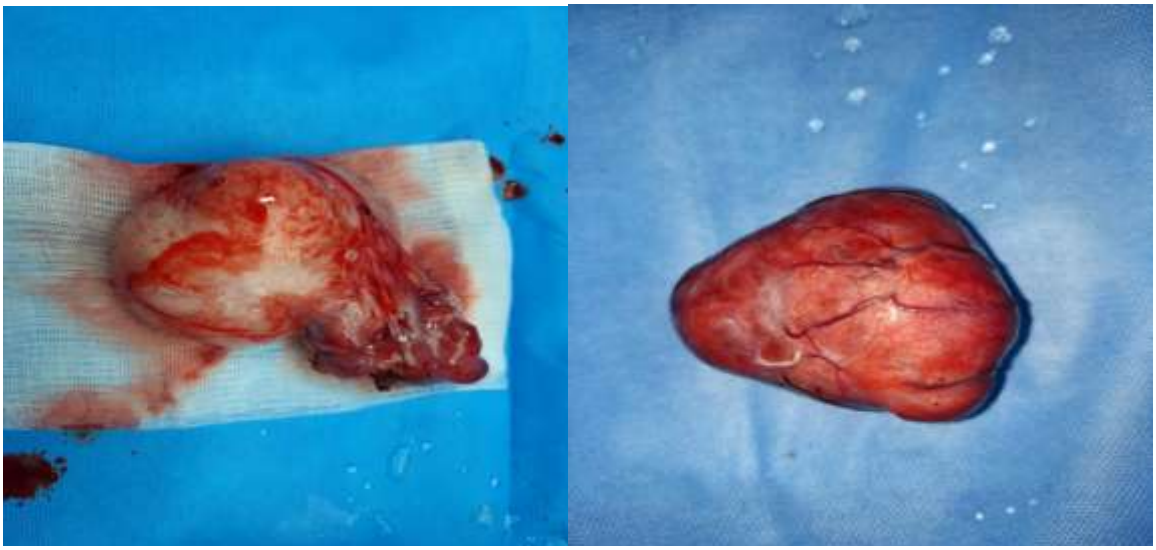
atrophic with 4cm fibroid in left adnexa noted. A 7x5cm mass over the Pouch of douglas peritoneum noted. No pelvic lymph node metastasis or para-aortic lymph node metastasis or omental metastasis was seen. Intra-operatively there was no surgical spill. Post operative period was uneventful.

Frozen section was sent and reported as benign spindle cell neoplasm.Immuohistological studies showed leiomyomatous origin.



III. DISCUSSION

Benign metastasizing leiomyoma (BML) is a rare entity. BML most commonly occurs in middle-aged women in the perimenopausal period. All patients have a history of uterine leiomyoma (ULM), with most women having had a hysterectomy. Most patients are asymptomatic, and the tumors are incidentally found when imaging studies are performed for unrelated reasons. The most common metastatic site is the lung; however, sites such as the heart, retroperitoneal lymph nodes, skin, and bones have been reported . As mentioned above, most cases are asymptomatic; however, patients may complain of chest pain, abdominal discomfort or pain abdomen. In our case, our patient was initially asymptomatic but later developed lower abdomen discomfort. Kayser et al. reported an average time of 14.9 years from time of hysterectomy to development of BML; however, Barnas et al. reported a median time of just under 9 years between hysterectomy and development of BML . Another study reported a median age of 46 years of which 82.6% of the patients had undergone a prior hysterectomy for ULM, with a median time from surgery to onset of BML being 10.5 years .

The pathogenesis of BML is not truly known. One theory is that there is hematogenous spread of benign endometrium cells into organs during the time of hysterectomy for ULM. Another theory is that they are leiomyosarcomas with low-grade malignant potential; as more cases are being reported, it seems unlikely that they are primary leiomyomas of the lung given the similarity between the uterine tumor and the metastatic site . Patton et al. reported a possible X-linked clonal inactivation suggesting that there is a monoclonality to this tumor . They also mention that telomere lengths do not play a role in this entity. Chromosomal abnormalities have been detected in BML. Nucci et al. described abnormalities in 19q and 22q while Lee et al. noted abnormalities in both the lung tumors and uterine tumors with translocations (12;14) and (1;2) noted . Abnormalities in chromosomes 1, 7, 13, and 14 have been reported as well . Mutations in BMP8B and MED12 genes have been identified, and mutations in ARID2 and NTRK1 and amplifications in BC11B and TCL1A have been noted too .

Microscopic findings of BML demonstrate characteristics of smooth muscle proliferation and tend to stain positive for actin, desmin, or vimentin. They do not to have a high mitotic rate, high Ki67, atypia, high degree of cellularity, or poor differentiation suggesting that they are benign tumors . They tend to be strongly positive for estrogen and progesterone, suggesting that they originate from the gynecological tract.

There are no standard treatment guidelines for BML. Since most cases are asymptomatic and tend to be slow-growing, they can be observed

unless symptoms develop. Surgical resection of metastatic site if feasible would be the preferred treatment modality . Given that these tumors are estrogen and progesterone positive, they tend to respond to endocrine therapy. Spontaneous regression during pregnancy and menopause has been noted suggesting that once patients attain menopause, there could be regression of these tumors . In nonresectable metastatic disease, manipulation of the hormones either via oophorectomy, GnRH analogs, ovarian ablation, tamoxifen, or aromatase inhibitors can be used. With endocrine therapy, most patients have stability of their masses or regression of the tumors. Most cases of BML have been reported in case reports; there has been no long-term follow-up of these patients to know if this entity can be fatal.

IV. CONCLUSION

Benign metastasizing leiomyoma is a rare condition usually seen in middle-aged perimenopausal women. Most women have a history of leiomyoma of the uterus with a median time to onset of BML approximately 10 years after definitive hysterectomy. Although these tumors by definition are metastatic deposits, they do not have any malignant features and tend to be slow-growing and asymptomatic. In our case, we presented a perimenopausal woman who developed BML 7 years after undergoing a hysterectomy. She had a vault lesion, and biopsy showed benign spindle cell neoplasm, and no carcinoma was seen. These tumors tend to be estrogen and progesterone positive suggesting a gynecological origin. Treatments include observation, surgery, or antiestrogen therapy.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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