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Recurrent Atypical Leiomyoma Arising From Vaginal Cuff after Hysterectomy- A Rare Case Report

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Introduction: *Leiomyoma are benign mesenchymal tumors presenting as the most common uterine neoplasm. Atypical leiomyoma (ALM), a nomenclature given in WHO classification[1], is a leiomyoma variant showing histopathological features ranging between a benign leiomyoma and leiomyosarcoma (LMS). It is a rare smooth muscle neoplasm detected mostly on histological examination in myomectomy and hysterectomy specimens. It usually arises from myometrium; however extrauterine sites such as cervix, vagina, retroperitoneum etc. have rarely been reported. ALM has a very low rate of extrauterine and intraabdominal recurrence (<2%) with a negligible risk for distant metastasis[2]. The case as being presented here has never been reported earlier, where ALM was not only recurrent but also arose at different sites, first from cervix, then uterine fundus and surprisingly twice from vaginal cuff after hysterectomy.*

Keywords: *Atypical leiomyoma, recurrence, extrauterine location*

I. Case Report

A 65 year old P4L4 postmenopausal hysterectomized patient presented in OPD at Dr. PDMMC hospital with chief complaints of something coming out per vaginum (SCOPV) and foul smelling vaginal discharge since one month. There were no associated bladder or bowel complaints. She had all vaginal home deliveries and last birth being 23 years back. She had attained menopause 11 years back. She was on anti-hypertensive drugs since 5 years.

Her past history suggested that she had recurrent complaints of SCOPV since seven years for which she had undergone surgery thrice during that period. The patient being illiterate, her past surgical history was extracted from the available records. In 2008, she had a pedunculated necrotic cervical polyp of size 5x4 cm prolapsing out through introitus for which she had undergone vaginal polypectomy, but the histopathology report (HPR) of the polyp was not available. On 26/1/2011, she presented with postmenopausal bleeding, discharge per vaginum and difficulty in micturition since 1 year. Her records showed that examination findings at that time were suggestive of 16 weeks firm mass arising from pelvis. On PS and PV examination, a huge 15x15 cm polypoidal mass was completely occupying the vagina with non- visualization of cervix. Her blood investigations were normal. Ultrasound study revealed a well defined mixed echogenic predominantly hypoechoic mass posterior to urinary bladder in the region of vagina and cervix most likely a malignant mass. CT scan corroborated with ultrasound findings, additionally showing bilateral hydronephrosis and hydroureter, possibly due to the above mentioned mass. With a provisional diagnosis of carcinoma cervix, she was posted for surgery on 3/2/2011. After opening the abdomen, a typical flower vase appearance of uterus was seen with a dimple present in place of fundus and both tubes and ovaries pulled into the center of the dimple. The diagnosis of chronic inversion of uterus was made. It was confirmed that the polypoidal mass felt through vagina was a large fundal fibroid causing uterine inversion. The ring of inversion was cut and the fibroid was pushed upwards to restore the normal anatomy. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was carried out. The HPR showed unremarkable features in endocervix, endometrium and bilateral adnexa. But the myometrial mass turned out to be Atypical Leiomyoma.

The patient was asymptomatic for the next 4 years. She presented in our hospital with a 5x5 cm pedunculated mass coming out through the introitus in Jan 2015. This time the polyp was arising from the vaginal vault for which she underwent polypectomy. HPR surprisingly confirmed atypical leiomyoma again.

Her present examination findings revealed a similar pedunculated mass of size 10x10 cm protruding out of introitus (Fig 1). The pedicle was 5cm long and 2cm thick attached to the vaginal vault. The lower pole was necrotic giving rise to offensive discharge PV (Fig 2). Under antibiotic cover, she was posted for surgery. Considering the recurrences in the past, a circular incision was taken on the vaginal vault and vaginal flaps were raised to get a clear margin around the pedicle. The pedicle was then excised and vaginal incision was sutured. Post operative period was uneventful and patient was discharged on 7th day. HPR revealed overlying stratified squamous epithelium of vagina immediately beneath which growth was seen showing spindle cell proliferation with areas more cellular than usual leiomyoma (Fig 3). The magnified view showed bizarre cells with diffuse nuclear atypia confirming it to be an atypical leiomyoma (Fig 4). On immunohistochemistry, growth was found to be smooth muscle cell actin (SMA) positive confirming it to be a smooth muscle tumor (Fig 5). There was no

involvement of pedicle and vaginal margins. Patient was advised to follow up regularly in OPD with the hope that she doesn't have recurrences in future.

II. Discussion

Atypical leiomyoma is also known as symplastic, bizarre and pleomorphic leiomyoma. It may occur spontaneously but is often seen in patients taking progestin compounds. This well circumscribed smooth muscle neoplasm has a gross appearance resembling that of a conventional leiomyoma but a few may have a more yellow to tan appearance, haemorrhagic areas, myxoid change and a softer consistency. This myoma variant is microscopically characterized by the presence of bizarre cells with enlarged pleomorphic often multiple nuclei which may be unifocal, multifocal or diffuse in their distribution. In addition to the presence of significant nuclear atypia, a defining histolological feature is a mitotic count less than 7 mitotic figures/ 10 high power fields (MF/10HPF). This is based on a series of Downes and Hart in 1997, in which tumors that met these criteria had a benign follow up for an average of 11 years with most patients being treated by hysterectomy[3].

These tumors, particularly with diffuse nuclear atypia treated by myomectomy alone should be diagnosed with care, as even in cases less than 2 MF/10HPF, recurrence at the surgical site and intraabdominal and pelvic recurrences have been noted. After adequate sampling ALM can be distinguished from leiomyosarcoma by the absence of coagulative tumor cell necrosis and <7 MF/10HPF, those tumors with 7-10 MF/HPF are best considered as smooth muscle tumors of uncertain malignant potential (STUMP). Additionally, LMS has >10 MF/10HPF, presence of aneuploidy, high MIB-1 activity and p53 positivity on immunohistochemistry while ALM are usually diploid with low MIB-1 activity and are negative for p53. It should be noted that LMS may show focal "bizarre" changes.

Majority of available data on ALM is related to those arising from uterus. A study by Sung C O et al on ALM of uterus with long term follow up after myomectomy showed local recurrence at the myomectomy site in one patient while none developing metastasis[4]. Similarly, in a clinicopathological study of 51 cases of ALM of uterus, Ly A et al inferred that these patients may be treated by myomectomy alone with successful pregnancy, but should be monitored for intrauterine residual or recurrent disease[2]. There is a definite scarcity of knowledge regarding ALM arising from vagina. Biankin SA et al reported a case of 44 year old patient with bizarre leiomyoma of the vagina in 2000[5]. A case of usual leiomyoma originating from vaginal cuff in a 70 year old woman, 25 years after total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH with BSO) was reported by Yarci A in 2010[6].

This case is unique because this patient had ALM in the fundus of uterus initially and within 4 years of TAH with BSO, she had recurrence of ALM from the vaginal cuff twice 6 months apart.

III. Conclusion

ALM can be considered to be a benign tumor for management purposes, although in cases with diffuse atypia, an intrauterine or extrauterine recurrence may be experienced, hence emphasizing on the regular follow up of these patients. In addition, adequate sampling of the myomectomy and hysterectomy specimens is required to differentiate ALM from leiomyosarcoma, also taking the help of IHC as there is a close window in differentiation between the two.

Declarations
Conflict of interest: None
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Ethical approval: Not required

References

[1]. WHO histological classification of tumors of the uterine corpus. IARC publications, Lyon, France, p 218. www.iarc.fr/en/publications/pdfs-online/pat-gen/bb4/bb4-chap4.pdf.
[2]. Ly A, Mills M, Mckenney J K et al. Atypical leiomyoma of uterus: a clinicopathologic study of 51 cases. Am J Surg Pathol.2013 May; 37(5):643-9.
[3]. Downes K A, Hart W R. Bizarre leiomyomas of the uterus: a comprehensive pathologic study of 24 cases with long term follow-up. Am J Surg Pathol.1997; 21: 1261-1270.
[4]. Sung C O, Ahn G, Song S Y et al. Atypical leiomyoma of uterus with long term follow- up after myomectomy with immunohistochemical analysis for p16INK4A, Ki-67, estrogen receptors and progesterone receptors. Int J Gynecol Pathol. 2009 Nov; 28(6):529-34.
[5]. Biankin S A, O'Toole V E, Fung C et al. Bizarre leiomyoma of the uterus- a case report. Int J Gynecol Pathol. 2000 Apr; 19(2):186-7.
[6]. Yarci A, Bayramov V, Sukur Y E et al. Vaginal vault leiomyoma: 25 years after total abdominal hysterectomy. J Minim Invasive Gynecol. 2010 Jan- Feb; 17(1):116-7.

Figures

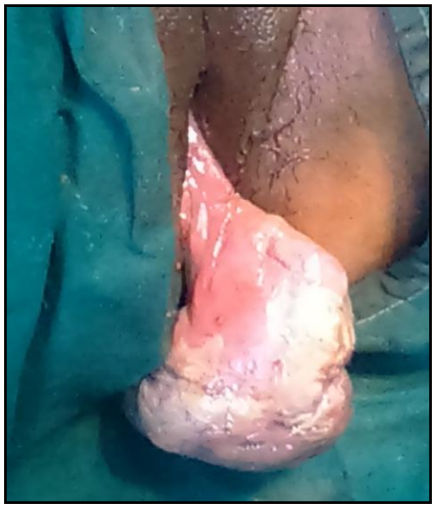

Fig. 1 Pedunclated myoma prolapsing out of introitus

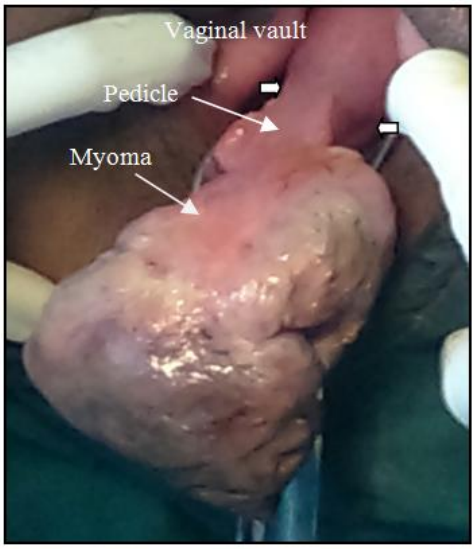

Fig.2 Myoma with thick pedicle attached to vaginal vault and a necrotic base. Block arrows denote the angles of vaginal cuff.

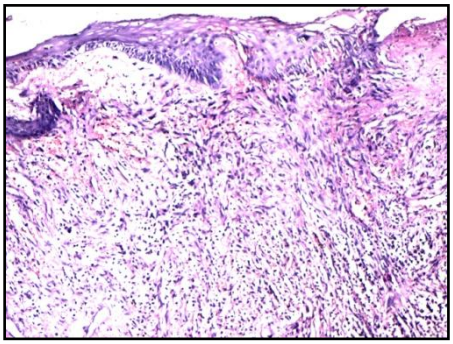

Fig 3. H&E stain section shows overlying squamous epithelium and beneath it is growth showing spindle cell proliferation

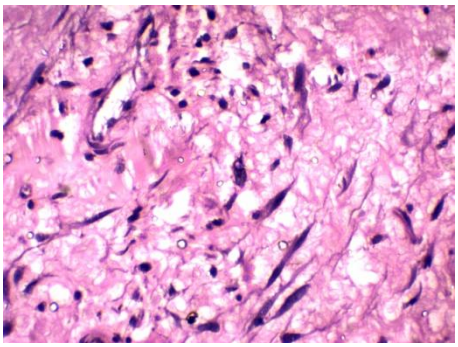

Fig. 4 Magnified view reveals hypercellularity and widespread atypical nuclei

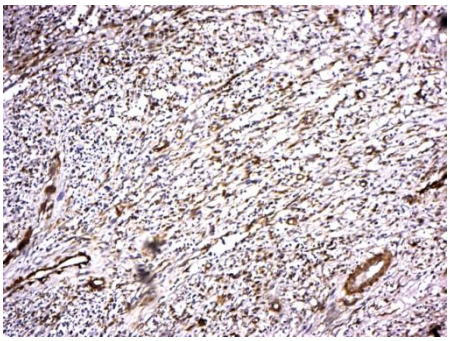

Fig. 5 Immunohistochemistry showed tumor to be SMA positive