

Primary vaginal leiomyoma in a hysterectomised woman presenting with urgency

Samettin CELIK¹, Canan SOYER CALISKAN¹, Safak HATIRNAZ², Huri GÜVEY³,

¹ Maternity Hospital, A Branch of Training and Research Hospital, Samsun, TURKEY

² IVF Center, Medicana International Hospital, Samsun, TURKEY

³ Obstetrics & Gynecology, Private Office, Kutahya, TURKEY

43

SUMMARY

Primary vaginal leiomyoma is a pathological diagnosis of a rarity which arises mostly from the anterior vaginal wall. Usually presented with symptoms related to the compression of the mass and clinical diagnosis seems a dilemma. We report a case of a 45 years old hysterectomised woman having a mass in the anterior vaginal wall with urinary symptoms who was initially evaluated as Gardner's cyst and operated vaginally but intraoperative findings and pathological report were correlated with leiomyoma. Surgical removal of the vaginal leiomyoma is the preferred treatment by ruling out the malignant potential. Any vaginal mass with or without symptoms should be evaluated carefully before surgical removal. Patients should be monitored carefully for the development of late recurrences following proper surgical removal.

Corresponding Author: Huri GÜVEY huriguvey@gmail.com

Received: March 17, 2021; **Accepted:** April 18, 2021; **Published Online:** April 21, 2021

Cite this case report as: Celik, S., Soyer Caliskan, C. & Hatirnaz, S. & Guvey, H. (2021). Primary vaginal leiomyoma in a hysterectomised woman presenting with urgency. *European Journal of Human Health* 1(1), 43-46.



Introduction

Vaginal masses are rare and range from Gardner's cyst, hemangioma, papilloma, mucus polyps, leiomyoma and even leiomyosarcomas (1-5). Primary leiomyoma of vagina may be associated with other leiomyomas.

The number of cases ever reported is around 350 which means that this is a quite rare clinical entity. First recorded case was reported in 1733(1-3). Bennet and Ehrlich evaluated 15000 autopsies and 50000 surgical specimens and reported 1/15000 9/50000 primary vaginal leiomyomas respectively (3). Symptoms and signs are related to the size and location of the mass and the patients may complain about wide variety of symptoms (4). Primary vaginal leiomyoma can mimic adnexial masses(5). It is reported that an atypical primary vaginal leiomyoma successfully removed can recur as a leiomyosarcoma which is much rarer compared to leiomyoma(6,7).

Case Report

Here we present an case of 45 years old woman who had a history of total abdominal hysterectomy and bilateral salpingooforectomy complaining about a mass in her vagina and urgency in micturition. The patient was evaluated vaginally and accepted as Gardner's cyst intially and operated for cyst removal but intraoperatively it was found a capsulated leiomyoma which was verified by pathological diagnosis.

Discussion

Uterine fibroids (also known as myomas or leiomyomas) are benign monoclonal neoplasms of the myometrium that represent the most common gynecologic tumor; they may be single or multiple, large (more than 10 cm) or small. Extra uterine leiomyomas are extremely rare tumors and commonly encountered in women aged between 30-50 years. Having a wide variety of symptoms, bit is a real diagnostic challenge and the diagnosis can be made during surgical procedure as in this case. The tumor may be palpated as solid or cystic which may be misleading as in our case. Location at the anterior

vaginal wall may result in urgency, stres urinary incontinance, disuria, disparouneia and a sense of fullness (8). In this case the mass made the patient complain about urgency in micturition and a mass filling the vagina.

Diagnostic measures like transvaginal ultrasound, Magnetic Resonance imaging (MRI) ve biospy of the mass may be of value but correct diagnosis can not be made in particular cases (5,9). Since the mass in the vagina was very smooth and cystic in nature and the patient was hysterectomised before, The surgeon's presumptive diagnosis was Gardner's cyst and surgical removal was decided. As in most of the cases, diagnosis could be made during surgical procedure and pathological report of the surgical specimen postoperatively. MRI is a valuable tool for detecting the rapid growth and leiomyoma recurrence since these findings may be related with sarcomatous transition of the tumor itself (6,10). Among the primary vaginal tumors, leiomyosarcoma is the leading tumor type and it may be primary tumor or a tumoral transition from atypical leiomyomas and leiomyosarcomas can be seen in patients with mullerian abnormalities due to close embryonic origin (11).

The development of leiomyomas either uterine or extrauterine may be correlated with the genetic variations. MED12 mutations, resulting in alterations in cell signaling and increased cell proliferation; found in about 70% of fibroids (12). Rearrangements at 12q14-15, resulting in overexpression of the HMGA2 protein, which inhibits cell death (OMIM 150699); found in about 7.5% of fibroids (12,13). Deletions within Xq22.3 locus of *COL4A5* and *COL4A6* genes, which results in enhanced cell proliferation (14). Inactivation of the fumarate hydratase gene; occurs in a small percentage of sporadic fibroids and in the rare hereditary syndrome of leiomyomatosis and renal cell cancer (14,15).

It is important to remember that primary vaginal leiomyoma may or may nor be seen with other leiomyomas. The behavior of vaginal leiomyoma may have some differences compared to uterine leiomyomas. Differantiation of vaginal leiomyomas

from leiomyosarcomas has no visible corners and smooth muscle neoplasms interspersed with atypical cells without mitotic figures and coagulative necrosis. These tumors are categorized as smooth muscle tumor of uncertain malignant potential (STUMP) subgroup of atypical leiomyoma of limited experience according to the world Health Organization revised criteria (16). Based on this knowledge, patients diagnosed as primary vaginal leiomyosarcoma need to be followed up carefully due to the potential to develop a recurrence as leiomyosarcoma.

In this case, the decision making for surgical removal was based on the smooth surface and cystic appearance of the anterior vaginal wall mass which was thought to be a simple Gardner's cyst in a hysterectomised woman. No possible malignancy or other vaginal tumors were discriminated by any of the diagnostic tools which is an absence for a correct diagnosis and proper management. However correct diagnosis can be put during surgery or histopathologically in most of the cases. The presumption of leiomyoma by the clinical findings is not easy and is mostly incidental.

In conclusion, surgical removal of the vaginal leiomyoma is the preferred treatment by ruling out the malignant potential according to revised WHO criteria. Vaginal surgery is the best option in hysterectomised patients. Patients should be monitored carefully for the development of late recurrences following proper surgical removal. Any vaginal mass with or without symptoms should be evaluated carefully before surgical removal.

Acknowledgement: The authors wish to thank to Dr Gülin Şimşek from pathology department due to contributions to study.

Funding: No funding for this case study

Conflict of interest

The authors declare that there is no conflict of interest.

References

1. Zuber I, Nadkarni PK, Nadkarni AA, Nadkarni A. Unusual clinical presentation of rare case of vaginal leiomyoma: a case report. *Int J Reprod Contracept Obstet Gynecol.* 2016;5(6):2047-2048.
2. Young SB, Rose PG, Reuter KL. Vaginal fibromyomata: Two cases with preoperative assessment, resection and reconstruction. *Obstet Gynecol.* 1991;78(5):972-974.
3. Bennett HG, Jr, Erlich MM. Myoma of the vagina. *Am J Obstet Gynecol.* 1941;42:314-320.
4. Curtin JP, Sorgo P, Slusher B. Soft tissue sarcoma of the vagina and vulva: a clinopathologic study. *Obstet Gynecol.* 1995;86(2): 269–272.
5. Gupta V and Arya P. A rare case of vaginal fibroid presenting as ovarian tumour. *Journal of Obstetrics Gynaecology of India.* 2006;5(6):537-538.
6. Cooney EJ, Borowsky M, Flynn C. Case report: Atypical, 'symplastic' leiomyoma recurring as leiomyosarcoma in the vagina. *Gynecologic Oncology Reports* 14 2015;14: 4–5.
7. Adeyemi AO, Aloy OU, Emmanuel O, Habibat FK, Luqman AA, Olaniyi AK, Jubril OK, et al. Primary vaginal leiomyosarcoma: case report of a rare gynaecological malignancy and diagnostic challenge in a resource-constraint setting, *Oxford Medical Case Reports.* 2020;2020(9):omaa081
8. Egbe TO, Kobenge FM, Metogo JAM, Wankie EM, Tolefac PN, Belley-Priso E. Vaginal leiomyoma: medical imaging and diagnosis in a resource low tertiary hospital: case report. *BMC Women's Health.* 2020;20:12.
9. Halder A and Mandal RD. Vaginal leiomyoma presenting as polyp. *Journal of Science.* 2015;5(10):915-916.
10. Shadbolt CL, Coakley FV, Qayyum A, Donat SM. MRI of vaginal leiomyomas. *J Comput Assist Tomogr.* 2001;25(3):355-357.

11. Zakashansky K, Peiretti M, Mahdavi A, Chun JK, Nezhat F. Combined laparoscopic and radical vaginal treatment of primary vaginal leiomyosarcoma in a patient with unicornuate uterus and pelvic kidney. *Journal of Minimally Invasive Gynecology*. 2007;4(14): 518–520.
12. Moravek MB and Bulun SE. Endocrinology of uterine fibroids: steroid hormones, stem cells, and genetic contribution. *Curr Opin Obstet Gynecol*. 2015;27(4):276-283.
13. Leiomyoma, uterine. Online Mendelian Inheritance in Man. OMIM website. Johns Hopkins University. Updated March 3, 2013. Accessed September 11, 2018. <https://www.omim.org/entry/150699>
14. Mehine M, Mäkinen N, Heinonen H-R, aaltoren LA, Vahteristo P. Genomics of uterine leiomyomas: insights from high-throughput sequencing. *Fertil Steril*. 2014; 102(3):621-629.
15. Fumarate hydratase. Online Mendelian Inheritance in Man. OMIM website. Johns Hopkins University. Updated September 9, 2016. Accessed September 11, 2018. [https://www.omim.org/entry/136850?search=fumarate hydratase gene&highlight=fumarate hydratase gene](https://www.omim.org/entry/136850?search=fumarate%20hydratase%20gene&highlight=fumarate%20hydratase%20gene)
16. Ip PP, Tse KY, Tam KF. Uterine smooth muscle tumors other than the ordinary leiomyomas and leiomyosarcomas: a review of selected variants with emphasis on recent advances and unusual morphology that may cause concern for malignancy. *Advances in anatomic pathology*, 17(2), 91-112.