

CASE REPORT

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Vaginal vault leiomyoma after hysterectomy. Case report

Leiomioma de cúpula vaginal posterior a histerectomía. Reporte de caso

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ABSTRACT

Leiomyomas are benign, mesenchymal tumors that usually arise from uterine smooth muscle cells, but can also occur in atypical sites such as the vagina, lungs and vascular structures. Post-hysterectomy vaginal vault leiomyomas are very rare and their etiology has not been determined. Transvaginal ultrasound, computed tomography and magnetic resonance imaging are useful tools for the diagnosis and follow-up of these patients. The definitive treatment is total removal of the tumor to avoid dissemination or inadvertent spillage of occult malignant neoplastic cells. Administration of gonadotropin-releasing hormone analogs or intravascular embolization may be alternative methods to reduce intraoperative blood loss. A case of vaginal vault leiomyoma following hysterectomy is presented.

Key words: Vagina, Leiomyoma, Hysterectomy.

RESUMEN

Los leiomiomas son tumores benignos, mesenquimales que generalmente surgen de las células del músculo liso uterino, pero también pueden aparecer en sitios atípicos como vagina, pulmones y estructuras vasculares. Los leiomiomas de cúpula vagina posterior a la histerectomía son muy raros y su etiología no ha sido determinada. La ecografía transvaginal, tomografía computarizada y la resonancia magnética son herramientas útiles para el diagnóstico y seguimiento de estas pacientes. El tratamiento definitivo es la extirpación total del tumor para evitar la diseminación o derrame inadvertido de células neoplásicas malignas ocultas. La administración de análogos de la hormona liberadora de gonadotropina o la embolización intravascular pueden ser métodos alternativos para reducir la pérdida hemática intraoperatoria. Se presenta un caso de leiomioma de cúpula vaginal posterior a histerectomía.

Palabras clave. Vagina, Leiomioma, Histerectomía.

INTRODUCTION

Leiomyomas represent benign clonal proliferation of smooth muscle cells. They are the most common benign mesenchymal tumors of the uterus and can occur in any tissue containing smooth muscle cells, such as soft tissues of the skin, lungs, intravascular space, retroperitoneal area, bowel, eye, heart, lung, breast and bladder⁽¹⁾.

The appearance of a leiomyoma in the vaginal vault long after hysterectomy is an extremely rare condition with symptoms that, depending on the size and anatomical location, can produce dyspareunia, dysuria and local pain. These must be surgically removed, due to possibility of malignancy and other complications⁽²⁾. A case of vaginal vault leiomyoma following hysterectomy is presented.

CLINICAL CASE

This was a 61-year-old menopausal patient, gestation 2, para 2, who consulted for presenting pain in the hypogastrium and left iliac fossa of 3 months of evolution, accompanied by dark brown vaginal discharge. She reported a history of vaginal hysterectomy with preservation of both adnexa due to metrorrhagia and uterine leiomyomas 15 years ago. The deliveries had been spontaneous, vaginally and without complications. The patient denied the use of hormone replacement therapy during menopause, smoking, alcohol consumption, illicit drugs, as well as personal or family history of neoplasms.

On physical examination, the abdomen was flat, depressible and there was a palpable tumor of pelvic origin, hard, solid, with irregular edges and immobile, approximately 9 centimeters, apparently adhered to the vaginal vault. Rectal examination showed that the tumor was located in the area corresponding to the right utero-sacral ligament. Speculotomy exposed super-anterior deviation of the vaginal vault, with the presence of a brown vaginal discharge, not foul-smelling, in scant to moderate quantity. No erosions or macroscopic lesions were observed.

The results of complete hematology, electrolytes, liver and renal function, urine and coagulation tests were normal. Values of tumor markers CA-125, CA 19-9 and carcinoembryonic antigen were within normal ranges. Transvaginal ultrasound showed a solid tumor in the pelvic area, 9 centimeters in diameter, with high vascularization and evidence of neovascularization on Doppler evaluation. Magnetic resonance imaging showed the presence of a single smooth-walled tumor in the vaginal vault, measuring approximately 10 x 8 centimeters, with homogeneous

signals on T1- and T2-weighted images (Figure 1). No intra-abdominal neoplasms or lymphadenopathies were observed. In view of the findings, it was decided to plan surgery to exclude the possibility of malignant pelvic neoplasia.

During exploratory laparotomy, a solid, well-circumscribed tumor measuring approximately 15 x 10 centimeters was found firmly attached to the vaginal vault, extending to the bilateral pararectal areas (Figure 2). Tumor was excised in its entirety along with both adnexa. There were no signs of abdominal or pelvic neoplastic disease.

Macroscopic evaluation by pathology showed that the tumor was rounded, firm and well circumscribed, approximately 14 x 10 centimeters. Microscopic examination found epithelioid cells with characteristic spindle-shaped, blunt-tipped nuclei and ample eosinophilic cytoplasm, without evidence of cellular atypia, with high isomorphism and cellular mitoses with low mitotic activity (0-1 / 10 high resolution fields) (Figure 3). Immunohistochemistry was positive for vimen-

FIGURE 1. MAGNETIC RESONANCE IMAGING. THE ARROWS INDICATE THE LESION IN THE PELVIS.

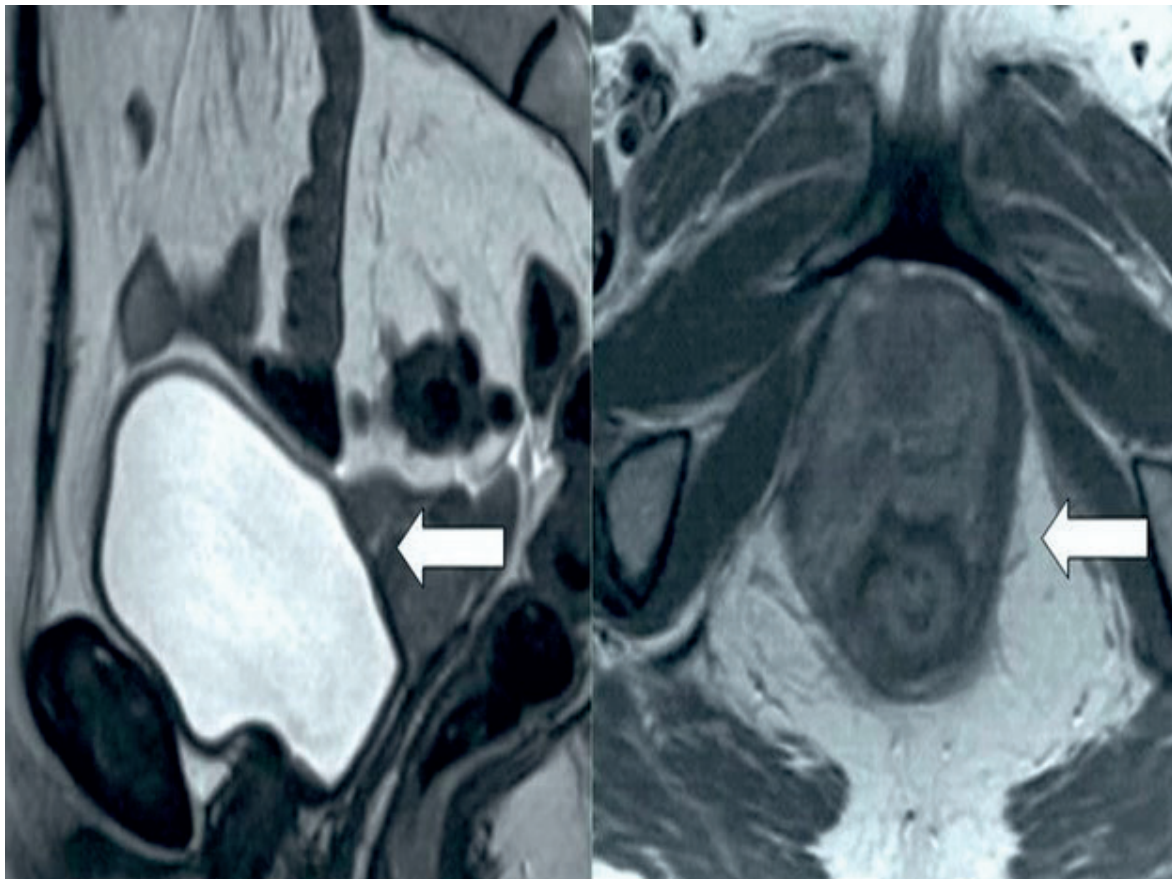




FIGURE 2. LEIOMYOMA ARISING FROM THE VAGINAL VAULT.

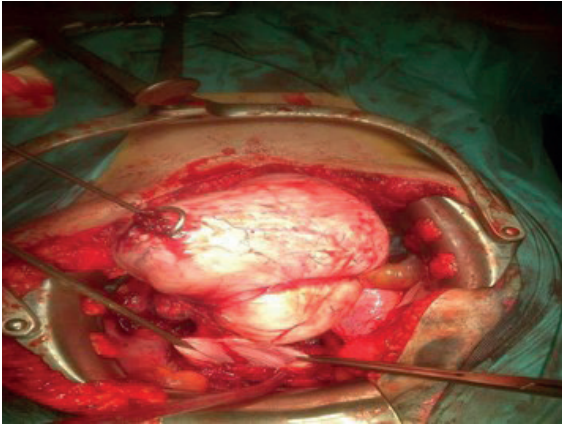
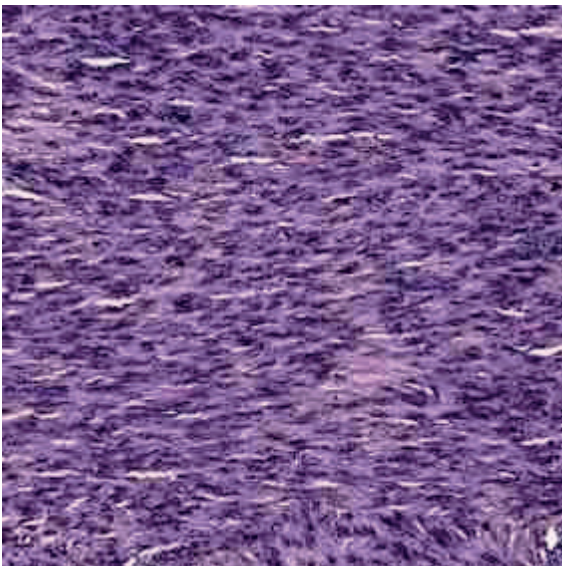


FIGURE 3. MICROSCOPIC IMAGE OF VAGINAL LEIOMYOMA WITH EPITHELIOID CELLS WITH CHARACTERISTIC BLUNT-TIPPED SPINDLE-SHAPED NUCLEI AND LARGE EOSINOPHILIC CYTOPLASM.



tin and smooth muscle actin. Both ovaries and fallopian tubes were normal. The final diagnosis was vaginal vault leiomyoma.

The patient had no postoperative complications and was discharged on the third day. During the 18-month follow-up she has had no symptoms or evidence of recurrence of leiomyomas in any location.

DISCUSSION

Myomas are the most common pelvic tumors in women. Despite its high frequency in the general population, vaginal leiomyoma is an uncommon entity⁽³⁾. The occurrence of these tumors following hysterectomy is extremely rare and there are only reports of less than 10 cases and

all patients had a history of uterine myomatosis. The clinical manifestation interval ranges from 1 to 25 years⁽⁴⁾.

The etiology of post-hysterectomy vaginal vault leiomyoma is unknown. Some hypotheses propose hematogenous expulsion of uterine leiomyoma cells to adjacent organs, which would lead to the appearance of the tumor. Another theory proposes that it may arise from vaginal smooth muscle layer, from structures in the submucosal planes, from the round ligament or from the differentiation of mesenchymal stem cells⁽²⁾. This process seems to be enhanced by polygenic hereditary predisposition to develop leiomyomatosis, probably due to ethnic predisposition. It is also important to highlight that the use of exogenous estrogen or in supraphysiologic quantity can be a predisposing factor for this condition⁽⁵⁾.

Vaginal vault leiomyomas after hysterectomy may be asymptomatic or produce symptoms such as dyspareunia, dysuria, pollakiuria, lumbar pain, constipation, genital discharge due to erosion and pressure in the genital region. The severity of the symptomatology depends on the site and size of the lesion, since they are closely related to the compression of other nearby organs^(6,7).

Imaging diagnosis of vaginal vault leiomyoma can be difficult, because it could extend beyond the area of origin and simulate a malignant neoplasm. The most commonly used diagnostic techniques are transvaginal ultrasound and magnetic resonance imaging. Ultrasonography has a diagnostic sensitivity and specificity of 96% and 95%, respectively⁽⁸⁾. The lesion is solid, with a spiral appearance and sometimes hypoechoic. It usually presents acoustic shadows with scattering to the surrounding tissues. On Doppler evaluation, it appears with circumferential vascularization and high velocity flow along with low resistance index, indicative of neovascularization⁽⁹⁾.

Magnetic resonance imaging is useful to show the extension and its relationship with neighboring structures. Leiomyoma may appear as a single smooth tumor with low and homogeneous signal intensity on T1- and T2-weighted images. However, those tumors of cellular and vascular



type may show hyperintensity in T2-weighted images and marked contrast enhancement⁽¹⁰⁾. Nevertheless, it is always necessary to consider the possibility of malignancy, so histopathological confirmation is the diagnostic standard to rule out the presence of malignant neoplastic components⁽⁶⁾.

The definitive treatment of post-hysterectomy vaginal vault leiomyoma is total resection to prevent inadvertent spread or spillage of occult malignant neoplasms. Surgical excision can be performed vaginally, laparoscopically or abdominally, depending on size, location and accessibility of tumor. Transvaginal myomectomy by transluminal endoscopy through natural orifices is also a feasible alternative. These fibroids are usually easily dissected without severe blood loss. In those cases with large tumors, administration of gonadotropin-releasing hormone analogs or intravascular embolization may be effective methods to avoid intraoperative bleeding⁽¹¹⁾. Although recurrence is uncommon, there is one report of a recurrent giant vaginal leiomyoma⁽¹²⁾.

Leiomyomas are hormone-dependent tumors that generally increase in size in premenopausal women and decrease at menopause. It has been proposed that the increase in volume depends on the complex relationship between hormones and locally active factors⁽¹³⁾. The presence of receptors for estrogen, progesterone, and epidermal growth factor has been documented⁽¹⁾. Several investigators have proposed that there is evidence of recurrence of benign lesions when adnexa are preserved during hysterectomy. These findings would indicate that tumor growth could be controlled with bilateral oophorectomy⁽²⁾. Long-acting gonadotropin-releasing hormone analogs suppress endogenous gonadotropin secretion and gonadal steroid production, so they may be useful in treatment in certain cases⁽¹⁴⁾. There are also reports indicating that progesterone may be effective in specific cases⁽¹⁵⁾.

In conclusion, although rare, vaginal vault leiomyomas can appear sometime after hysterectomy. This condition should be considered as a differential diagnosis of pelvic mass in patients with a history of hysterectomy with or without adnexal preservation, as they can develop up to 25 years after surgery. Surgical removal is the definitive treatment.

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