Benign Metastasizing Leiomyoma with Multiple Metastasis of Lungs and Bones

Leiomioma Benigno Metastático con Múltiples Metástasis en Pulmones y Huesos

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ABSTRACT

Background: benign metastasizing leiomyomas (BML) represents a rare disease consisting of the extra-uterine metastasizing of smooth muscle cells with similar histological, immunological, and molecular patterns to those of benign uterine leiomyomas. They are characterized by low mitotic activity, lack of anaplasia or necrosis and bad vascularization. BML is a rarely found entity with few documented cases in the literature, usually occurring in women of reproductive age with a history of myomectomy or hysterectomy. The leiomyomas can metastasize to several organs, the lungs being the most commonly affected. Case presentation: we present a case of a 40 year old infertile woman, with BML with multiple metastasis (lungs and bony: lumbar spine, pelvis, knee, and humerus), firstly she presented low back complaints, refractory to medical treatment, and was submitted to percutaneous L3-L5 fixation and transpedicular biopsy. Later on she was submitted to hysterectomy and bilateral ovariectomy with 7 years follow up. Good resolution of pain after surgery, 5 years after surgery she had SARS-CoV2 pneumonia, associated with coinfection with Aspergillus niger and Cryptococcus laurenti with 37 days of hospitalization, including 13 days in Intensive Care Unit (ICU) with good evolution.

Keywords: Leiomyoma; Benign; Hysterectomy

RESUMEN

Introducción: la BML representa una enfermedad rara (trastorno) que consiste en la diseminación extratérmina de las células del músculo liso. Estas lesiones exhiben patrones histológicos, inmunológicos y moleculares similares a los observados en los leiomiomas uterinos benignos. Las BML se caracterizan por una baja actividad mitótica, ausencia de anaplasia o necrosis y vascularización inadecuada. A pesar de su rareza, las BML se han documentado en un número limitado de casos, afectando principalmente a mujeres en edad reproductiva con antecedentes de miomectomía o histerectomía. Entre los órganos susceptibles de metástasis, los pulmones son el sitio más comúnmente afectado. Reporte de caso: presentamos el caso de una mujer infértil de 40 años diagnosticada de LMB con múltiples metástasis en ambos pulmones y en varias localizaciones óseas, entre ellas columna lumbar, pelvis, rodilla y húmero. Inicialmente la paciente presentaba dolencias lumbares refractarias al tratamiento médico. La biopsia transpedicular se obtuvo durante la fijación percutánea de L3-L5, seguida de histerectomía y oforectomía bilateral. La paciente ha estado en seguimiento durante 7 años. La paciente experimentó una reducción significativa del dolor después de la cirugía y no se observaron metástasis adicionales. Sin embargo, cinco años después de la cirugía, desarrolló neumonía por SARS-CoV2, asociada a coinfección por Aspergillus niger y Cryptococcus laurenti, que requirió 37 días de hospitalización, incluidos 13 días en Unidad de Cuidados Intensivos (UCI). Alentadoramente, la paciente demostró una evolución favorable durante este período difícil.

Palabras-Clave: Leiomioma; Benigno; Histerectomía

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Case Report

INTRODUCTION

BML is an infrequent condition that consists of metastatic dissemination of uterine leiomyomas to distant sites while maintaining benign pathologic characteristics. BML primarily affects menopausal or postmenopausal women who have previously undergone hysterectomy for leiomyomas, often several years after initial surgery\(^1\)\(^-\)\(^3\). While spinal involvement is uncommon\(^1\), the occurrence of multiple leiomyomas in BML patients is exceedingly rare\(^6\). BML is a rare cause of extra pelvic metastasis, predominantly to the lungs, often associated with prior gynecological instrumentation\(^7\). According to some authors, BML presents itself in extra-uterine sites, namely lungs (79.5%), lymph nodes, abdominal and pelvic cavity, nervous system, and bone\(^8\). Although it is relatively uncommon, BML accounts for approximately 4.4% of all benign soft tissue tumors\(^9\).

CASE PRESENTATION

A 40 years old woman, married, no children, previously healthy, presented with severe lumbar pain, unresponsive to analgesics. No localizing neurological deficit was detected. Radiological investigations, including X-Ray, CT scan and MRI of lumbar spine, revealed tumoral infiltration of L4 vertebral body with canal stenosis (Figure 1). Further examinations detected tumours on additional sites, namely on the knee (Figure 2A), pelvis and uterus (Figure 2B), lung (Figure 2C), and humerus (Figure 2D).

The patient underwent percutaneous fixation after transpedicular biopsy, accompanied by an attempted of anterior and posterior cementation procedure, which was aborted due to leakage (Figure 3). Under the same anesthesia, a biopsy of the pelvic tumoral mass of the pelvis was also performed. Histopathological examination confirmed metastasizing benign leiomyoma. Hysterectomy and bilateral oophorectomy followed after 2-month recovery.

The patient has been closely monitored for 7 years and has displayed excellent overall outcome. Adequate pain control has been achieved, requiring only occasional analgesics. The remaining tumoral mass have remained stable throughout the follow-up period.

Additionally, the patient overcame a challenging infection 5 years after surgery, when she developed SARS-Cov2 pneumonia, complicated by coinfection with Aspergillus niger and Cryptococcus laurenti. At this juncture, she required a 37-day of hospitalization, including 13 days in the IUC, and exhibited favorable evolution.

Figure 1. X-Ray, CT scan and MRI revealing mass involving L4 vertebra.
Figure 2. A. Left, X-ray; Right, MRI: tumor involving tibia. B. Left, X-ray - tumor of left iliac crest; Right, CT scan: tumor of iliac crest and uterus. C. Left pulmonary nodule. D. Humerus tumor.
CONCLUSION

Our case report highlights the diagnostic and therapeutic intricacies and challenges associated with BML, which is an exceptionally rare condition. Despite some literature suggesting an indolent nature, our patient debuting symptom was severe lumbar pain, refractory to analgesic treatment. Unlike the majority of reported cases, which involve patients who have undergone hysterectomy and partial oophorectomy, our case was presented with symptomatic spine metastasis. Successfully management, required lumbar spine fixation, followed by hysterectomy and oophorectomy, radiotherapy and long-term hormonal therapy.

The diagnosis of BML relies on meticulous medical history evaluation, (appropriate radiology) along with histopathologic and immunohistochemical analysis of metastatic lesions or primary tumor. To date, no standard treatment approach has been established. The clinical course of BML varies depending on the number and sites of metastases, emphasizing the need for an individualized (tailored) approach. Furthermore, our case underscores the importance of multidisciplinary collaboration and long-term follow-up in effectively managing BML patients.

REFERENCES

944428. PMid:35339186.


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