



Laparoscopic Hybrid Right Hepatectomy and Left Lobe Non-Anatomic Resection for Treatment of Bilateral Liver Metastasis from Uterine Leiomyosarcoma: Case Report and Review of the Literature

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Abstract

Background: Uterine leiomyosarcoma is a rare uterine neoplasm with an extremely aggressive malignancy associated with a poor overall prognosis. When only methacronic hepatic metastases are presented surgical resection may be offered.

Case Report: A 50-years old asymptomatic female patient who had undergone a posterior pelvic exenteration 3 years ago due ULMS, presented in follow-up examinations the appearance of exclusive liver metastases (after 1 year later from her follow-up). The resection of the lesions in single time was successfully proposed and performed by means of right hepatectomy and non-anatomical resection of the left lobe lesion. To date, two years after hepatic resection, she is alive without either symptoms or recurrence.

Conclusion: Although Uterine LMS are rare and aggressive tumors with generally poor prognosis, the surgical treatment of metastatic liver lesions can offer not only a better long-term survival, but also attain more progression free survival, such as we have observed in this case report. The overall management of these cases must always be individualized in order to achieve optimal treatment.

Keywords: Uterine leiomyosarcoma; Hepatic metastasis/surgery; Liver/Surgery; Hepatectomy

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Introduction

Uterine LMS tends to be an aggressive neoplasm, with only a 50% in 5-year survival rate with patients whose tumor is confined to the uterus, and about a third of them have already spread at the time of diagnosis. They tend to be often asymptomatic and diagnosed as an incidentally discovered abdominal mass. Surgical resection of the primary lesion is considered the standard therapy; however, there is no general consensus on the treatment for the liver metastasis of uterine leiomyosarcoma [1,2].

Although there is a high chance of lesions recurrence, small series have shown the benefit of liver metastasectomy, therefore a long-term survival and cure potential can be achievable on this scenario when free margins are attained [3]. Chemotherapy is usually offered to treat the metastasis; however, its advantages have not been well documented [4]. Present authors report a case of long-term survival (24 months) in a patient with Uterine LMS with multiples hepatic metastasis who was successfully submitted to a single time hybrid metastasectomy.

Case Presentation

A 50-years old asymptomatic female patient with a clinical history of 4 years ago had undergone a posterior pelvic exenteration with cystectomy, right ureterectomy and partial resection of the common external iliac artery and vascular reconstruction due a locally advanced moderate grade uterine leiomyosarcoma followed adjuvant treatment (pelvic radiotherapy and systemic treatment with chemotherapy). After one year from his surgery, in routine follow up was observed an appearance of three liver lesions in the both liver lobes. He presented at this time no extrahepatic disease at her staging (as tomography as PET-CT). These hepatic metastases presented the following location: one lesion (around 5 cm of diameter) in segments VI/VII, one lesion ((around 5 cm of diameter) in segments V/VIII and another small lesion (1 cm of diameter) superficially positioned

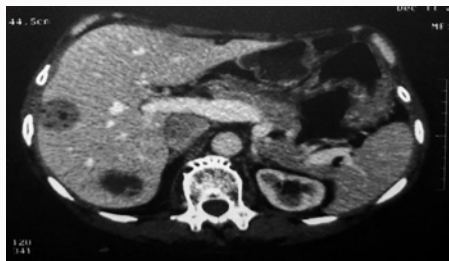


Figure 1: Preoperative Ct- hepatic metastases in both hepatic lobes.

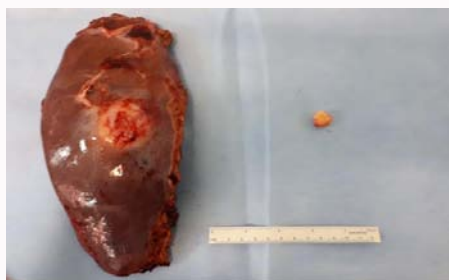


Figure 2: Surgical species- Right Hepatectomy with Non-anatomical resection of left lobe lesion.

on the segments II/III of the left lobe (Figure 1). At this time we have proposed and performed a single stage resection off the all lesions by means of a straightforward right hepatectomy and non-anatomical resection of the left lobe lesion (Figure 2).

Hybrid anterior approach was chosen because these right lobe lesions were bulky and to mobilize the right lobe would be dangerous with risk of vena cava injure. So, we performed all identification, dissection and ligation of the right hilar structures by classical extrahepatic access and anterior laparoscopic section of the liver parenchyma, and finally to mobilize all right lobe of the retrohepatic vena cava and perform the dissection and ligation of the right hepatic vein was used a small open right subcostal incision. She presented a good recovery without postoperative complications and she was discharged on the 5th postoperative day. A histological analysis showed mesenchymal tumor cells in right lobe lesions (surgical margins were free) and just fibrosis and necrosis in left lobe lesion. Immunohistochemical analysis confirmed uterine leiomyosarcoma origin.

To date, two years after hepatic resection, she is alive without either symptoms or clinical-radiologic recurrence (Figure 3). Additionally, she also presents an excellent quality of life.

Discussion

Leiomyosarcomas (LMS) are a heterogeneous group of smooth muscle neoplasm whose outcome has historically been confounded by the inclusion of gastrointestinal stromal tumors, and it can arise in the uterus, extremity, or other primary sites [5]. Uterine leiomyosarcoma is an extremely rare tumor, with an estimated incidence of approximately 1% of all uterine malignancies, although it represents around 70% of all uterine sarcomas. Women can be affected in variable ages, but the occurrence is more frequent in perimenopausal years like observed in present case. Symptoms are usually nonspecific, and not *easily distinguished* from other benign uterine conditions, which can often result in delay of the diagnosis [6]. They can attain great dimensions and invades loco-regional structures like observed



Figure 3: Late postoperative Ct (24 months after resection of hepatic metastases).

in this present case. This way, multivisceral becomes necessary to attain free margins aiming to ameliorate the final prognosis. Uterine LMS have a high mortality rate and propensity for hematogenous dissemination, frequently metastasizing to the lungs, abdominal cavity and retroperitoneum. However, isolated liver metastases like observed in the present case are very rare. The overall prognosis is dismal, mainly in locally advanced or metastatic disease, therefore is estimated only 10% to 15% of overall survival at 5 years for those with metastatic disease [7]. The prognosis factor includes mainly absence of residual tumor following primary surgery, tumor size and Mitotic Index (MI) above 10 mitosis per 10 HPF [8].

The standard therapy in primary uterine leiomyosarcoma is the complete surgical resection (total hysterectomy) with or without adjuvant therapy to follow. Surgery correctly performed is imperative and can be curative. Routine oophorectomy or lymph node dissection remains controversial, and do not appear to be associated with clinical advantages. Chemotherapy may be used with an adjuvant scenario or even for treating metastatic disease [9-11]. In metastatic tumors, especially isolated lung or liver lesions, retrospective studies have demonstrated better outcomes when there is a chance of surgical option regarding a possible complete resection, which may contribute to a prolonged survival [12-15]. Because of the lack of cases and aggressive behavior, resectable hepatic metastases are uncommon in patients with leiomyosarcoma, and constitute a highly select group [8,13,14].

In this present case, we could observe that total resection of the lesions with a free margins was the very important for final prognosis like has been described in the literature because it's high tumor recurrence potential [1-5,13,14]. Surgical cytoreduction has also been an important strategy in patients who present with metastatic uterine LMS at initial diagnosis with metastatic leiomyosarcoma, and may present survival advantage [15].

Adjuvant therapy in LMS after primary resection is still experimental. Many studies have been performed in order to evaluate the role of adjuvant chemotherapy and has not shown significantly survival improvement for patients with stage I or II disease [9,10]. Doxorubicin and ifosfamide were the most recommended between the chemotherapeutic options with the largest response rates, but currently new regimes with the combination of gemcitabine and docetaxel are being chosen as most active therapy for patients with advanced or recurrent uterine LMS [15]. Adjuvant radiotherapy is also undefined, and can be indicated in R1 and R2 resections in locally advanced tumors confined to the pelvis; Several studies have shown that it may reduce the risk of local recurrence in patients receiving pelvic radiation therapy, although such treatment has not been

proved to have advantage in women with early leiomyosarcomas, because pulmonary metastases are more prevalent than pelvic recurrences [11].

In same, although there is no consensus in metastatic uterine LMS treatment, many studies have shown the advantages of the surgical approach in selected cases, with minimal morbidity, prolonged time to first recurrence and possible cure.

Conclusion

Although Uterine LMS are rare and aggressive tumors with generally poor prognosis, the surgical treatment of metastatic liver lesions can offer not only a better long-term survival, but also attain more progression free survival, such as we have observed in this case report. The overall management of these cases must always be individualized in order to achieve optimal treatment.

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