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Brain Metastasis of Uterine Leiomyosarcoma: A Case Report and Review of the Literature

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Abstract

The metastases of uterine leiomyosarcoma to the brain are exceptional and such cases are seldomly reported in the literature. The diagnosis is based on CT and brains MRI findings, in association with a gynecological history of cancer. Their management still stays without guidelines, and the surgical total resection is known to be the only way to influence positively the prognosis and save patient lives. We report a rare case of a 46 years old woman who underwent a hysterectomy and bilateral salpingo-oophorectomy 5 years earlier and presented with the right hemiparesis and whose CT scan and MRI of the brain showed cerebral lesions related to brain location of uterine leiomyosarcoma. The patient underwent surgery for gross total tumor resection, followed by adjuvant radiotherapy, and was doing well after surgery. Three months later she was admitted for recurrences and died after two months of palliative care.

Keywords

Uterine Leiomyosarcoma, Brain Metastasis, Case Report

1. Introduction

Brain metastases frequently occur secondary to poorly controlled primary lung, breast, melanoma, colorectal and renal tumors [1]. These tumors are the most common sources of brain metastases in adults. Uterine leiomyosarcoma (ULMS) is a rare entity among malignant gynecologic tumors with a very poor prognosis

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and the highest prevalence in the pre-and menopause period. Only early-stage tumors have an acceptable prognosis, provided the patient has been treated without injuring the uterus [2]. In contrast to endometrial carcinomas which most commonly metastasize to the lymph nodes, ULMS has a high propensity for hematogenous spread most commonly to the lungs [3]. We report a very rare case of a 46 years old woman with a gynecologic history of uterine leiomyosarcoma operated and treated by adjuvant radiotherapy five years earlier, presented with agressive brain tumor with extension to the lungs and who underwent surgery, followed by adjuvant radiotherapy for metastases of this tumor to the brain with a good immediate outcome, and died a few months later because of the brain tumor recurrences.

2. Case Presentation

A 46-year-old woman with a past medical history of uterine myomas revealed by heavy menstrual bleeding (menorrhagia). Exploratory surgery for a simple myomectomy was done for sample collection and the diagnosis of uterine leiomyosarcoma was confirmed by the histopathology five (5) years ago. The patient underwent an abdominal hysterectomy and bilateral salpingo-oophorectomy, followed by adjuvant radiotherapy with a good outcome. Then, there was no evidence of any other lesions. The patient denied any history of arterial hypertension or diabetes. Her current medical history started with sudden onset of right hemiplegia, associated with moderate headaches without signs of intracranial hypertension neither comitial crisis. The patient was alert, in good general status without a fever.

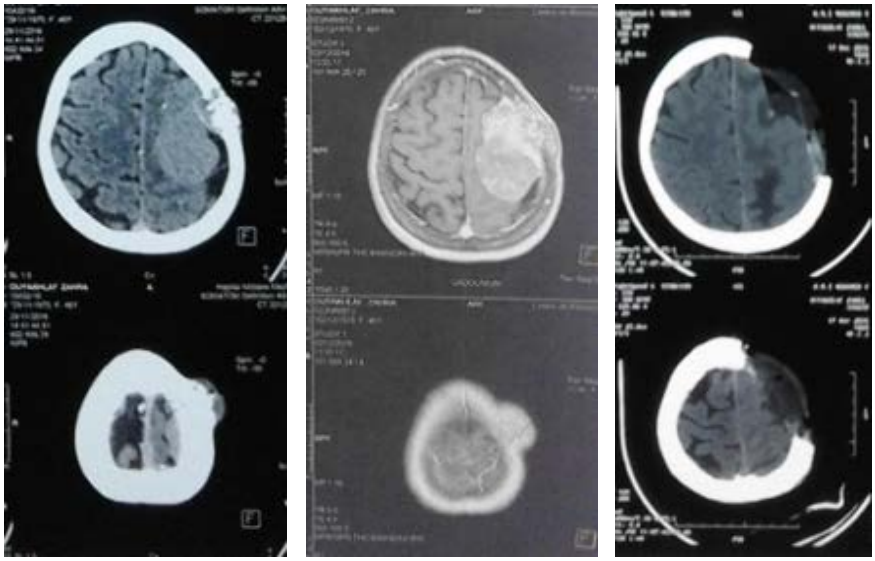
2.1. Investigation

A brain CT-Scan showed two cerebral lesions in the left parieto-occipital region (**Figure 1(a)**), confirmed by brain MRI (**Figure 1(b)**) showing a heterogenous left parietal very aggressive lesion with bone extension and another small parieto-occipital one. The most probable radiological diagnosis was the brain metastases in relation to her past medical history. The Positron Emission Tomography (PET) scan revealed metastatic lesions of both lungs. Laboratory findings were normal.

2.2. Management and Outcome

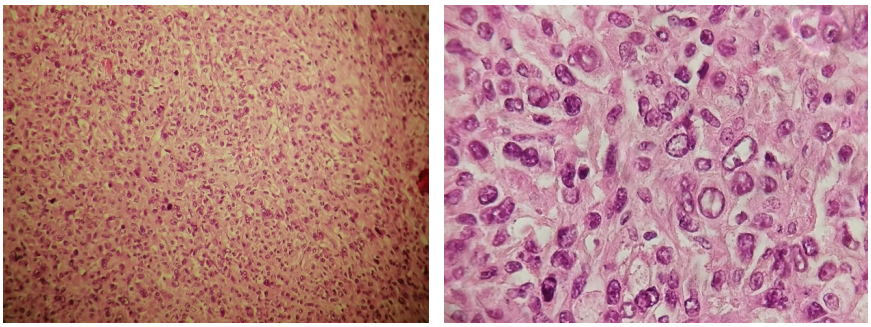
The patient underwent surgery, and gross total resection of the largest and very aggressive lesion was done through a classical parietal approach with bony resection (**Figure 1(c)**). Immediate postoperative recovery of the motor palsy was observed. Histopathology analysis of the tissue sample confirmed the diagnosis of metastasis of the uterine leiomyosarcoma to the brain. Histologically, multiple serial sections showed a highly pleomorphic tumor arranged in irregular fascicles (**Figure 2(a)**). The tumor cells were spindle shaped and had characteristically elongated and blunt-ended cigar-shaped nuclei with eosinophilic cytop-

lasm. Tumor cells showed marked nuclear pleomorphism and hyperchromatism with prominent nucleoli, showing numerous mitotic figures (**Figure 2(b)**), including frequent atypical forms. The tumor was massively infiltrated into dura mater, cranial bone and surrounding soft tissue. Immunohistochemical staining revealed that tumor cells were strongly positive for h-caldesmons (**Figure 3(a)**), smooth muscle actin (**Figure 3(b)**), and vimentin (**Figure 3(c)**), but negative for pancytokeratin, epithelial membrane antigen, desmin, S-100, Melan A, CD34, CD30, and CD10. Thus, the diagnosis of a brain metastasis from uterine leiomyosarcoma was made.



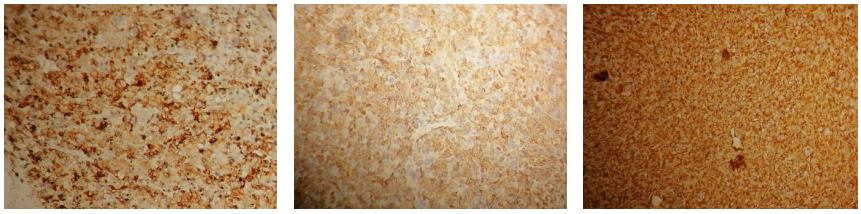
(a) (b) (c)

Figure 1. (a): Pre-operative brain CT-scan showing an agressive left parietal lesion spontaneously hyperdense. (b): Brain MRI with the same lesion, enhanced heterogeneously after gadolinium injection with an infiltration of the bone and the subcutaneous cell tissue. (c): Post-operative CT-scan showing the large and total removal of the tumor and the loco-regional invasion of the bone.



(a) (b)

Figure 2. Photomicrographs showing spindle and irregular-shaped cell tumor proliferation forming irregular fascicles ((a): hematoxylin and eosin stain; original magnification ×200) exhibiting pleomorphic and bizarre tumor cells with foamy cytoplasm and marked atypia, including numerous mitotic figures ((b): hematoxylin and eosin stain; original magnification ×400).



(a) (b) (c)

Figure 3. Photomicrographs showing strong immunostain to H-caldesmon ((a): Immunochemistry stain, original magnification ×200), Smooth muscle actin (SMA) ((b): IHC stain, original magnification ×200), and vimentin ((c): Immunochemistry stain, original magnification ×200).

This successful surgery is followed by adjuvant radiotherapy and the patient was doing well. Three months later she presented with the same neurologic symptoms and the brain MRI revealed recurrences of the metastasis. The patient's general status was not favorable for surgery and she was then transferred to the palliative care department where she died after two months of palliative care.

3. Discussion

The diagnosis of ULMS often occurs in retrospect after surgical resection of a presumed benign uterine neoplasm and therefore, patients often do not undergo preoperative staging workup [3]. Sandruck and colleagues [4] published a case of a 39-year-old patient who underwent a myomectomy 18 months before the diagnosis of ULMS. Our patient was first operated on for probable uterine myomas. A simple tumorectomy was performed and pathology showed a uterine leiomyosarcoma. In a second time, an abdominal hysterectomy and bilateral salpingo ovariectomy were done. Unfortunately, control of the primary disease in uterine leiomyosarcoma is poor. The gold standard for management is the total abdominal hysterectomy. However, these tumors cannot be considered a local disease even in the early stages because of the local aggressiveness and propensity for early lymphatic and hematogenous [5] [6]. Currently, neither chemotherapy nor radiation therapy has demonstrated clear effectiveness as adjuvant treatment [5] [7]. Uterine leiomyosarcoma is a rare malignancy of mesenchymal origin. It usually develops in the fifth decade of life. Nevertheless, authors as Prussia and colleagues [8], Sandruck and colleagues [4] reported respectively a case of a 36 and 39 years old woman with uterine leiomyosarcoma and cephalic metastasis. There are no specific clinical signs and symptoms found in these patients. Distant metastases to the central nervous system are usually associated with the widespread dissemination of the disease. In fact, between 20% and 40% of patients with systemic cancer develop brain metastases [9]. Our patient was 46 years old, close to the second half of life, and presents similar dissemination characteristics of the disease related to the central nervous system associated with the widespread dissemination to other organs. Of note, pulmonary metastases appear to be the most frequent immediate prior site of disease for patients

with sarcoma developing brain metastases [4] [10]. Other studies cite cases of soft tissue sarcoma or leiomyosarcoma with non-uterine primaries metastatic to the brain [10] [11] [12]. The literature suggests that the treatment of choice for brain metastases from uterine leiomyosarcomas is neurosurgical resection of all known lesions, single or multiple, followed by postoperative whole-brain irradiation as adjuvant therapy, even if radiation therapy appears to improve local control without any significant impact on survival because most recurrences involve distant sites [9] [13] [14]. The presence of concurrent lung metastases is not a contraindication to surgery [9] [13]. Our management was the same as that proposed by the literature. Some authors as Wronski *et al.* [14] and Bindal *et al.* [15] found a survival linked to the quality of surgical resection estimated between 6 and 14 months. The prognosis remains very poor.

4. Conclusion

Uterine leiomyosarcoma is a very rare entity in gynecologic malignant disease. Often metastasized in the lung, the brain's secondary location is exceptional. The surgery of metastases is the rule and the adjuvant treatment still being uncodified. The uterine leiomyosarcoma is very aggressive in general, with a high potential of recurrence despite the quality of surgical resection locally and for the brain metastases, with a poor prognosis.

Author Contributions

Fernand Nathan Imoumby: Conceptualization, Writing an original draft, & editing. **Yao Christian Hugues Dokponou:** Writing, review & editing. **Loukou Franck Kouakou:** Writing, review & editing. **Hajar El Agouri:** Writing, review. **Abad Cherif El Asri:** Review & editing. **Miloud Gazzaz:** Supervision, review, & Validation.

Ethics and Reporting Guidelines

Informed consent and verbal permission were obtained from the family of the patient prior to the submission of this article. Also, this article respects both the Consensus-based Clinical Case Reporting Guideline and the Recommendations for the Conducting, Reporting, Editing, and Publication of Scholarly Work in Medical Journals [16] [17].

Conflicts of Interest

The authors declare not having any conflict of interest in this case report and there are no financial resources.

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