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Leiomyosarcoma of the uterus: a clinicopathological report of two cases and review of literature.

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Summary

Leiomyosarcomas of the uterus are highly malignant tumours that are rare and represent slightly over 1% of all uterine malignancies. The clinical presentation is fairly constant and includes abnormal vaginal bleeding, pains or both. Only two cases (0.6%) out of 337 uterine malignancies seen over a period of 15 years in Ile-Ife had histological diagnosis of leiomyosarcoma. Both patients were postmenopausal, aged 49 years and presented in advanced disease stage. Two months after surgery, one of the patients died from disease progression while the other defaulted while on chemotherapy. Leiomyosarcoma of the uterus though rare in our environment, when it occurs, presents with advanced stage of the disease. Poor compliance coupled with high cost of treatment portends poor prognosis thus making it rapidly fatal.

Keywords: Leiomyosarcoma, uterus, poor prognosis.

Résumé

Les léiomyosarcomes de l'utérus sont les tumeurs les plus contagieuses qui sont rare et représentent environ 1 % de tous les cas de tumeurs utérin. Les symptômes cliniques sont constants et inclus d'hémorragie vaginal anormale et des douleurs. Seulement 2 cas (0.6%) sur les 337 cas de tumeurs utérin étaient examinés histologiquement durant une période de 15 ans a Ilé Ifé. Les patients étaient à leur post ménopause, d'age moyenne de 39 ans et représentaient une étape avancée de la maladie. Deux mois après la chirurgie, un des patients mourrait de la progression de la maladie alors que les autres sous traitement ne complétaient pas leur suivi. La léiomyosarcomes de l utérus bien que rare dans notre environnement parait à une étape avancée de la maladie. Le faible de conformité des patients joint au taux élevé des médicaments conduit à une prognosie faible rendant cette maladie fatale.

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Introduction

Leiomyosarcomas are aggressive malignant tumours of smooth muscle origin and can affect virtually any part of the body where smooth muscle is present. They represent 25% of all uterine sarcomas and slightly over 1% of all uterine malignancies [1].

The majority of patients with leiomyosarcoma of the uterus present above 40 years of age and the usual complaint are abnormal vaginal bleeding, gradually increasing chronic pelvic pain or both [2]. Leiomyosarcomas may show some similarity to leiomyomas but are usually less-well circumscribed, soft and may show areas of haemorrhages or necrosis [3].

Microscopically, leiomyosarcoma is composed of hypercellular oval to spindle shaped cells arranged in interlacing bundles with large hyperchromatic, irregular nuclei and high mitotic count [4,5]. An average of 5 or more mitotic figures per 10 high power fields in the most active area is agreed among most authors as the distinguishing feature of leiomyosarcoma [6]. In addition to the morphology, the ultrastructural and immunohistochemical features of smooth muscle cells, actin and myosin are consistently present in the cytoplasm of the tumour cells [7]. We present two case reports of leiomyosarcoma of the uterus with the aim of correlating the clinical presentations and histopathological findings with the prognosis.

Case reports

Case 1

A 49 year old P₄⁺¹, 4 alive postmenopausal woman presented with progressive abdominal swelling of two years, bleeding per vagina of three weeks and lower abdominal pain of a day duration. The main finding on clinical examination was an abdomino-pelvic mass measuring 20 weeks size that moved with the uterus. The external genitalia, the vagina and the cervix were grossly normal. Abdomino-pelvic ultrasound scan findings were suggestive of uterine fibroid. A diagnosis of uterine fibroid to rule out endometrial carcinoma

was made and the patient was prepared for total abdominal hysterectomy and bilateral salpingooophorectomy.

Post-surgery, she was to be commenced on adjuvant chemotherapy (cis-platinum and cyclophosphamide) following histopathological diagnosis of leiomyosarcoma. The immunohistochemistry showed smooth muscle antigen positivity (SMA+) which confirmed leiomyosarcoma. The International Federation of Gynaecologists and Obstetricians (FIGO) staging was IIB. However, she did not report for readmission until ten weeks later. A repeat abdomino-pelvic ultrasound scan and chest x-ray showed metastatic lesions in the lungs, liver and bowels. Approximately 5 months post-surgery, she had completed 3 courses of chemotherapy with slight clinical improvement before she defaulted.

Case 2

A 49 year old postmenopausal P₅⁺¹ 5 alive, presented with a year history of progressive abdominal swelling; weight loss and early satiety of six months duration. Clinical examination revealed a middle aged woman, pale with massive ascites. Abdominal ultrasound revealed a partly cystic and solid right adnexae mass with a suggestion of ovarian malignancy. A clinical diagnosis of ovarian malignancy was made, and she was admitted for exploratory laparatomy. At surgery, 1.5 litres of serous fluid was drained, an encapsulated huge mass, partially solid and cystic measuring 40cm x 40cm was attached to the fundus of the uterus.

A total abdominal hysterectomy with bilateral salpingo-oophorectomy and partial omentectomy was done and she was discharged a week after surgery. The histopathology report was leiomyosarcoma of the uterus with metastasis to the omentum; immunohistochemistry was positive for SMA, caldesmon and desmin. The FIGO stage was IVB. She was to commence adjuvant chemotherapy but her clinical condition remained unstable. She developed abdominal distention and vomiting, a repeat ultrasound scan done revealed multiple abdominal masses with ascites, and her condition deteriorated further and she died 10 weeks post-surgery.

Discussion

The two cases seen over a 15 year period indicates that leiomyosarcoma of the uterus is uncommon in our environment. This represents 0.6% of the total 337 uterine malignancies seen over a 15 year period. The mean age of 49 years observed in this review is in agreement with the findings from other studies [1,8].

The two patients were postmenopausal and the predominant presenting symptoms include abnormal vaginal bleeding, abdominal mass and discomfort. These symptoms are similar to what has been documented in previous studies, the only difference being the late presentation observed in our patients [2,8].

Definitive treatment for uterine leiomyosarcoma is surgery with adjuvant chemotherapy and or radiotherapy. In this series, one of our patients

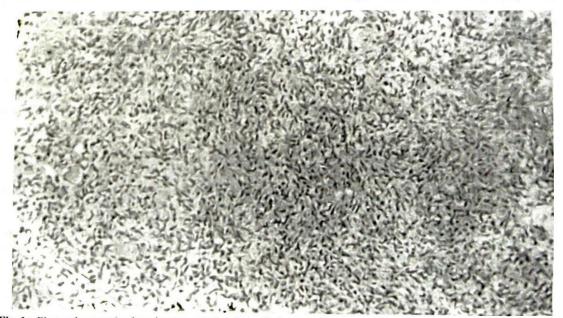


Fig. 1: Photomicrograph of uterine tumour showing spindle shaped tumour cells with nuclear atypia H&E X144

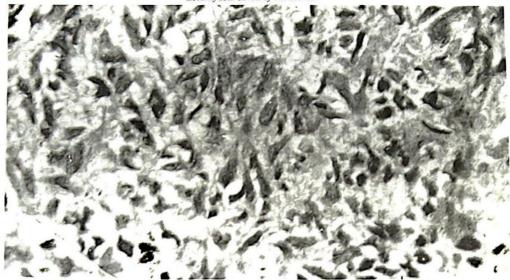


Fig. 2: Higher magnification of figure 1. Note the pleomorphism, hyperchromatism and nuclear atypia. H&EX576

had surgery plus adjuvant chemotherapy while the other had surgery alone. The histology from the two cases fulfilled the criteria for the diagnosis of leiomyosarcoma, and they showed spindle shaped tumour cells arranged in fascicles with nuclear atypia and abnormal mitosis [9] [Fig.1 and 2]. There were more than 10 mitotic figures per 10 high power fields. Adjuvant chemotherapy administered to one of the patients does not seem to have much effect on prognosis as this patient presented with tumour metastases within 2 months of surgery. There is presently no standard chemotherapy protocol for leiomyosarcoma because of their rarity, lack of large clinical trials and poor response of individual patients to chemotherapy, however modest activity has been reported with cis-platinum which this patient had [10]. The choice of chemotherapy in our environment is also largely determined by availability and cost.

Advanced stage of disease and tumour necrosis found in both cases are poor prognostic factors that have important bearings on the patients' survival [11,12]. It is also important to note that both cases presented had tumours in the uterine corpus and cervix and this has also been shown to be a poor prognostic index regardless of the mode of therapy [13].

Conclusion

Leiomyosarcoma of the uterus is rare in our environment; advanced stage disease, high cost of treatment and high default rate in a depressed economy like ours will be major limitations for some years to come in the adequate management of this category of patients.

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