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Case Report

The shoulder's hidden alarm: metastatic uterine leiomyosarcoma in disguise

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ABSTRACT

Uterine Leiomyosarcoma is a rare but aggressive soft tissue tumour arising from smooth muscle fibres of the uterus. Leiomyosarcoma is one of the most common types of smooth muscle-derived malignancies in uterus and it is an aggressive malignancy. We are reporting a unique case of a 54-year-old woman who presented with uterine leiomyosarcoma along with humeral metastasis manifesting as a right shoulder swelling, an uncommon site of metastasis. This case report emphasizes the rarity of this presentation and underscores the importance of thorough clinical and radiological evaluations in atypical presentations. This patient also had a significant symptom relief and improvement in quality of life following palliative care and chemotherapy.

Keywords: Uterine leiomyosarcoma, Metastasis, Palliative care, Atypical

INTRODUCTION

Leiomyosarcoma primarily originates from smooth muscle or mesenchymal cells and often involves the retroperitoneum, uterus, or extremities.[1,2] Leiomyosarcoma is the most common type of uterine sarcoma accounting for approximately 30% of all uterine sarcomas. Incidence of uterine leiomyosarcoma is highest in the perimenopausal age group and it carries a poor prognosis especially in patients with advanced stage and higher grade.[3] Surgery is the cornerstone of curative therapy, with chemotherapy being used in adjuvant setting for advanced disease. However, for inoperable and metastatic disease, palliative chemotherapy and radiotherapy are the preferred treatments. Its metastasis pattern typically includes the lungs, liver, and kidneys, metastasis to brain and bones is rare.[4,5] Primary presentation with metastasis to humerus, as seen in this case report, is very rare.

CASE REPORT

A 54-year-old postmenopausal woman (P1L1) presented with a progressively enlarging swelling in the right shoulder from six months. With gradual increase in the size of the lesion, the movements in the right shoulder were severely restricted. The swelling was associated with mild pain earlier but at the time of presentation, the pain was severe and her sleep was disturbed as a result of it. She also complained of bleeding per vagina and foul-smelling white discharge per vagina from last three months. There was no history of fever, weight loss, loss of appetite, or early satiety. She had no history of diabetes, hypertension, tuberculosis, or previous malignancies. Patient had attained menopause two years prior, with no history of use of oral contraceptives, hormone therapy or radiotherapy ever. There was no history of malignancies in the first and second degree relatives in the family.

On examination patient had pallor with a 6.5×8.2 cm firm-to-hard, tender swelling in the right shoulder (Figure 1) involving the head and proximal shaft of humerus, associated with localized edema. The movements of shoulder joint in all axis were severely restricted. Patient couldn't bear the weight of the right arm hence she supported and carried the right arm with her left arm. On per abdominal examination, a pelvic mass was palpated, consistent with the size of an 18-week gravid uterus. No other organomegaly or tenderness was present on per abdominal examination. On gynaecological examination, cervix was atrophic and pulled up. No lesion was visualised however bleeding PV was noted. On Per-rectal examination, the uterus was found to be hard and bulky.

Figure 1: Swelling in the right shoulder involving Head and the proximal head of humerus.

X ray of shoulder showed expansile lesions completely involving head and proximal shaft of humerus (Figure 2). CT scan of the right shoulder was performed which showed a neoplastic mass of size 8.5×9.4×12 cm involving and completely destroying the head and proximal shaft of right humerus (Figure 3). 2.2×4.9×3.2 cm expansile lesion was noted in posterior aspect of 7th rib in PET CT (Figure 6). No evidence of pulmonary or hepatic metastases was found (Figure 4). MRI Pelvis showed a well defined multilobulated lesion (16.4×14.5×10.1 cm) originating from the uterus, involving the cervix (Figure 5A, B). PET-CT showed a hypermetabolic uterine mass with metastases to the right humerus, 7th rib, and ilium (Figure 6). Histopathology of uterine biopsy revealed spindle cells tumor with necrosis. Biopsy from shoulder swelling showed spindle cell sarcoma with Immunohistochemistry positive for desmin and negative for S-100, SMA, h-caldesmin, CD34, BCl2, myogenin and CK, favouring metastatic leiomyosarcoma.

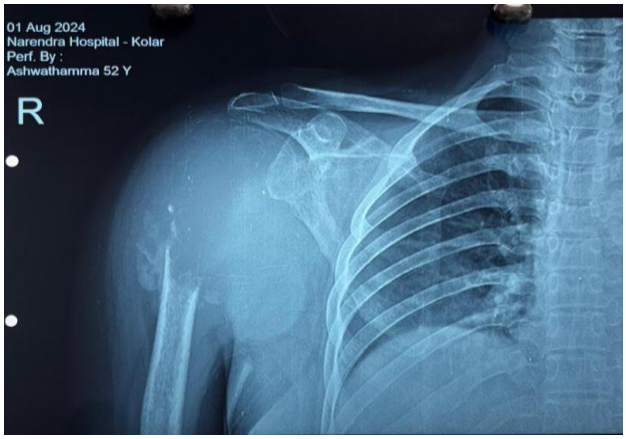

Figure 2: X ray shoulder ap view showing expansile lesion involving head and proximal shaft of shoulder.

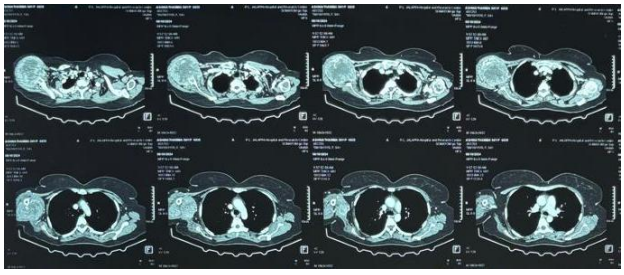

Figure 3: CT thorax showing the expansile lesion of humerus.

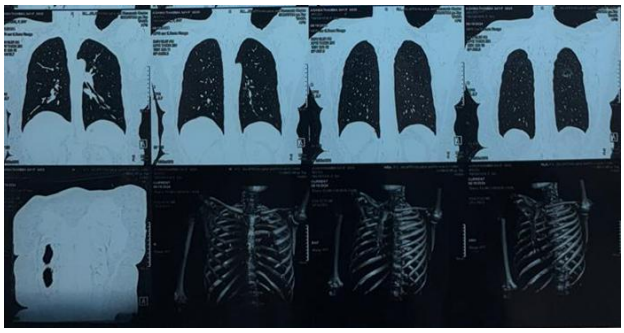

Figure 4: CT thorax showing no involvement of lung or liver.

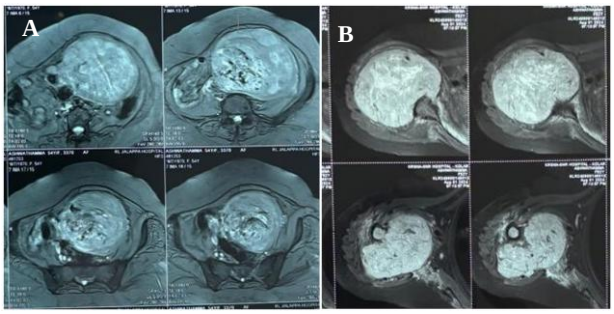

Figure 5: (A) MRI scan of pelvis showing the lesion arising from the uterus and (B) MRI scan of shoulder showing the expansile lesion.

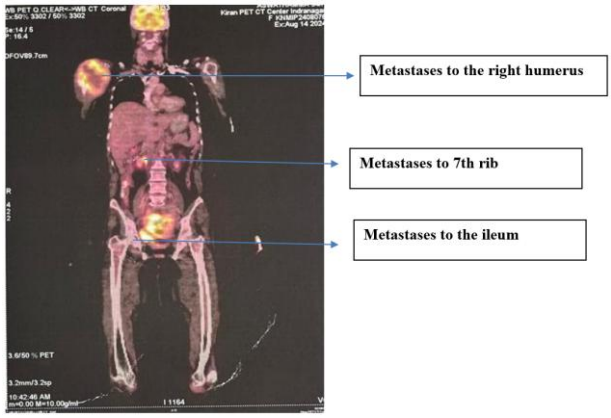

Figure 6: PET CT showing hypermetabolic uterine mass with metastases to the right humerus, 7th rib, and ilium.

Based on history and examination findings along with these investigations, patient was diagnosed with uterine leiomyosarcoma stage IVB with metastatic involvement of the right humerus. After correction of anaemia with blood transfusions and adequate symptomatic relief, patient was discussed in tumour board and planned for palliative chemotherapy. Two cycles of Adriamycin and Ifosfamide regimen, palliative chemotherapy have been given. After two cycles, significant clinical improvement was noted with >50% reduction in shoulder swelling along with significant reduction in pain and improvement in shoulder mobility. Patient also had complete relief from bleeding and white discharge per vagina. Arm Sling was provided for immobilisation and supporting the right shoulder, this minimised movement of arm and discomfort. These treatments led to a significant improvement in the quality of life of the patient.

DISCUSSION

Leiomyosarcoma is a rare and aggressive type of soft tissue sarcoma with an incidence of 1 per 100,000 population annually.[6] Most common site of leiomyosarcoma is uterus.[7] Uterine leiomyosarcoma is most common among women in 5th and 6th decades of life. Previous history of exposure of pelvis to radiotherapy especially at younger age, tamoxifen use for management of breast cancer and genetic conditions such as retinoblastoma and Li-Fraumeni syndrome are recognised as risk factors for development of uterine leiomyosarcoma.[8,9] Patients usually present with abnormal vaginal bleeding, pelvic mass, pelvic and abdominal pain.[10] Stage, grade and mitotic index are proposed to be prognostic factors for uterine leiomyosarcomas. However, regardless of stage and grade, histology carries a poor prognosis.

Stage I and II disease are treated with surgery alone. Stage III uterine leiomyosarcoma is treated with surgery followed by adjuvant chemotherapy. Uterine leiomyosarcoma has propensity to spread via hematogenous route and very low propensity to spread via lymphatics hence lymphadenectomy is not done routinely when patient has no lymphnodes visualised on imaging. Adjuvant radiation is used only in advanced disease or when the tumor resection is incomplete. Stage IV uterine leiomyosarcoma involving metastatic and very advanced tumours are treated with palliative chemotherapy and radiotherapy.[11] The five-year survival rate is 50-55% for stage I and 8-12% for stage II-IV.

Nearly 1/3rd of all patients with uterine leiomyosarcoma present upfront with distant metastasis. Metastasis are usually seen in the lungs, liver, and pelvis, metastasis to brain and bones are not commonly seen.[6] Typically, sarcomas metastasize hematogenously to lung and liver however presentation with only bone metastasis is rare. This patient had only skeletal metastasis and also this patient's presentation with a metastasis in humeral head highlights an unusual metastatic pattern.[12] This case report underscores the possibility of atypical presentations where thorough clinical evaluation and further investigations including MRI /CT scan of metastatic site, PET-CT and biopsy along with IHC from the metastatic sites may be required to accurately diagnose and plan further treatment.

Palliative chemotherapy planned for this patient was Adriamycin and Ifosfamide. Patient is tolerating this chemotherapy well. Patient had a clinically good response to the chemotherapy and there was a significant improvement in quality of life. Chemotherapy will be further continued for this patient. Other chemotherapeutic agents effective in this condition are gemcitabine and docetaxel.[13] Palliative radiotherapy to bone metastasis is useful for pain relief and prevention of pathological fractures and spinal cord compression when vertebral metastasis are present. Palliative radiotherapy to brain is effective in symptomatic relief and prevention of convulsions. Steriotactic radiotherapy is useful in management of metastasis at various sites. Uterine leiomyosarcoma are known to express Estrogen and Progesterone receptors with less than 10% objective response rate to hormone therapy.[14] Targeted therapies such as pazopanib and immunotherapy drugs namely Pembrolizumab and nivolumab are promising newer drugs being investigated in clinical trials.

CONCLUSION

This case report highlights a rare presentation of metastatic uterine leiomyosarcoma with only bone metastasis, emphasizing the need for vigilance in identifying and managing atypical metastatic patterns. Cancer chemotherapy and palliative care can significantly improve the quality of life even in advanced cases.

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