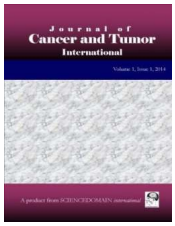

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Granulocytic Sarcoma of Ovary Associated with Chronic Myelogenous Leukaemia Mimicking Carcinoma: A Rare Case Report

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Authors' contributions

This work was carried out in collaboration between all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Granulocytic sarcoma is a rare haematological neoplasm due to extramedullary leukemic deposits. Granulocytic Sarcoma of ovary is even rare associated with chronic myeloid leukaemia and responds to imatinib treatment. A strong vigilance is required. Here we report a case of granulocytic sarcoma of ovary in a 31 year old female with primary infertility being provisionally diagnosed as

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ovarian tumour. Later on, after complete haematological investigation and fine needle aspiration cytology of ovarian tumour, the case is found to be of granulocytic sarcoma associated with chronic myeloid leukaemia. The aim of documenting this case report is to establish the possibility of granulocytic sarcoma in unusual site which was initially thought to be of nonhematological malignancy and proved by tissue diagnosis to be of haematological malignancy.

Keywords: Chronic myeloid leukaemia; extramedullary; fine needle aspiration cytology; granulocytic sarcoma.

1. INTRODUCTION

Granulocytic sarcoma is an extramedullary neoplasm frequently seen in acute and chronic leukaemia and myeloproliferative disorders, and is composed of primitive granulocytic cells [1]. It is a rare occurrence with an estimated incidence of 0.7 per one million in children and 2 per one million in adults [2]. It occurs in 3-9% of patients with acute and chronic myeloid leukaemia (CML) [3]. Bones, perineural tissue, lymph nodes, skin, gum and almost every site of the body are possible sites for granulocytic sarcoma. But granulocytic sarcoma of ovary is a rare entity. We hereby report a case of granulocytic sarcoma of ovary in a 31-year old female of CML who was mimicking ovarian tumour due to its rarity and diagnostic dilemma.

2. CASE REPORT

A 31 year old female patient presented with lower abdominal pain and dyspepsia for 2 weeks. She had history of hypomenorrhea and primary infertility. On clinical evaluation, she had pallor and spleenomegaly. Clinical evaluation of other systems did not reveal any abnormality. USG of abdomen and pelvis revealed spleenomegaly and a right hypoechoic adnexal mass (?Multicystic enlarged ovaries). USG Doppler also showed right ovarian enlargement with predominant solid component and low resistance flow in ovarian artery, suspicious of neoplastic lesion of right ovary. Her serum CA125 was raised (56.19 units/ml). Based on imaging and biochemical parameters patient was provisionally diagnosed as carcinoma of right ovary. During routine haematological study, incidentally peripheral smear report revealed a total WBC count of 1.4 lakhs with premature cells (Myeloblasts 2%, Myelocytes 15%, Metamyelocytes 13%), suggestive of CML and diagnosis was further straightened with LAP (leukocyte alkaline phosphatise) score which was 3 (normal range being 20-150). With further evaluation of ovarian status with CT scan abdomen and pelvis, patient was found to have right adnexal lesion (5×3.6 cm) with spleenomegaly (Fig.1) and USG guided fine needle aspiration cytology (FNAC) of right ovarian solid mass lesion revealed clusters of benign mesothelial cells and blood cells comprising of myelocytes, metamyelocytes, occasional promyelocyte, normoblasts, basophils and neutrophils (Fig. 2). Further cytogenetic analysis showed presence of Philadelphia chromosome confirming of CML. Hence, the case was diagnosed as CML with granulocytic sarcoma involving ovary and started with Imatinib 400mg per day. Response evaluation after 6 months of imatinib treatment revealed complete haematological response and major molecular response. Radiologically, CT scan of abdomen and pelvis revealed disappearance of right adnexal mass with normal spleen (Fig. 3). Now, the patient is on imatinib treatment and attended complete molecular response.

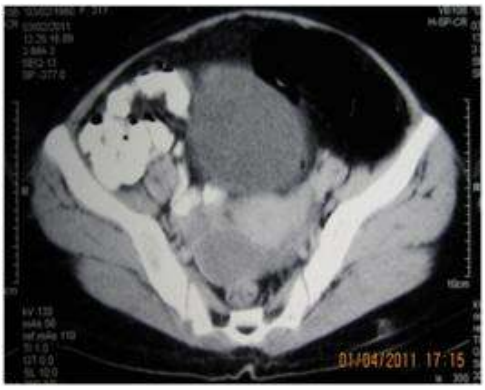

Fig. 1. CT scan of abdomen and pelvis showing heterogeneously hypodense mass lesion in right adnexa

3. DISCUSSION

Granulocytic sarcoma is a rare extramedullary tumour consisting of premature granulocyte precursor cells. Myeloblasts and myeloid precursor cells are present in varying degree of differentiations. They arises de novo, or may arise in association with other hematologic disorders such as AML (acute myeloid

Table 1. Showing similarities and difference between granulocytic sarcoma and epithelial carcinma

Similarities between granulocytic sarcoma and epithelial carcinoma	Difference between granulocytic sarcoma and epithelial carcinoma		
	Points to differentiate	**Granulocytic sarcoma**	**Epithelial carcinoma**
- Clinical presentation of abdominal pain and swelling - Radiological findings of solid ovarian mass - CA-125 may or may not rise in both the cases - Stage of presentation in both the cases may be early or advanced - Prognosis in granulocytic sarcoma is unknown and difficult to compare with epithelial carcinoma	Stage at presentation	unknown	Majorities advanced
	Serum CA-125	May or may not raised	Commonly raised
	Premature granulocytic precursor cells in ovarian tumour	Present	Absent
	Association of AML, CML, other myloproliferative disorders	Present	Absent
	Myloperoxidase	Present [7]	Absent
	CD45	Present [7]	Absent
	CD43	Present [7]	Absent
	Treatment with chemotherapy regimen used in leukaemia	Good response	No role
	Prognosis	Unknown	Known

leukaemia), myelodisplastic syndrome or myeloproliferative disorders. Association of granulocytic sarcoma with CML is extremely rare [4]. Granulocytic sarcoma can arise anywhere in body; it frequently involves perineural tissues, bones, spines, lymph nodes, skin and gums [5,6]. However granulocytic sarcoma in female genital tract is uncommon. Ovarian granulocytic sarcoma is a rare entity in daily practice leading to under diagnosis of the case [5,7]. Primary ovarian presentation of granulocytic sarcoma is very rare [3,8]. Granulocytic sarcoma can occur in female genital tract and may be the first clinically significant manifestation of a haematological malignancy [9]. It is further rarer to diagnose granulocytic sarcoma by FNAC [6,10-12]. This case is reported because of its uncommon location and dilemmatic way of presentation where provisional diagnosis was thought of as ovarian tumour. In this case, the aspirate obtained from ovary (USG-guided) is of paramount importance because it is showing evidence of myeloid differentiation. It arises high index of suspicion of granulocytic sarcoma because our patient presented with infertility and lower abdominal pain as primary symptom and establishment of haematological malignancy i.e. CML. Besides, after 6 months of Imatinib treatment, CT evaluation of abdomen and pelvis was showing complete disappearance of ovarian tumour which indirectly but strongly supports the ovary being presented as granulocytic sarcoma in CML.

In general, granulocytic sarcoma is not a differential diagnosis for ovarian tumour due to paucity of occurrence [3]. Granulocytic sarcoma can occur in ovary even though rare and may mimic ovarian tumour as in the present case. The similarities and difference between granulocytic sarcoma and epithelial carcinoma of ovary is highlighted in Table 1 (above).

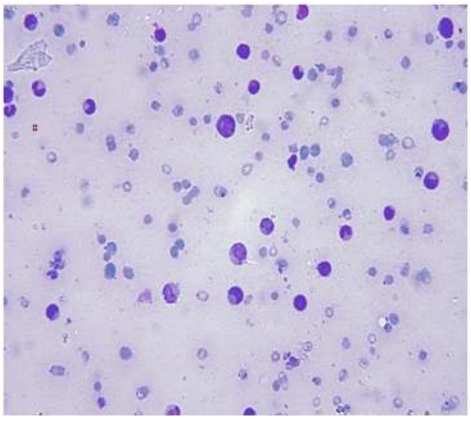

Fig. 2. (Diff quik stain -100X) showing granulocytic precursor cells like promyelocytes, myelocytes, metamyelocytes and blast cells

The present case is interesting due to finding as ovarian mass with simultaneous finding of CML. CML can be associated with granulocytic sarcoma. Most frequently, the tumour presents later in the natural history of CML as the disease progresses. Patients in relapse after chemotherapy or stem cell transplantation can also present with granulocytic sarcoma [13]. Granulocytic sarcoma that develops with chronic phase CML confers a worse prognosis i.e. higher risk of rapid blastic marrow transformation [13]. Granulocytic sarcoma associated with CML responds to imatinib treatment which is evident in the present case in which there is disappearance of ovarian pathology after 6 months course of Imatinib treatment. Therefore, early diagnosis is required to manage the case and to improve survival.

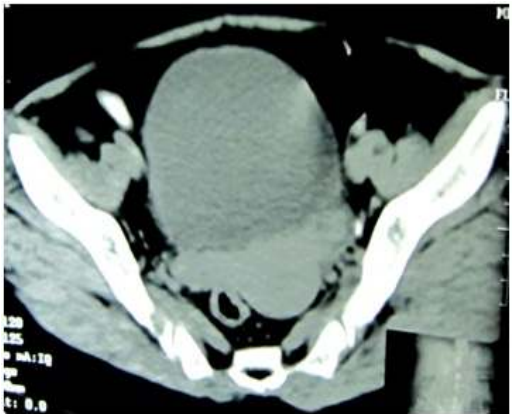

Fig. 3. CT scan of abdomen and pelvis showing disappearance of the ovarian lesion

4. CONCLUSION

Granulocytic sarcomas are rare haematological neoplasms due to extramedullary leukemic deposits, and primary granulocytic sarcoma of ovary even further rare. Granulocytic sarcoma associated with CML responds well to imatinib treatment. Hence a strong vigilance is required to suspect ovarian involvement in CML as granulocytic sarcoma. Patient having CML with ovarian involvement should be evaluated to rule out the possibility of granulocytic sarcoma.

CONSENT

It is not applicable.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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