Case Report

Burkitt's lymphoma in pregnancy: Unusual presentation of a rare case

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ABSTRACT

The association of pregnancy with cancer is rare. This case shows pregnancy exhibiting an inhibitory effect on Burkitt's lymphoma (BL) as well as demonstrates the extremely aggressive nature of the disease.

Key words: Burkitt's lymphoma, ovary, preganacy

INTRODUCTION

The association of cancer with pregnancy is rare (1 in 1,000 births).[1] The most common types of cancer diagnosed during pregnancy are breast, cervical, melanoma, Hodgkin's disease, leukemia, and non-Hodgkin's lymphoma.[1] Burkitt's lymphoma (BL) is an aggressive variety of B cell non-Hodgkin's lymphoma with a rapidly fatal course if untreated. BL is associated with rapid growth of the tumor, increased involvement of the central nervous system, risk of relapse, and high risk of tumor lysis. BL during pregnancy is rare. Case reports of BL in pregnancy are limited, and typically neither the mother nor the infant survives.[2] We report the unusual case of a patient who was diagnosed with BL in the immediate postpartum period.

CASE REPORT

A 26-year-old lady, para 1 (previous vaginal delivery six years back) had an uneventful second pregnancy without any medical and obstetric complications and delivered a live baby of 2.6 kg by lower segment cesarean section. During cesarean section (indication: Nonprogress of labor), a right-sided solid ovarian tumor was found and a right-sided ovariotomy was done. Histopathology revealed high-grade non-Hodgkin's lymphoma of right ovary. She was immediately referred to our hospital.

We received the patient on the third postpartum day with preauricular lymphadenopathy, fever, abdominal swelling, respiratory distress, and bilateral pedal edema. A review of ovarian tumor histopathology as well as biopsy from preauricular lymph node showed sheets of round lymphoid cells having scanty cytoplasm. Some of the cells had cytoplasmic vacuoles. There were numerous tingible body macrophages imparting a 'starry sky appearance' [Figure 1]. The cells were positive for CD79a (CD: Cluster of differentiation), CD20, CD10, and bcl-6, and were negative for CD3 and myeloperoxidase. The lymphoma exhibited a high mitotic rate with 100% of the cells staining positive for Ki67. She was found severely anemic with a hemoglobin level of 5.2 g%. Contrast enhanced computed tomography (CECT) of thorax and whole abdomen showed mediastinal adenopathy, multiple hypodense hepatic lesions suggestive of secondaries with a thick clumped gut in the pelvis with blurring of adjacent fat planes [Figure 1]. Bone marrow aspiration confirmed stage IV BL.

Gradually, she developed sixth nerve palsy with posterior uveitis with vitreous opacities. She was started on CODOX-M/IVAC regime but died on the 14th postpartum day before the first cycle could be completed due to rapid progression of the disease.

DISCUSSION

Adult BL is a rare but aggressive type of B cell non-Hodgkin's lymphoma that accounts for only 1–2% of all cases of non-Hodgkin's lymphoma in immunocompetent adults.[3] Because of such a high growth fraction of BL,

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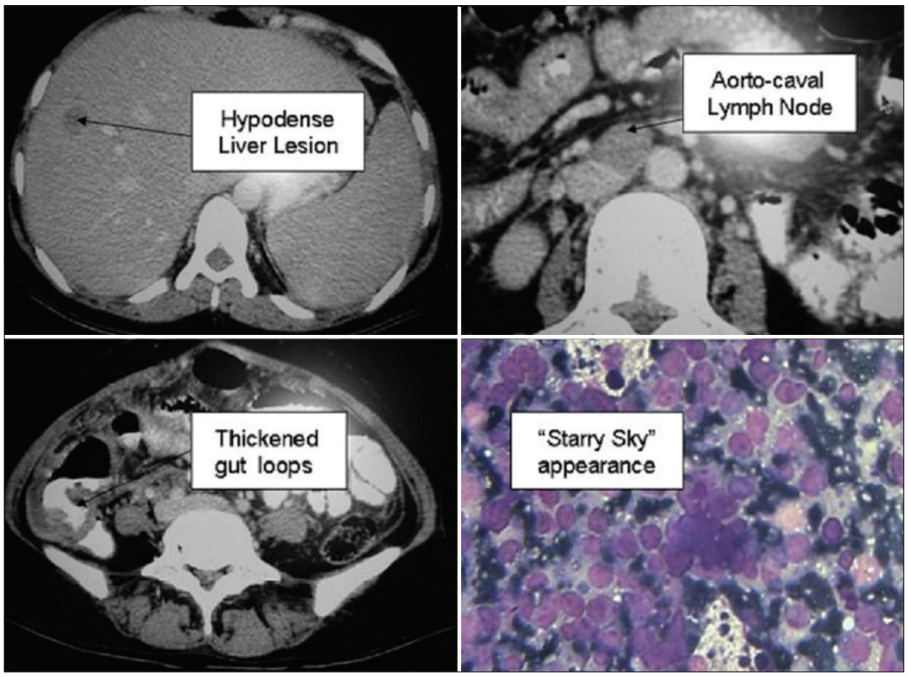

Figure 1: Computed tomography scan of abdomen showing extensive intra-abdominal disease and photomicrograph of ovarian biopsy slide showing starry sky appearance of Burkitt's lymphoma

with a doubling time of 25 hours, the disease carries a very high mortality or relapse rate. Most case reports show that majority of the pregnant patients with BL present with bilateral breast infiltrations, and/or a few present with ovarian masses. They receive either no treatment or only cyclophosphamide monotherapy and ultimately die of the rapidly progressive disease.[2] A very few number of cases with favorable outcome exist in the literature. One report described the successful treatment of a 21-year-old woman diagnosed with stage II disease at 26 weeks with CODOX-M/IVAC, with the baby delivered by Caesarean section at 32 weeks (after two cycles) and the last two cycles of therapy subsequently given with the addition of methotrexate. Fourteen months later, both baby and mother were reported alive and well.[4] Another report described a 20-year-old woman, 12 weeks pregnant, found to have BL of the right ovary with ascites. She underwent surgery to remove all visible disease, electively terminated the pregnancy, and then received multiagent combination chemotherapy with the Cancer and Leukemia Group B 9521 regimen. She was reported to be disease-free one year later.[5]

This case presents certain unique features despite the negative outcome. First, the disease did not exhibit its true nature until puerperium, leading us to speculate the possible biological effects of the pregnancy on the lymphoma. Data from animal experiments with serially transplantable virus-induced lymphoma inoculated into female rats suggests a protective effect against growth of the tumor which is greater during early pregnancy, less accentuated during late pregnancy, and ceases entirely postpartum and particularly during lactation, when the growth of the tumor resumes on an accelerated course.[6] We believe this case presents an *in vivo* correlate of this phenomenon. Second, the aggressiveness of BL is evident from this case, given the speed at which the patient deteriorated despite institution of treatment.

CONCLUSION

BL is a very aggressive disease and is very rarely associated with pregnancy. Aggressive chemotherapy is helpful in a minority of patients. There may be an inhibitory effect of the pregnancy on the lymphoma as is evident in this case.

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