

Course of Non-AIDS Kaposi's Sarcoma During Pregnancy: Case Report

AIDS'le İlişkili Olmayan Kaposi Sarkomunun Gebelik Seyri

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ABSTRACT Kaposi's sarcoma (KS) is common disease in AIDS patients and can occur in non-AIDS patients. KS accompanied by pregnancy is a rare condition and its course during pregnancy is a controversial issue. We report a pregnant woman with non-AIDS KS and her pregnancy outcome. A multiparous woman presented at first trimester of her pregnancy with the lesions of KS on the dorsum of the feet. The lesions were extended to the lower legs and arms during the second and third trimesters. Pregnancy was uneventful throughout the pregnancy. Delivery was performed by caesarian/section at 38th week of gestation. The baby was healthy with no signs of KS or congenital anomalies. The decision of a pregnancy should be considered cautiously in women with non AIDS-related KS.

Key Words: Sarcoma, kaposi; pregnancy

ÖZET Kaposi sarkomu (KS) yaygın olarak AIDS'li hastalarda görülmekle birlikte AIDS'li olmayan hastalarda da rastlanabilir. Gebelikte Kaposi sarkomu ender bir durumdur ve gebelikteki seyri de tartışmalıdır. Biz burada AIDS'le ilişkili olmayan KS'li bir gebeyi ve hastalığın gebelikteki seyri sunduk. Multipar hasta gebeliğinin birinci trimesterinde ayak sırtında lezyonlarla başvurdu. Lezyonlar 2. ve 3. trimesterde bacaklara ve kollara kadar yayıldı. Gebelikte herhangi bir sorun kaydedilmedi. Gebelik 38. gestasyon haftasında sezaryenle sonlandırıldı. Bebekte herhangi bir konjenital anomali kaydedilmedi. AIDS'li ilişkili olmayan KS'de gebelik kararı dikkatli bir şekilde verilmelidir.

Anahtar Kelimeler: Kaposi sarkomu; gebelik

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Kaposi's sarcoma (KS) is the most common tumor found in patients with the acquired immune deficiency syndrome (AIDS). Although the cellular origin of KS has not yet been fully elucidated, it is thought to be derived from endothelial cells, either of vascular or of lymphatic origin.^{1,2} In total, four types of this tumor have been described. The classical variant of KS occurs more frequently in men than in women. This form is not associated with the HIV virus and this type is distinguished clinically by multiple red to purple plaques or nodules on the lower extremities. It may be associated with malignant neoplasm and disorders of the immune system. KS accompanied by pregnancy is a rare condition and its course during pregnancy is a controversial issue. In the literature, many

pregnant women with AIDS-associated KS have been reported, but there are limited data about the pregnancy associated with non-AIDS KS.³⁻⁶ It has been shown that clinical-grade preparations of human chorionic gonadotropin could have a protective effect in immunodeficient mice.⁷ However, the role of pregnancy-related factors is not clear due to the descriptions of KS development or progression during pregnancy.^{8,9} In this case, we described a pregnant woman with non-AIDS KS and discussed the course of this classical variant during pregnancy.

CASE REPORT

A 31-year-old woman, G3 P2, presented for routine antenatal care at the 12th weeks of gestation. She gave a history of Kaposi's sarcoma for the last 4 years. Minimal lesions only on the dorsal region of the feet were observed on examination. In the biopsy of the skin lesion, there was a patchy, sparse, dermal perivascular infiltrate consisting of lymphocytes and plasma cells (Figure 1). HIV serology and Western blot test gave result negative. The plasma analysis was detected positive for human herpes virus 8 (HHV8) by immunofluorescence assay (IFA). On examination, she had purplish plaques skin lesions which were present mainly on the dorsum of the feet. Oral mucosa was normal. An ultrasound scan revealed a viable fetus with normal appearance at the 12th weeks of gestation and

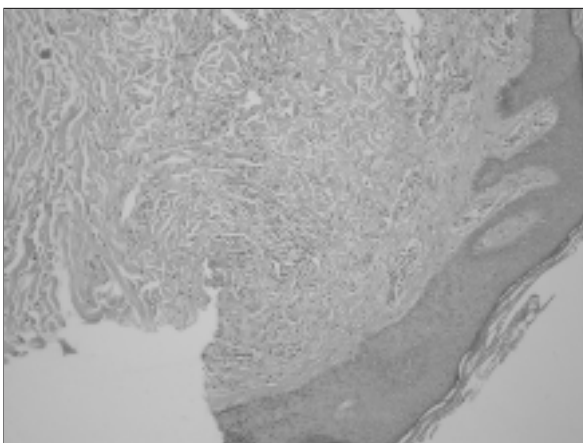


FIGURE 1: Patch stage. Numerous slit-like spaces throughout the dermis HE, x40.

beta-human chorionic gonadotropin (β -HCG) level was recorded as 154.000 mIU/mL in this gestational week.

An informed consent was obtained from patient and then we investigated the patient's all clinical previous records since onset of KS. The previous treatment regimen included six cures of chemotherapy with adriamycine, bleomycine and vincristine and three cures of radiotherapy and interferon- α 2 2 years ago before pregnancy. No treatment was given to the patient within last two years. A computerized tomography (CT) scan of the abdomen and thorax showed no pathology. The patient had minimal lesions only on the dorsal region of the feet during last two years before pregnancy. She decided to have a pregnancy without any consultation of her physician.

On examination at the 20th week of gestation, her lesions were extended to the legs and increased in size. β -HCG level decreased and was detected as 26400 mIU/ml. We consulted the patient with oncology team and they did not recommend any treatment. Her pregnancy was uneventful on the ultrasonography. Routine blood tests and viral serology for HBV and HCV were normal. Her chest examination revealed no respiratory complaints suggesting a possible involvement of the lung. We did not perform the chest radiography due to the pregnancy. At the 32nd week of pregnancy, the lesions were more extensive and she had also lesions on her both arms and dorsum of the hands (Figure 2, 3). A more precise decrease in β -HCG level was determined and the value of this hormone was 9600 mIU/mL. During this time, oncology team recommended to perform a chemotherapy but patient did not accept to take treatment and the treatment of the KS was delayed until delivery. She was delivered by emergency caesarian section for the deterioration of the disease at 38 weeks of pregnancy. A 2900 gram, male baby was healthy with no signs of KS and congenital anomalies. The baby's plasma test for HHV8 by indirect IFA gave negative result. Deterioration of the disease continued after delivery and radiotherapy was applied to the patient throughout the one month but no recovery was obtained. Recently, chemotherapy has been started.



FIGURE 2: Purplish plaques and nodules on the feet.



FIGURE 3: Purplish plaques on the hands.

DISCUSSION

Classic KS has a peak incidence of about 40–70 years and occurs more frequently in men than in women. KS-AIDS patients tend to be younger, averaging 20–40 years at age of onset. Subsequently, a progressive rise in incidence occurred in women aged 25–29 years and in men aged 35–39 years.¹⁰ KS is usually evident as multiple vascular nodules in the skin and other organs and is diagnosed by histopathology. The systemic medications that are American FDA-approved are liposomal daunorubicin, liposomal doxorubicin, paclitaxel and interferon- α .¹⁰ The male-to-female ratio is 10–13/1 and this ratio has led to the hypothesis that female hormones could control KS.

Although many pregnant women with AIDS-related KS have been reported, classic KS associated with pregnancy has been reported infrequently.^{3–6} The effect of pregnancy on KS remains controversial. Although a protective effect of pregnancy against KS in two women was observed by Lunardi-Iskandar et al, other authors reported an absence of a clear effect of pregnancy on KS.^{6,8,9,11} In our case, an increase in size and number of KS lesions started within 2nd trimester and continued during 3rd trimester. Report of cases of AIDS-associated KS regressing during pregnancy evoked the role of pregnancy-related factors such as human chorionic gonadotrophin (hCG). There

are conflicting reports about the effect of hCG on the occurrence and progression of KS in pregnant women. HCG can be found in the blood before the first missed menstrual period, as early as six days after implantation. Levels of hCG increase steadily in the first 14 to 16 weeks and peak around the 14th week of pregnancy, and then decrease gradually. Some studies revealed that clinical-grade preparations of hCG could inhibit the growth of the neoplastic KS-Y1 cell line in vitro and in immunodeficient mice. These results suggest that hCG may have a protective effect against KS.^{2,11,12} However, there are highly controversial results of other clinical studies using commercial hCG preparations. These studies showed that KS in human remained in partial remissions or stabilization.^{13,14} In addition, absences of effect or even progression were observed in other ones.^{15,16} The course of disease was deteriorated within second and third trimesters which may be due to HCG decrease.

A direct antiviral activity of hCG has also been reported. Along with an inhibition of HIV infection rate of macrophages, a considerable decrease in HIV-1 expression in a murine KS-Y1 model in response to impure hCG have been reported.^{12,17,18} This synchronous effect of hCG may play a role in pregnant women with AIDS-associated KS for regression or stabilization of KS. For these reasons, determination of the effects of hCG on only

KS in pregnant women with AIDS-associated KS may be difficult. The fact that the incidence of the disease has drastically declined with the introduction of antiretroviral therapy may support this suggestion. Because any change in the sarcoma was not observed during the first trimester in our non-AIDS KS case, increase in hCG level may have stabilized the disease in this period. However, we observed an increase in size and number of KS lesions during the 2nd and 3rd trimesters. Although a vertical transmission from mother to infant in KS has been reported, we did not observe KS in the infant of our patient.⁹

In conclusion, pregnancy does not lead to any changes in classic KS during the first trimester in our case. However a progression occurs in the sarcoma during the following trimesters. We suggest that women with classic KS should carefully consider the decision to have a pregnancy due to possible progression of the disease during the mid and last trimesters.

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