Kaposi's sarcoma developing in a geriatric patient with autoimmune hepatitis shortly after immunsupressive treatment

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Dear Editor.

Kaposi sarcoma (KS) is an angioproliferative low-grade neoplasm caused by human herpesvirus-8 (HHV-8), commonly observed in immunocompromised patients. It was first described by Moriz Kaposi in 1872 as idiopathic multiple-pigmented sarcoma of the skin.^[1] KS is more prevalent in individuals with autoimmune diseases due to either immunosuppressive therapy or immune system dysregulation. Autoimmune hepatitis (AIH) is a chronic inflammatory liver disease that can occur at any age.^[2] Here, we report a case of Kaposi sarcoma developing in an elderly patient undergoing corticosteroid and azathioprine (AZA) therapy for autoimmune hepatitis.

A 70-year-old woman was referred for evaluation due to persistently elevated liver function tests detected during routine screening. She reported fatigue and unintentional weight loss. Laboratory investigations showed markedly elevated transaminases; AST: 1708 U/L [Upper Limit of Normal (ULN): 35 U/L], ALT: 1624 U/L [ULN: 50 U/L], with normal albumin 4.7 g/dL [ULN: 4.5 g/dL] and total bilirubin 17 mg/dL[ULN: 1.2 mg/dl]), γ -Glutamyltransferase (GGT) and alkaline phosphatase (ALP) were also elevated at 158 U/L [ULN: 38 U/L]) and 214 U/L [ULN: 98 U/L]) respectively.

Additional findings included a normal blood count, an activated partial thromboplastin time of 33.8 seconds (reference range: 26–37 seconds), and a prothrombin time of 13 seconds (reference range: 10–13 seconds). Hypergammaglobulinemia was present, with elevated serum IgG 17.8 g/L; (ULN: 16 g/L). Serological tests for viral hepatitis and celiac disease were negative. Ceruloplasmin, copper, and α -1 antitrypsin levels were within normal limits. Autoimmune serology revealed negative antinuclear antibodies (ANA), antimitochondrial antibodies (AMA), and anti-liver/kidney microsomal type 1 (anti-LKM1) antibodies, while anti-smooth muscle antibodies (ASMA) were positive at a titer of 1:320.

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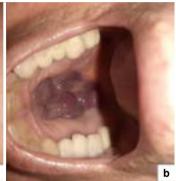


Figure 1. A brown nodular lesion, 5–6 mm in size, on the medial aspect of the forearm **(a)**, a purple tumoral lesion in the oral mucosa, 2–3 cm in size, on the hard palate **(b)**.

Abdominal ultrasound showed a homogeneous liver echotexture and a normal-sized spleen. A liver biopsy revealed portal lymphocytic and plasmocytic infiltration, moderate interface hepatitis, and normal bile ducts. No cholangiographic study was performed. The patient was diagnosed with autoimmune hepatitis and started on methylprednisolone (0.8 mg/kg) followed by AZA (1.5 mg/kg). Thiopurine S-methyltransferase (TPMT) activity was confirmed as normal before initiating AZA.

Four months after starting corticosteroids and AZA, the patient developed small, painless, red-to-purple lesions on her forearm and oral mucosa (Fig. 1). Lesions later appeared on her ears, legs, and perianal region. An excisional skin biopsy confirmed Kaposi sarcoma, with further evaluation ruling out visceral and lymph node involvement. Further evaluation revealed no involvement of the lungs, abdominal organs, or lymph nodes. Laboratory testing was positive for HHV-8 and negative for Human immondeficiency virus. Immunosuppressive therapy was tapered, and the patient was referred to a medical oncology clinic for further management. After one year of follow-up, the patient remained clinically stable, with no progression in laboratory or imaging findings.

Kaposi sarcoma has four clinical-epidemiological types: the original classic type, African or endemic type, iatrogenic or immunosuppressant-associated type, and epidemic or AIDS-related type. [3] Classic KS is relatively common among elderly men in some regions such as Turkiye. [4] Iatrogenic KS is frequently reported in patients receiving immunosuppressive therapy for autoimmune diseases, particularly rheumatologic conditions and inflammatory bowel disease. [5] Although Kaposi sarcoma is typically associated with long-term immunosuppression, there are documented cases of KS developing



after short-term corticosteroid and AZA therapy in patients with autoimmune diseases, including Crohn's disease. [6] However, only a few cases of KS following short-term steroid use have been reported in the literature.

The development of Kaposi sarcoma in an elderly patient receiving short-term steroid and AZA treatment for autoimmune hepatitis is a rare and remarkable case. The duration and dose of immunosuppression, as well as the patient's age, can increase the risk of Kaposi sarcoma, requiring a treatment approach tailored to the stage of the disease.

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