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Varicella Zoster Meningoradiculitis in Crohn's Disease Treated with 6-Mercaptopurine

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To the Editor

Thiopurine immunosuppression is associated with an increased risk of infection, including varicella zoster (VZV). To our knowledge there are no reported cases of varicella zoster meningoradiculitis in an inflammatory bowel disease (IBD) patient treated with these agents.

We report a 49-year-old male with a history of ileocolonic Crohn's disease (CD) who presented to the emergency department (ED) with a diffuse skin rash, myalgias, and fever. The patient had been taking 6-mercaptopurine (6-MP) 100 mg daily for his CD for over 10 years. He had two previous ileocolonic resections; the most recent was in 1999. He had mild anastomotic recurrence but his CD was in clinical remission.

The patient stated that he developed an erythematous rash over 3 weeks on his upper limbs and trunk. He noticed numerous small circular lesions on his posterior right thigh 2 weeks later. One week prior to his ED presentation, he developed myalgias and headache, and on the day of presentation he developed fever of 102°F.

On physical examination his temperature was 100°F. There were a number of diffuse coalescing small pink dry scaly papules and thin plaques across the trunk and arms. There were a number of edematous nodules (>1 cm) on the posterior aspect of his right thigh that were nonscaly and had a central violaceous hue. The remainder of the physical examination was unremarkable.

Laboratory examination was significant for a total white blood count of 2.9 K/μL with normal differential. Liver and renal function were normal. Testing for borrelia burgdorferi, human immunodeficiency virus (antibody and RNA), cytomegalovirus (CMV), Epstein–Barr virus (EBV), and hepatitis B and C were negative. Skin biopsy from the left upper arm demonstrated findings consistent with pityriasis rosea. Skin biopsy from the posterior right thigh showed evidence of a herpes virus infection with follicular involvement and underlying vascular injury. 6-MP was discontinued on admission. The patient defervesced and was discharged with instructions to take 10 days of valganciclovir.

When he was seen in the office 1 week after discharge the patient reported paresthesia and numbness affecting his feet, lower legs, the posterior aspect of his thighs up to the buttocks bilaterally and his penis. He had no episodes of incontinence, urinary retention, or back pain.

His headache persisted. On examination he was afebrile and there were no signs of meningism. The pityriasis persisted and the herpetic lesions were partially crusted. There was decreased sensation to light touch over the sacrum, buttocks, posterior thighs, feet and toes, sparing the anterior proximal thighs, and decreased pinprick on the scrotum. Reflexes were 2+ and symmetric throughout and the toes were downgoing bilaterally.

The patient was seen by the infectious disease service who suspected herpes (herpes simplex virus [HSV] or VZV) lumbosacral radiculitis. A lumbar puncture was performed and the patient was commenced on IV acyclovir. The cerebrospinal fluid (CSF) contained 26 white blood cells per μL (96% lymphocytes). Total protein was 97 mg/dL (normal range 15–45 mg/dL) and glucose was 47 mg/dL. The CSF was positive for VZV by polymerase chain reaction. The patient was diagnosed with VZV meningoradiculitis and was discharged on 21 days of IV acyclovir. Two weeks into his course a repeat VZV PCR was negative, although his symptoms persisted. He remains off 6-MP.

VZV is a neurotrophic herpesvirus that causes chickenpox during primary infection, after which the virus becomes latent in cranial nerves, dorsal roots, and autonomic ganglia.[1] The virus may reactivate many years after primary infection to cause herpes zoster (shingles), a painful vesicular skin rash confined to one or more sensory dermatomes.[2] The incidence of zoster is increased in those older than 60 years and immunosuppressed individuals. The most common complication of zoster is post-herpetic neuralgia, but VZV reactivation can lead to a variety of neurologic and ocular disorders, including vasculopathy, myelopathy, herpes zoster oticus, meningoencephalitis, polyneuritis cranialis, cerebellitis, and necrotizing retinitis.[2] Neurological manifestations of zoster infection can occur without the associated rash (zoster sin herpete). Zoster is thought to be due to retrograde transport of the virus from ganglia to the skin but isolation of the virus from the blood of immunocompromised patients suggests that it can also spread hematogenously.[3] Treatment with intravenous acyclovir for 5–7 days is indicated for any immunosuppressed patient with zoster infection, although longer courses may be necessary if there are signs of ongoing viral replication and progressive end-organ injury.

Zoster meningoradiculitis is a recognized manifestation of zoster infection but is infrequently reported in the literature. There is a case of a fatal varicella meningoradiculitis (without skin involvement) reported in a 77-year-old male following chemotherapy.[4] Varicella pneumonia has been reported in IBD patients taking thiopurines and antitumor necrosis factor agents (alone and in combination), with a number of fatalities.[5,6]

The diagnosis in this case was confused by the coexisting pityriasis rosea, which led the patient to delay reporting the herpetic lesions on his leg. Even after presentation, the symptoms of arthralgias, myalgia, and mild headache were not linked to the herpetic infection until the patient developed more apparent neurological symptoms. This case highlights the risks associated with immunosuppression in IBD and reminds gastroenterologists to be aware of atypical presentations and rare complications of infection in this group.

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