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Case study

Rhomboid glossitis in disseminated CMV infection

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A 53 year-old woman with newly diagnosed AIDS and simultaneous diagnosis of diffuse large B-cell lymphoma was admitted to the hospital for neutropenic fever and diarrhea after her second cycle of R-EPOCH (Rituximab, etoposide, prednisone, vincristine, cyclophosphamide, doxorubicin). She had been recently started on antiretroviral treatment with raltegravir, tenofovir and emtricitabine with appropriate virologic response (HIV PCR 115 copies/ml down from 9 million on diagnosis with CD4 count of 75/ml), and was receiving trimethoprim/sulfamethoxazole prophylaxis, azithromycin prophylaxis and fluconazole for oral thrush. She remained febrile after neutrophil recovery and despite broad-spectrum antibacterials; routine blood tests showed transaminitis. On physical exam, she had persistent thrush, and a large, punched-out, painless mid-tongue ulcer (Fig. 2A). Computerized tomography of the chest showed multiple sub-centimeter nodules, concerning for an opportunistic infection. Fungal cultures of the thrush grew *Candida lusitaniae* with intermediate susceptibility to fluconazole (minimal inhibitory concentration 1 mirco/L). Bronchoalveolar lavage (BAL) bacterial, mycobacterial and fungal cultures, *Pneumocystis* direct fluorescent antibody and cytological examination for malignancy were negative.

A biopsy of the central tongue lesion showed numerous intranuclear and cytoplasmic inclusions (Fig. 1A), consistent with cytomegalovirus (CMV) cytopathic effects. Bacterial and fungal stains were negative. Immunohistochemical stains were also positive for CMV (Fig. 1B; M; DDG9 and CCH2; DAKO and Chemicon cocktail), CMV culture from BAL was positive, and plasma CMV viral load was 75,000 IU/mL. The patient was started on treatment with valganciclovir and caspofungin, with complete resolution of her fever, diarrhea, thrush and lingual ulcer and resumed chemotherapy uneventfully (Fig. 2B).

CMV disease usually presents with constitutional symptoms and signs (generalized malaise, fever, flu-like syndrome) and/or diarrhea, as illustrated in the present case. However, this case also highlights that CMV can have unusual manifestations, such as pneumonitis with minimal respiratory symptoms and oral ulcers. The rapid improvement of the patient's oral thrush could be due to appropriate treatment of fluconazole-nonsusceptible *C. lusitaniae*. However, we cannot exclude a contribution of anti-CMV treatment, given the immunomodulatory properties of the virus and their potential interference with host-pathogen interactions across different kingdoms.

In conclusion, clinicians should have a high clinical suspicion for CMV as a treatable cause of non-specific and protean clinical syndromes, including oral ulcers, in T-lymphocyte-depleted patients who are not on anti-CMV prophylaxis. In immunocompromised patients, it is important to obtain tissue specimen for definitive pathologic diagnosis, in order to diagnose uncommon syndromes and guide management.

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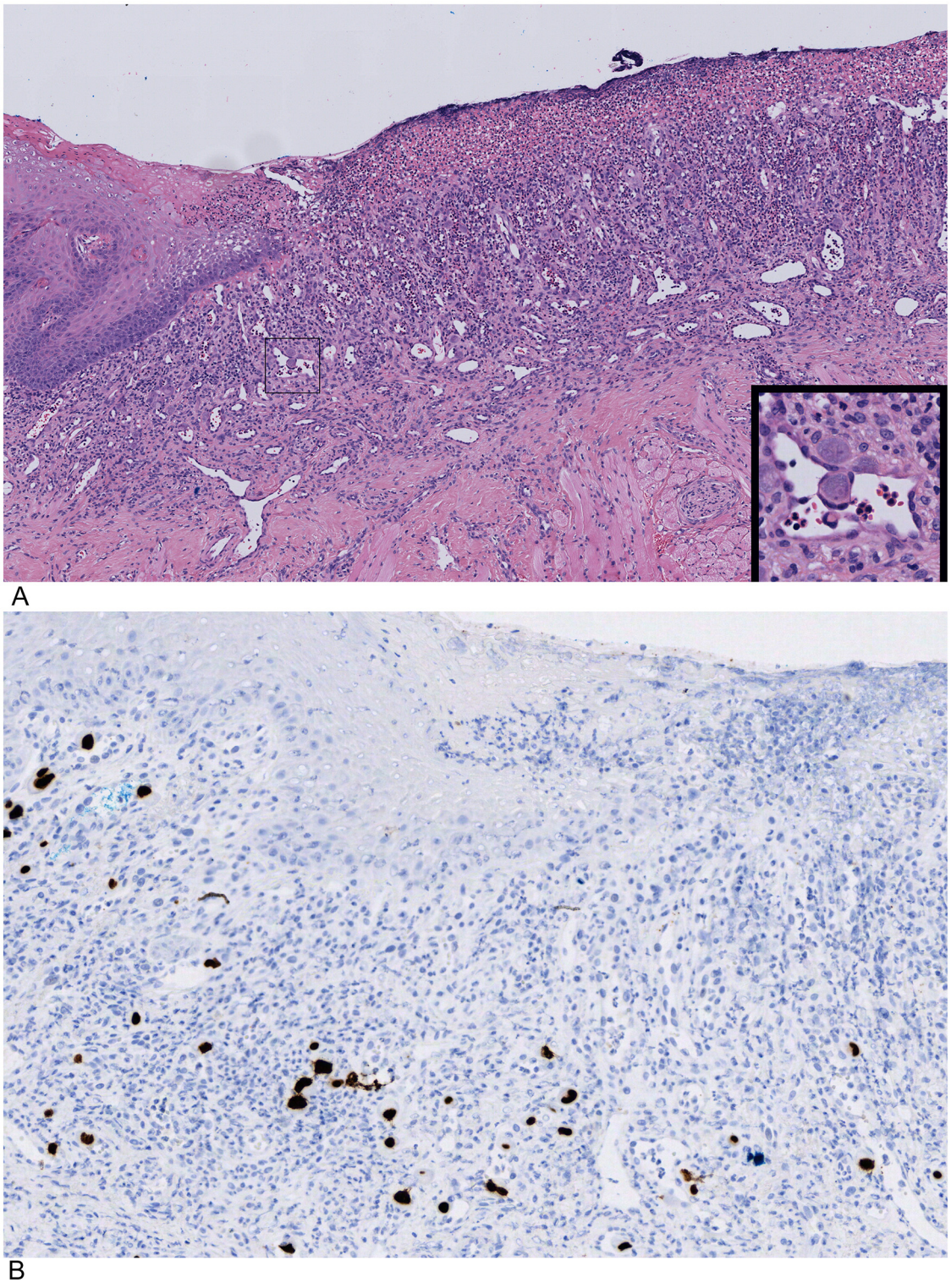

Fig. 1. (A) H + E stain of biopsy of central tongue ulcer,with magnification window showing intranuclear inclusion bodies within glandular tissue. (B) M; DDG9 and CCH_2; DAKO and Chemicon cocktail stain of tongue biopsy showing positive stain for CMV inclusions.

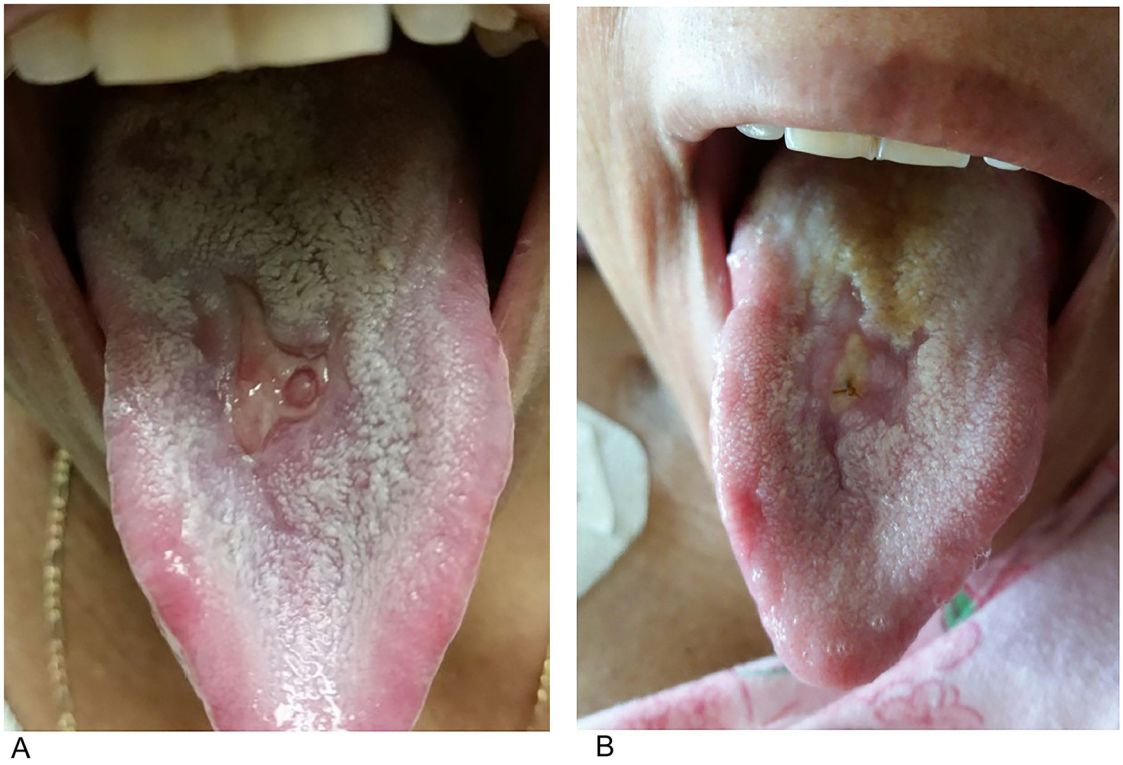

Fig. 2. (A) Clinical image of lingual ulcer,on diagnosis and before treatment. (B) Clinical image of tongue,1 week following with valganciclovir.