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CORRESPONDENCE

Stevens-Johnson syndrome without skin lesions extensively involving the digestive tract

Stevens-Johnson syndrome (SJS) is a severe adverse cutaneous reaction characterized by extensive skin detachment and mucosal erosions, posing a potential risk to life.[1] A rare variant, known as SJS without skin lesions (SWSL), presents only with mucosal membrane involvement, leaving the skin unaffected.[2] We here report a case of SWSL caused by the sulfamethoxazole–trimethoprim (SMX–TMP) medication, notably affecting the digestive tract.

An 84-year-old Japanese woman was admitted to our hospital to treat with SMX–TMP for a urinary tract infection. Following her COVID-19 vaccination, she developed a fever, subsequently progressing to a severe sore throat and eyelid swelling accompanied by deteriorating mucosal erosions, warranting her referral to our hospital. Her symptoms included a fever of 38.7°C and pain spanning from her lips to her upper abdomen. A physical examination identified conjunctival redness, lip swelling, and oral mucosal membrane erosions, yet no skin rash was observed (Figure 1A–C). Endoscopic examination revealed extensive ulcerations in the esophagus and stomach (Figure 1D,E).

Laboratory investigations showed elevated levels of serum C-reactive protein (8.4 mg/dL; normal range, <0.2 mg/dL) and soluble IL-2 receptor (1430 U/mL; normal range, <474 U/mL). The absence of an Anti-*Mycoplasma* antibody (detected via the particle agglutination method), anti-streptolysin O antibody, and autoantibodies to desmogleins/BP-180 were noted. Clinical features supported the diagnosis of SWSL, but unfortunately, histologic confirmation was not available. The patient responded well to a corticosteroid pulse therapy with 1000 mg of methylprednisolone and oral administration of prednisolone, which significantly improved the mucosal lesions and

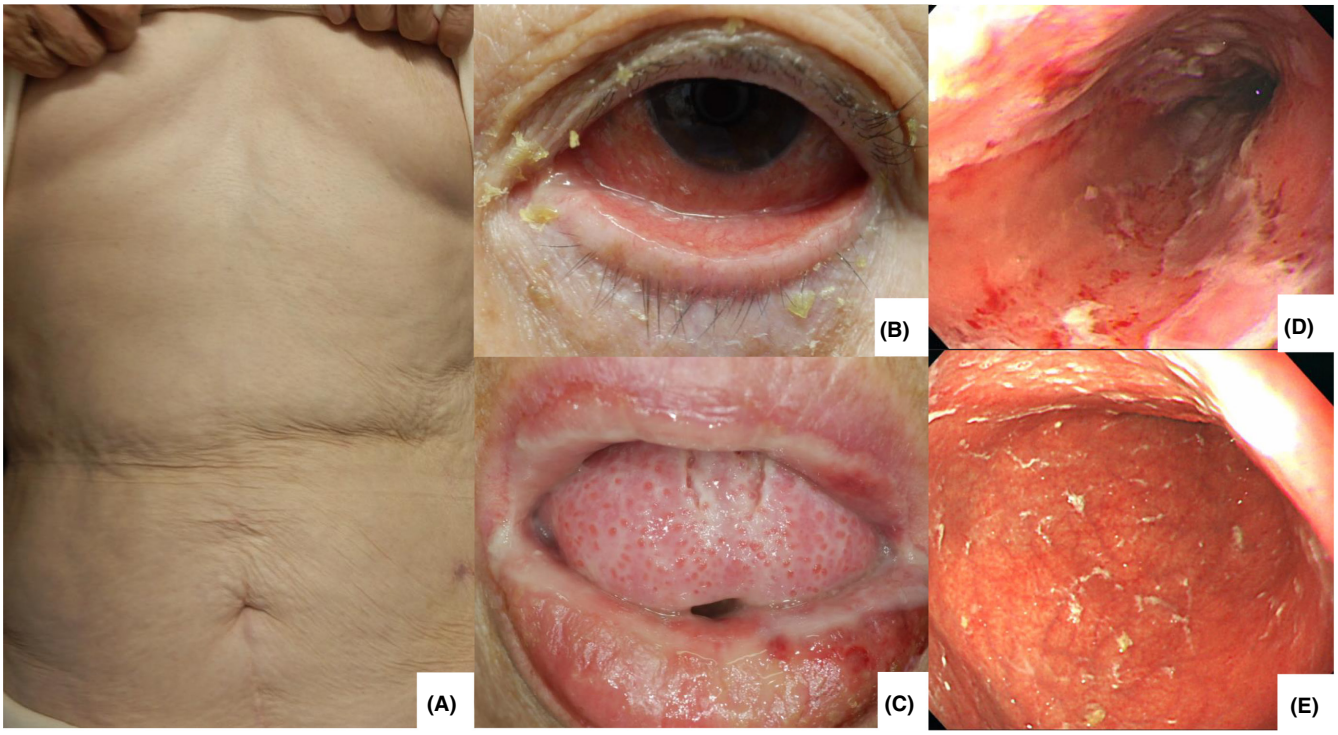

FIGURE 1 Clinical and endoscopic images. (A) No erythema was seen on the body at all. (B, C) Photographs showing erosions of the ocular conjunctiva, lips, and oral mucosa. (D, E) Endoscopic photographs showing extensive ulceration of esophageal and gastric mucosa.

overall condition. Approximately a month later, the patient's urinary tract infection recurred, necessitating the reintroduction of SMX–TMP treatment, which was not suspected of being the causative agent at the time. However, within 2 days of resuming the medication, she again developed lesions in the lips and oral cavity. This confirmed the strong correlation between this drug and its development. Following the discontinuation of the medication, the patient was successfully treated with half-dose corticosteroid pulse therapy.

The current case presents two extraordinary features that warrant discussion. First, it represents drug-induced SWSL, which is quite uncommon. In the published medical literature, only four such cases have been documented, while SWSLs themselves were reported in some numbers; these are primarily associated in mycoplasma infections.[2] Second, the presence of severe gastrointestinal involvement, which occurs in roughly 10% or fewer SJS/TEN cases, is also rare.[3] The unique aspects of similar cases might perplex the diagnostic process and treatment decisions. Although it might be rare, acknowledging the clinical resemblance to autoimmune bullous disease and the risk of serious, latent gastrointestinal complications, as demonstrated in the current case, is key to preventing diagnostic and treatment delays.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

ETHICS STATEMENT

Approval of the research protocol: No human participant was involved in this study.

Informed Consent: The patient provided informed consent for the publication of the images submitted with this article.

Registry and the Registration No.: N/A.

Animal Studies: N/A.

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