



Letter to the Editor

Colonic involvement in Stevens–Johnson syndrome-like mucositis without skin lesions



Dear Editor

Stevens–Johnson syndrome (SJS) is an acute disease characterized by severe inflammation and necrosis of two or more mucosal membranes and systemic symptoms such as fever and malaise.¹ SJS has most often been associated with a drug reaction and less frequently with infections such as *Mycoplasma*.² While skin manifestation is commonly seen in SJS patients, SJS-like mucositis without skin lesions has been rarely reported. Such cases have been called SJS without skin lesions, Fuchs syndrome or atypical SJS.³ This condition is believed to be a variant of erythema multiforme majus and usually non-life-threatening because of lacking widespread skin involvement. Although there is an opinion that such cases should not be diagnosed as SJS,⁴ attention should be paid to this variant from the practical view point. Meanwhile, this mucosal condition has been increasingly recognized as having a close association with *Mycoplasma pneumoniae* infection and is currently known as *Mycoplasma pneumoniae*-associated mucositis (MPAM).⁵ Here, we report a rare case of SJS-like mucositis without skin lesions, presenting with severe melena caused by colon membrane involvement.

A 44-year-old Japanese female was referred to our hospital (day 0) because of high fever, cough, and sore throat. Seven days prior to our first examination (day-7), she developed conjunctival injection, followed by severe ulceration on the lips and oral mucosa (day-5). She had taken medicines including acetaminophen and codeine phosphate since day-7. A rapid antigen test using swabs from her nasal mucosa was performed with a positive result for influenza type B, and she was given laninamivir on day-4. The patient was admitted to another hospital and all medicines were ceased on day-2, but her eye and mucosal symptoms deteriorated rapidly.

On our physical examination, her conjunctiva was hyperemic with marked eye discharge and visual disturbance in both eyes (Fig. 1a). There were painful ulcerative lesions on the lower lip (Fig. 1b) and oral mucosa (Fig. 1c). Her skin and genital mucosa were not affected. Histopathologically, a mucosal biopsy specimen taken from the lower lip showed a mild lymphocytic infiltrate in the upper lamina propria with lymphocytic invasion into the epithelium (Fig. 1d). Ophthalmological consultation indicated typical SJS findings including corneal erosions, pseudomembranes and syncanthus in both eyes. Chest roentgenological examination displayed pneumonia in the right lower lung field. Laboratory examination showed elevated levels of neutrophil count (9048/

mm³; normal, 1550–7000/mm³) and CRP (9.08 mg/dL; normal, <0.51). *M. pneumoniae* IgM was negative by enzyme immunoassay, and culture of a pharyngeal swab was negative for bacteria. Herpes simplex virus IgG and IgM titers were negative.

A diagnosis of SJS-like mucositis without skin lesions was made, and corticosteroid pulse therapy with methylprednisolone 1000 mg daily for 3 days was initiated on day 1, followed by prednisolone 60 mg daily. Intravenous administration of sulbactam/ampicillin and oral azithromycin was also started on day 1 for pneumonia. On day 4, when the mucosal lesions were alleviated, the patient had bloody stool with rapid progression of anemia as her serum hemoglobin level decreased from 13.1 mg/dl to 11.2 mg/dl (normal, 11.5–14.6 mg/dl) in the 8-day duration. A colonoscopy revealed multiple erythematous erosions on the alimentary canal mucosa of rectum (Fig. 2a). A biopsy from a rectum lesion showed apoptotic cells in the glandular epithelium with lymphocytic infiltration into the lamina propria mucosae (arrowheads, Fig. 2b). Upper gastrointestinal endoscopy showed only mild gastritis. A diagnosis of SJS-like mucositis with intestine involvement was made. Since melena was spontaneously improved, no therapy was added for proctitis. Because of improvement of pneumonia, the antibiotics were discontinued on day 9. The erosions of lower lip and oral mucosa were epithelized, and her eye lesions were cleared and her vision was recovered to her original levels. Prednisolone was begun to taper on day 17. On day 31, she was discharged at the prednisolone dose of 40 mg daily, which was then discontinued on day 78 without recurrent. The mucositis of infection-associated mucositis is usually milder than that of SJS and respond well to the therapy,⁴ but the drug was carefully tapered.

The patient's clinical manifestations, including the ocular and oral lesions, were compatible with SJS-like mucositis without skin lesions. The cause of mucositis is speculative in our patient. Since the chest radiograph showed a typical finding of *Mycoplasma pneumoniae* infection, we consider that *Mycoplasma pneumoniae* infection is the most likely cause of mucositis despite the negative results of clinical investigations for *Mycoplasma pneumoniae* infection. The lung involvement of SJS could be excluded by the chest radiograph. Other possibilities include drugs and influenza type B infection. Although acetaminophen is known to cause of SJS, lymphocyte stimulation test for this culprit drug was negative. We can not negate the possibility of influenza infection, as three cases of SJS have been reported after influenza infection.⁶

The present case showed widespread colon erosions with asymptomatic anemia and melena. The differential diagnoses were infectious colitides, antibiotic-associated diarrhea,

Peer review under responsibility of Japanese Society of Allergy.

<http://dx.doi.org/10.1016/j.alit.2014.08.010>

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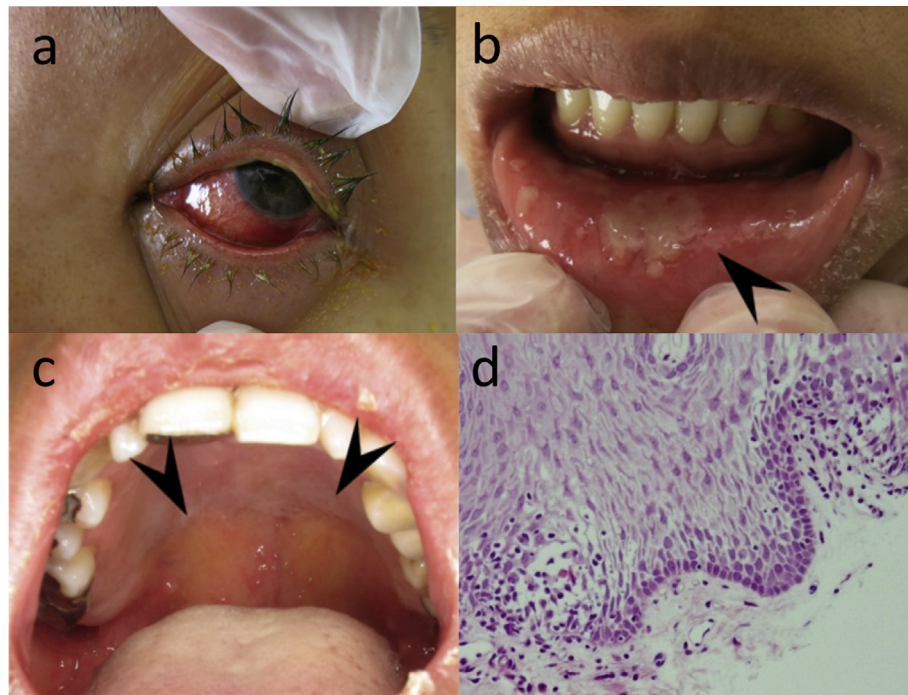


Fig. 1. Clinical appearances of left eye (a), lip (b), and oral mucosa (c). A conjunctiva of both eyes was hyperemic with a marked eye discharge. Lower lip and oral mucosa showed painful ulcerative lesion (arrowhead). Histopathological findings of the lower lip (d). A mild lymphocytic infiltrate in the upper lamina propria with lymphocytic invasion into the epithelium (hematoxylin – eosin stain, original magnification $\times 400$).

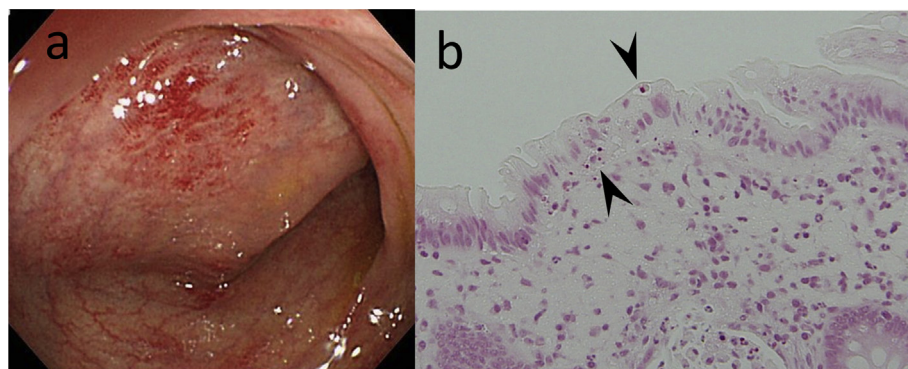


Fig. 2. Colonoscopy showing multiple erythematous erosions on the alimentary canal mucosa of rectum (a). Histopathological findings of a rectum lesion (b). Apoptotic cells (arrowheads) in the glandular epithelium with lymphocytic infiltration into the lamina propria mucosae (hematoxylin – eosin stain, original magnification $\times 400$).

pseudomembranous colitis, and inflammatory bowel disease. However, all laboratory data showed negative results for bowel infection and the patient lacked the histories and symptoms ordinarily presented in these diseases. Furthermore, the anemia and melena disappeared after the systemic corticosteroid therapy. We thus diagnosed the intestinal condition as colon involvement of SJS-like mucositis. Intestinal involvement has not been reported in either SJS-like mucositis without skin lesions or MPAM. Severe gastrointestinal involvement was rarely reported in SJS or toxic epidermal necrosis (TEN). Approximately 10 cases have been reported to have colonic involvement of SJS.⁷ The colonic lesions may occur long after the initial cutaneous lesions in SJS. Our case suggests that abdominal symptoms, anemia, and feces should be carefully checked in patients with SJS and TEN, especially with SJS-like mucositis without skin lesions.

Conflict of interest

The authors have no conflict of interest to declare.

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Received 16 June 2014

Received in revised form 7 August 2014

Accepted 15 August 2014

Available online 16 December 2014