International Journal of Contemporary Pediatrics
Bansal R et al. Int J Contemp Pediatr. 2026 Dec;13(12):2604-2608
https://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20264335

Case Report

When skin speaks for the gut: pediatric ulcerative colitis presenting as Sweet syndrome

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Received: 09 October 2026
Revised: 05 November 2026
Accepted: 20 November 2026

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ABSTRACT

Pediatric ulcerative colitis (UC) is an uncommon inflammatory bowel disease that may initially present with atypical extraintestinal manifestations, resulting in delayed diagnosis. Sweet syndrome (acute febrile neutrophilic dermatosis) is a rare dermatological manifestation of UC in children and may closely mimic severe bacterial sepsis.

Keywords: Pediatric ulcerative colitis, Sweet syndrome, Neutrophilic dermatosis, Inflammatory bowel disease, Extraintestinal manifestations, Colonoscopy, Case report

INTRODUCTION

Ulcerative colitis (UC) is a chronic inflammatory bowel disease that may be accompanied by extraintestinal manifestations (EIMs), including cutaneous disease. Cutaneous manifestations may precede, accompany or follow intestinal symptoms and can occasionally dominate the clinical presentation, obscuring the underlying bowel disorder.[1,2] Sweet syndrome (acute febrile neutrophilic dermatosis) is a rare reactive neutrophilic dermatosis characterized by fever, tender erythematous papules, plaques or nodules, peripheral neutrophilia and a dense dermal neutrophilic infiltrate on skin biopsy.[3] Although skin EIMs are recognized in pediatric IBD, Sweet syndrome is distinctly uncommon in children.[4,5]

The present case is notable because prolonged fever, systemic inflammation and painful pustular skin lesions initially raised concern for severe infection. The eventual diagnosis was supported by the combination of continuous colitis on colonoscopy, radiologic evidence of inflammatory bowel disease, exclusion of major infections, and cutaneous biopsy demonstrating dense neutrophilic dermatosis. Similar pediatric cases have described Sweet syndrome in association with active UC, including presentations that initially prompted evaluation and treatment for infection.[6,7]

CASE REPORT

A previously healthy 7-year-old girl presented with persistent high-grade fever for 12 days, abdominal pain, vomiting and 6–7 episodes of greenish loose stools per day. On examination, she was febrile and pale and subsequently developed multiple painful pustular skin lesions over the trunk and extremities.

Laboratory investigations revealed microcytic hypochromic anemia with hemoglobin 8.5 g/dl, neutrophilic leukocytosis, markedly elevated C-reactive protein (179.8 mg/l), hypoalbuminemia (2.9 g/dl) and hypokalemia (2.6 mEq/l). Fecal occult blood was positive. Stool microscopy demonstrated mucus, occult blood, pus cells and budding yeast cells. Stool and urine cultures were negative, while blood cultures showed no persistent bacteremia. Extensive infectious evaluation,

including testing for malaria, dengue, scrub typhus, leptospirosis, Widal reaction, HIV and tuberculosis by GeneXpert, was negative. Tissue transglutaminase antibodies and autoimmune work-up were also negative. Contrast-enhanced CT of the abdomen demonstrated circumferential bowel-wall thickening involving the terminal ileum, caecum, ascending colon, sigmoid colon and rectum, with loss of haustration, mesenteric lymphadenopathy and minimal pelvic free fluid, suggestive of inflammatory bowel disease. Colonoscopy demonstrated multiple healing superficial ulcers extending continuously from the rectosigmoid colon to the caecum, with caecal ulcers and backwash ileitis; multiple biopsies were obtained.[8]

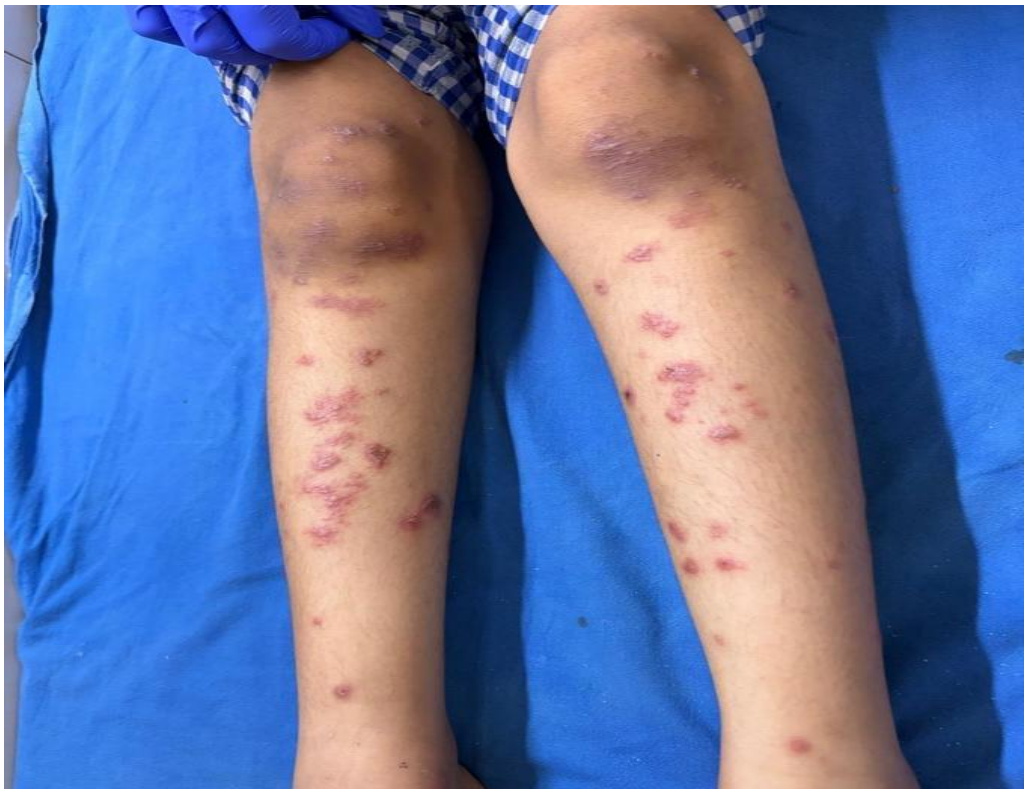


Figure 1: Multiple erythematous/pustular and crusted cutaneous lesions over the lower limbs, with areas of post-inflammatory hyperpigmentation.

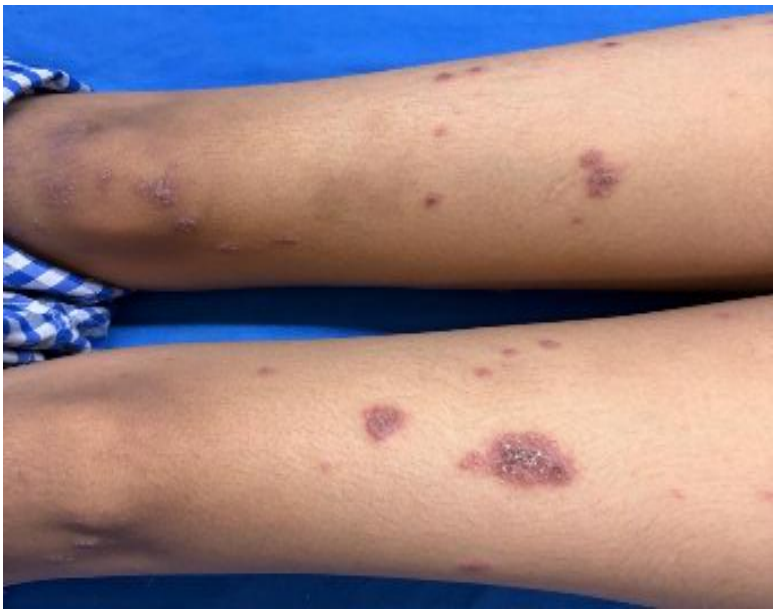


Figure 2: Close-up view of painful inflammatory cutaneous lesions over the lower extremities.

During hospitalization, the patient received broad-spectrum antibiotics because of persistent fever and concern for multidrug-resistant infection. Despite appropriate antimicrobial therapy, fever and skin lesions persisted. Punch biopsy of the cutaneous lesions demonstrated dense neutrophilic dermatosis, confirming Sweet syndrome.Following initiation of oral prednisolone along with mesalamine therapy, fever rapidly subsided, cutaneous lesions resolved, inflammatory markers improved, electrolyte abnormalities were corrected, and gastrointestinal symptoms progressively improved.

Colonic biopsies showed focal surface erosion/ulceration with focal active colitis, cryptitis and crypt distortion, but no granuloma, crypt abscess, prominent basal plasmacytosis, dysplasia or malignancy. Importantly, the colonic biopsy was obtained after systemic prednisolone had already been started; therefore, the relatively nonspecific and treatment-modified histology was not considered sufficient to overturn the clinical and endoscopic diagnosis of ulcerative colitis. GeneXpert Ultra for Mycobacterium tuberculosis on colonic tissue was negative. Taken together, the clinical course, extensive continuous colitis on colonoscopy, backwash ileitis, systemic inflammatory phenotype, exclusion of major infections, skin biopsy demonstrating dense neutrophilic dermatosis, and response to therapy supported a diagnosis of ulcerative colitis with associated Sweet syndrome/neutrophilic dermatosis.

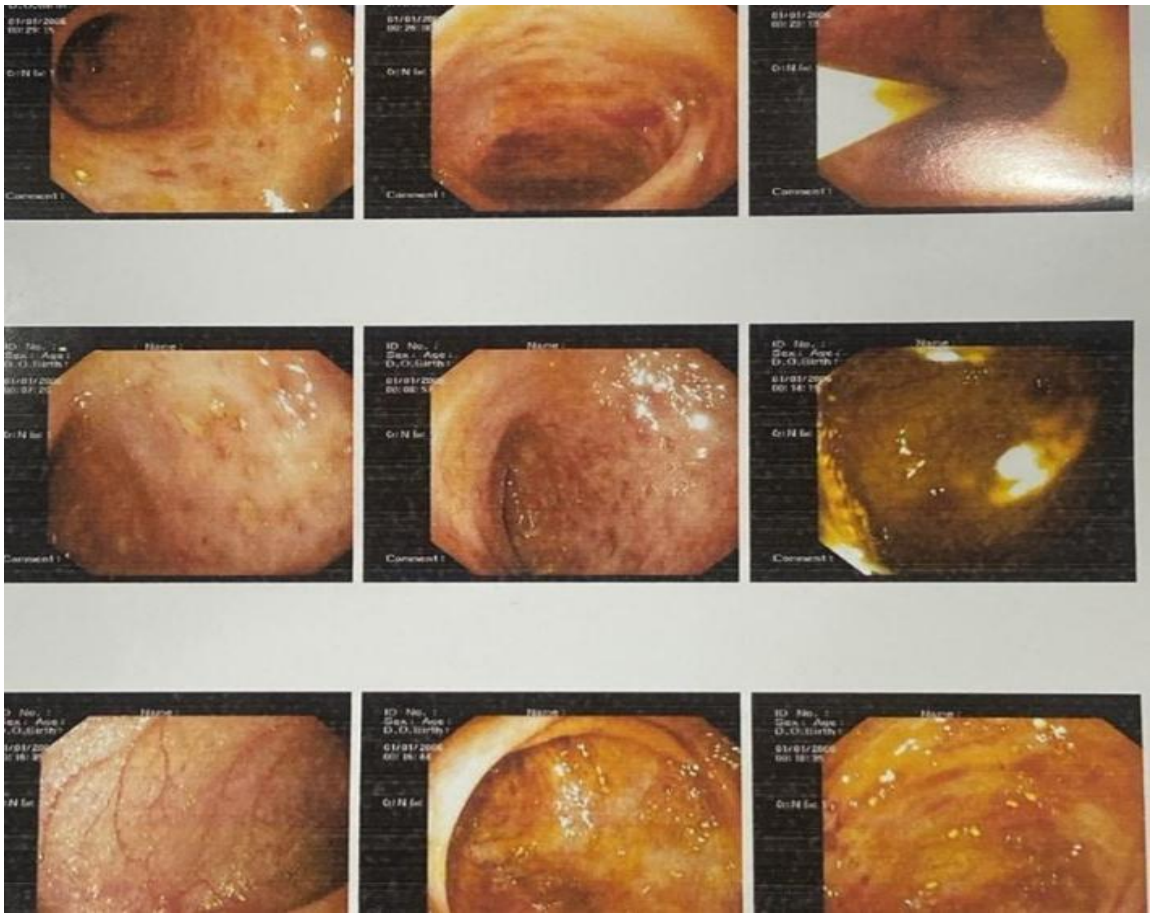


Figure 3: Representative colonoscopic images demonstrating inflammatory and ulcerative changes. The original patient-identifying header was cropped for manuscript use.

DISCUSSION

Sweet syndrome is a rare reactive cutaneous manifestation of inflammatory bowel disease. It classically presents with acute fever, neutrophilia and tender erythematous papules, plaques or nodules; histopathology typically demonstrates a dense dermal neutrophilic infiltrate, often with papillary dermal oedema.[3,4] In a systematic review of Sweet syndrome associated with IBD, fever, painful polymorphic lesions, elevated inflammatory markers and neutrophil-rich skin biopsies were frequent findings. However, the evidence base is dominated by adult reports, and pediatric cases remain sparse.[5] The present child showed a similar clinical pattern, with prolonged fever, neutrophilic leukocytosis, markedly raised CRP and biopsy-confirmed neutrophilic dermatosis.

The present case also illustrates the diagnostic overlap between Sweet syndrome and severe infection. Persistent high-grade fever, markedly elevated inflammatory markers and pustular skin lesions led to broad-spectrum

antimicrobial treatment. As in published pediatric reports, Sweet syndrome may initially be suspected to represent infection; failure to improve with antibiotics, a negative infectious evaluation and the demonstration of a sterile neutrophilic infiltrate should prompt consideration of an inflammatory dermatosis associated with IBD.[6,7] In our patient, the skin lesions and fever persisted despite antimicrobial therapy and improved rapidly after systemic corticosteroid treatment.

The gastrointestinal diagnosis was based on the overall clinicoradiologic and endoscopic picture rather than on the post-treatment biopsy alone. Continuous superficial ulceration extending from the rectosigmoid region to the caecum, associated caecal inflammation and backwash ileitis, together with CT evidence of extensive colitis, supported UC. The revised ESPGHAN Porto criteria identify continuous colonic inflammation beginning distally, with backwash ileitis as a possible accompanying feature in extensive colitis, as findings compatible with UC; they also recognize that pediatric UC may show atypical or less established histological features.[8]

In this case, the colonic biopsy was obtained after prednisolone had already been initiated and showed focal active colitis, cryptitis and crypt distortion without granulomas or prominent basal plasmacytosis. This timing is an important limitation when interpreting the histology. Accordingly, the relatively nonspecific biopsy findings were considered alongside the clinical, radiological and endoscopic evidence rather than in isolation. The association between Sweet syndrome and UC has been described in both adult and pediatric literature, although individual reports are uncommon. A pediatric case report described Sweet syndrome in active UC, while other reports have documented drug-associated Sweet syndrome in children receiving azathioprine; therefore, medication exposure should also be reviewed when evaluating a child with IBD and new neutrophilic skin lesions.[7,9]

In the present patient, the skin manifestations occurred before initiation of mesalamine and prednisolone, and there was no reported preceding thiopurine exposure. The rapid improvement of fever and skin lesions after systemic corticosteroid therapy, together with improvement in gastrointestinal symptoms on prednisolone and mesalamine, was consistent with an inflammatory process. Current IBD guidance classifies Sweet syndrome among reactive cutaneous EIMs and recommends dermatology involvement and control of the underlying intestinal inflammation.[2,10,11]

This case adds to the limited pediatric literature by describing Sweet syndrome as the presenting clue to newly recognized UC, rather than as a complication arising during established disease or after immunomodulator exposure. A previous adult case report and recent pediatric reports similarly emphasize that Sweet syndrome may accompany active intestinal inflammation and can be mistaken for infection.[6,7,12] The case reinforces the value of multidisciplinary assessment involving pediatrics, gastroenterology, dermatology and pathology when infection does not adequately explain prolonged fever and cutaneous inflammation. Early recognition may reduce unnecessary antimicrobial exposure and facilitate timely treatment of both intestinal disease and its extraintestinal manifestation.

CONCLUSION

This case highlights the diagnostic challenge of pediatric ulcerative colitis presenting initially as prolonged fever with sweet syndrome. Persistent fever despite broad-spectrum antibiotics, a negative infectious work-up, characteristic radiological findings, colonoscopic evidence of continuous colitis with backwash ileitis, and skin biopsy demonstrating neutrophilic dermatosis were pivotal in establishing the diagnosis. Sweet syndrome is a rare reactive cutaneous EIM of IBD, and pediatric reports remain limited. Early multidisciplinary evaluation and treatment of the underlying inflammatory disease resulted in favorable clinical recovery. Clinicians should maintain a high index of suspicion for IBD in children with prolonged fever, colitis and unexplained neutrophilic skin lesions.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Bansal R, Inamdar P, Ponathil A. When skin speaks for the gut: pediatric ulcerative colitis presenting as Sweet syndrome. Int J Contemp Pediatr 2026;13:2604-8.