2-22-2022

Bowel-Associated-Dermatosis-Arthritis Syndrome (BADAS) as early presentation of ulcerative colitis in an adolescent girl

Hannah Russell Johns

Nayha Shetty
Henry Ford Health, nshetty1@hfhs.org

Joshua Cash

Tejesh Patel

Crystal Pourciau

Recommended Citation
Johns HR, Shetty N, Cash J, Patel T, and Pourciau C. Bowel-Associated-Dermatosis-Arthritis Syndrome (BADAS) as early presentation of ulcerative colitis in an adolescent girl. Pediatr Dermatol 2022.

DOI: 10.1111/pde.14954

Pediatric Dermatology WILEY

BRIEF REPORT

Bowel-Associated-Dermatosis-Arthritis Syndrome (BADAS) as early presentation of ulcerative colitis in an adolescent girl

Abstract

A 14-year-old girl presented with fevers, joint pain, leukocytosis, and painful, fluctuant skin lesions, preceded by a 2-week history of abdominal cramping and diarrhea. Workup revealed bowel-associated-dermatosis-arthritis syndrome (BADAS) in the setting of ulcerative colitis, a rare finding in the pediatric population.

1 | CASE REPORT

A 14-year-old girl presented with several, painful, violaceous nodules, and indurated plaques on the upper and lower extremities after a 1-month history of malaise, fever, and migratory arthritis, preceded by 2 weeks of abdominal cramping and bloody diarrhea. The cutaneous lesions began as erythematous, indurated, subcutaneous nodules, some of which expanded into hemorrhagic abscesses (Figure 1).

Laboratory investigation revealed worsening anemia (Hgb 8.2 g/dl, Hct 26.4%), leukocytosis with neutrophilia (WBC 23.4 × 10^3/μl and ANC 19.4 × 10^3/μl), elevated CRP (193.2 mg/L), and increased ESR (104 mm/h). Initial colonoscopy exhibited eosinophilic colitis from the rectum to the sigmoid colon. Punch biopsy from a nodule edge demonstrated suppurative panniculitis with septal and lobular inflammatory infiltrate characterized by predominant neutrophils, lymphocytes, eosinophils, and histiocytes that stained negative for CD15 and myeloperoxidase but positive for CD163 (Figure 2A,B). There was no evidence of vasculitis. Tissue cultures were negative. A diagnosis of bowel-associated-dermatosis-arthritis syndrome (BADAS) in the setting of possible inflammatory bowel disease (IBD) was established. The patient was given two doses of methylprednisolone at 40 mg (0.6 mg/kg) followed by a prednisolone taper and sulfasalazine, with rapid improvement of her skin, gastrointestinal, and joint symptoms. A few months later, she was readmitted for abdominal pain and hematochezia. Repeat colonoscopy confirmed ulcerative colitis (UC). Subsequently, the patient's cutaneous, gastrointestinal, and joint symptoms were well-controlled with sulfasalazine and infliximab.

2 | DISCUSSION

BADAS is a neutrophilic dermatosis typically characterized by recurrent erythematous papules and vesiculopustules in the setting of IBD or bowel bypass surgery.[1] Atypical cutaneous manifestations of BADAS include nodules, plaques, and abscesses.[2] Diagnosis of BADAS can be challenging as there are no established diagnostic criteria, and it is known to be a mimicker of several other dermatoses such as aseptic abscess syndrome (AAS), Sweet's syndrome (SS), pyoderma gangrenosum, panniculitis, and hidradenitis suppurativa.[3]

Tender erythematous plaques and nodules noted in our patient are often observed in SS; however, diarrhea and polyarthralgia are more consistent with clinical findings of BADAS.[3] Although

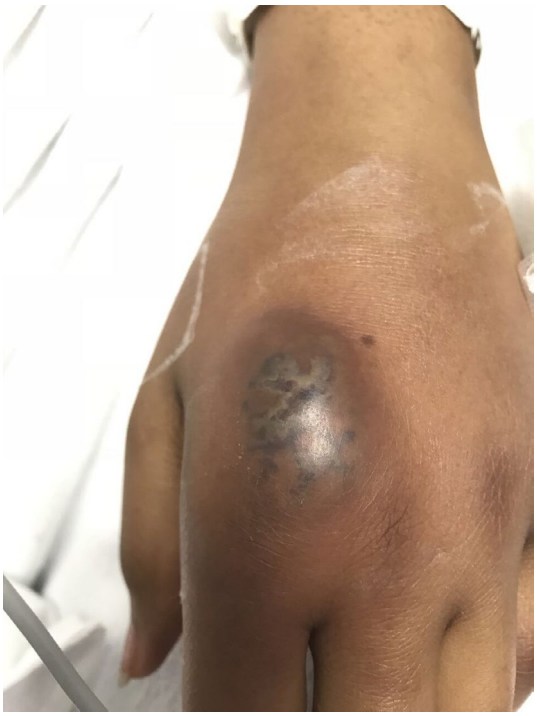

FIGURE 1 On the dorsum of the left hand, there is an exquisitely tender, fluctuant nodule with surrounding erythema and edema

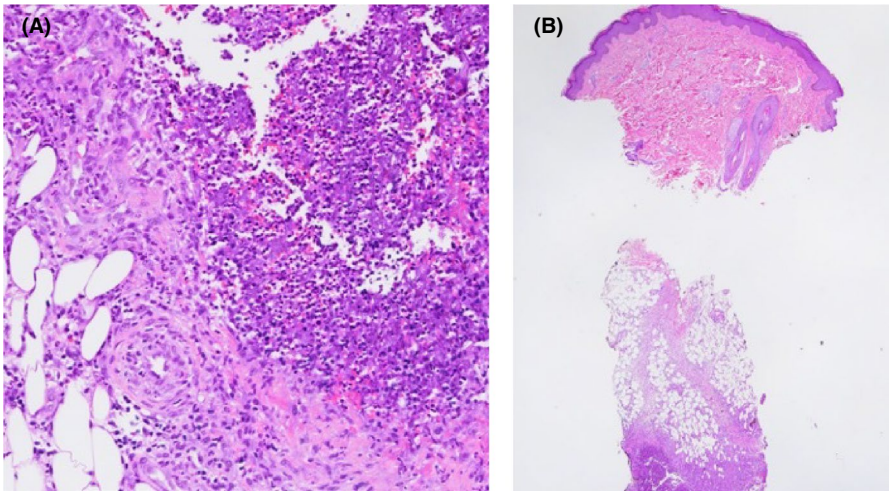

FIGURE 2 A, Histopathology shows panniculitis with a septal and lobular inflammatory infiltrate of small abscesses, eosinophils, and histiocytes with positive staining for CD163 and negative staining for both CD15 and myeloperoxidase (H&E, ×40); B, Low power view of figure showing the depth of infiltrate (H&E, ×2). Photographs are courtesy of Dr. Maria Tirado Gonzalez

cutaneous abscesses and neutrophilic infiltrate extending into the hypodermis, as seen in our patient, can be observed in AAS, the absence of abscesses in viscera such as the spleen, pancreas, and lungs made the diagnosis of AAS less likely.[2] BADAS has histologic manifestations consistent with neutrophilic infiltration that can extend from the epidermis into the adipose tissue and septa.[2] A diagnosis of BADAS associated with IBD was supported by skin eruptions coinciding with polyarthralgia, fever, diagnosis of UC, pathology demonstrating predominant neutrophilic infiltrates without evidence of infection, and rapid improvement with steroids and UC treatment.

BADAS in the pediatric population is extremely rare. In contrast to our patient, previous pediatric BADAS cases were diagnosed with IBD prior to the presentation of BADAS.[1,4] BADAS has been reported with the initial presentation of Crohn's disease (CD) in an adolescent,[5] but to our knowledge, it has not been reported concurrently with UC diagnosis. Cutaneous manifestations of IBD are much less common in UC compared to CD.[2] As IBD becomes more prevalent in pediatric patients, it is important to recognize BADAS as a potential complication associated with IBD in children.

KEYWORDS

arthritis, Crohn's disease, dermatitis, inflammatory bowel disease, ulcerative colitis

ACKNOWLEDGMENTS

We would like to thank Dr. Maria Tirado Gonzalez for the production of the histopathology photographs used in this case.

CONFLICT OF INTEREST

Hannah Johns, B.S., Nayha Shetty, MD; Joshua Cash, MD; Tejesh Patel, MD, and Crystal Pourciau, MD certify that they have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.

CONSENT FOR PUBLICATION

This manuscript has been reviewed and approved for submission by all authors.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

Hannah Russell Johns BSc[1]
Nayha Shetty MD[2]
Joshua Cash MD[1]
Tejesh Patel MD[1]
Crystal Pourciau MD[1]

[1]*Department of Dermatology, University of Tennessee Health Sciences Center, Memphis, Tennessee, USA*
[2]*Department of Dermatology, Henry Ford Health System, Detroit, Michigan, USA*

Correspondence
Crystal Pourciau MD, MPH, FAAD, 49 N. Dunlap St. Suite 103, Memphis, TN 38105, USA.
Email: cpourcia@uthsc.edu

ORCID

Hannah Russell Johns https://orcid.org/0000-0002-2489-3734
Nayha Shetty https://orcid.org/0000-0002-8309-2654
Crystal Pourciau https://orcid.org/0000-0003-4678-1183

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