

Bowel-associated dermatosis arthritis syndrome: A case report with first positron emission tomography analysis



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Key words: arthralgia; bacterial overgrowth; bowel-associated dermatosis-arthritis syndrome; positron emission tomography; skin eruption.

INTRODUCTION

Bowel-associated dermatosis-arthritis syndrome (BADAS) is a rare syndrome that was described for the first time in 1971¹ in patients who received bypass obesity surgery,² taking the name of *bowel bypass syndrome*. Similar cases were reported with patients who had inflammatory bowel disease³ leading to the name of *BADAS*. It causes both arthralgia and skin eruptions, which, in context of bowel disease, has to put us in mind of the possibility of the diagnosis. Here we report a case of a positron emission tomography (PET) scanner description.

CASE REPORT

A 46-year-old woman presented with red skin eruptions and inflammatory polyarthralgia. She had no medical history except by-pass surgery for obesity 8 years before. Symptoms suddenly started in March 2017 with inflammatory arthralgia and swelling of the digital joints accompanied by epigastric pain. The patient went to the emergency department and started 20 mg/d of prednisone, which led to substantial clinical improvement. She received prednisone for one week. After prednisone was stopped, she developed a new flare of symptoms during the next 72 hours. At that time, arthralgia reached the knees and wrists. Ultrasonography found edema with positive Doppler signal confirming arthritis in metacarpophalangeal and metatarsophalangeal joints. At the same time, a painful skin eruption developed over the face, legs, and arms and consisted of erythematous macules and papules with a diameter of about 1 cm (Fig 1). There was neither pruritus nor fever.

Abbreviations used:

BADAS: bowel-associated dermatosis-arthritis syndrome
PET: positron emission tomography



Fig 1. Skin eruption on arms and legs of a 46-year-old woman.

Blood investigations found increased white cell count ($11.8 \times 10^9/L$, differential including mild neutrophilia at $8.3 \times 10^9/L$) and C-reactive protein level of 71 mg/L. The following investigations were performed and had normal results: serum electrolytes, liver markers, blood culture, test for *Mycobacterium tuberculosis*, Lyme disease serology, HIV and hepatitis C and B virus serologies, B9 and B12 vitamin deficiency, anti-nuclear, DNA, anticyclic citrullinated peptide, antimodified citrullinated vimentin, antiextractable nuclear antigen, antineutrophil cytoplasmic, anti-Saccharomyces cerevisiae antibodies, antibodies for celiac disease, angiotensin-converting enzyme, fecal calprotectin, thyroid-stimulating

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Funding sources: None.

Conflicts of interest: None disclosed.

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JAAD Case Reports 2019;5:140-3.

2352-5126

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<https://doi.org/10.1016/j.jidcr.2018.10.025>

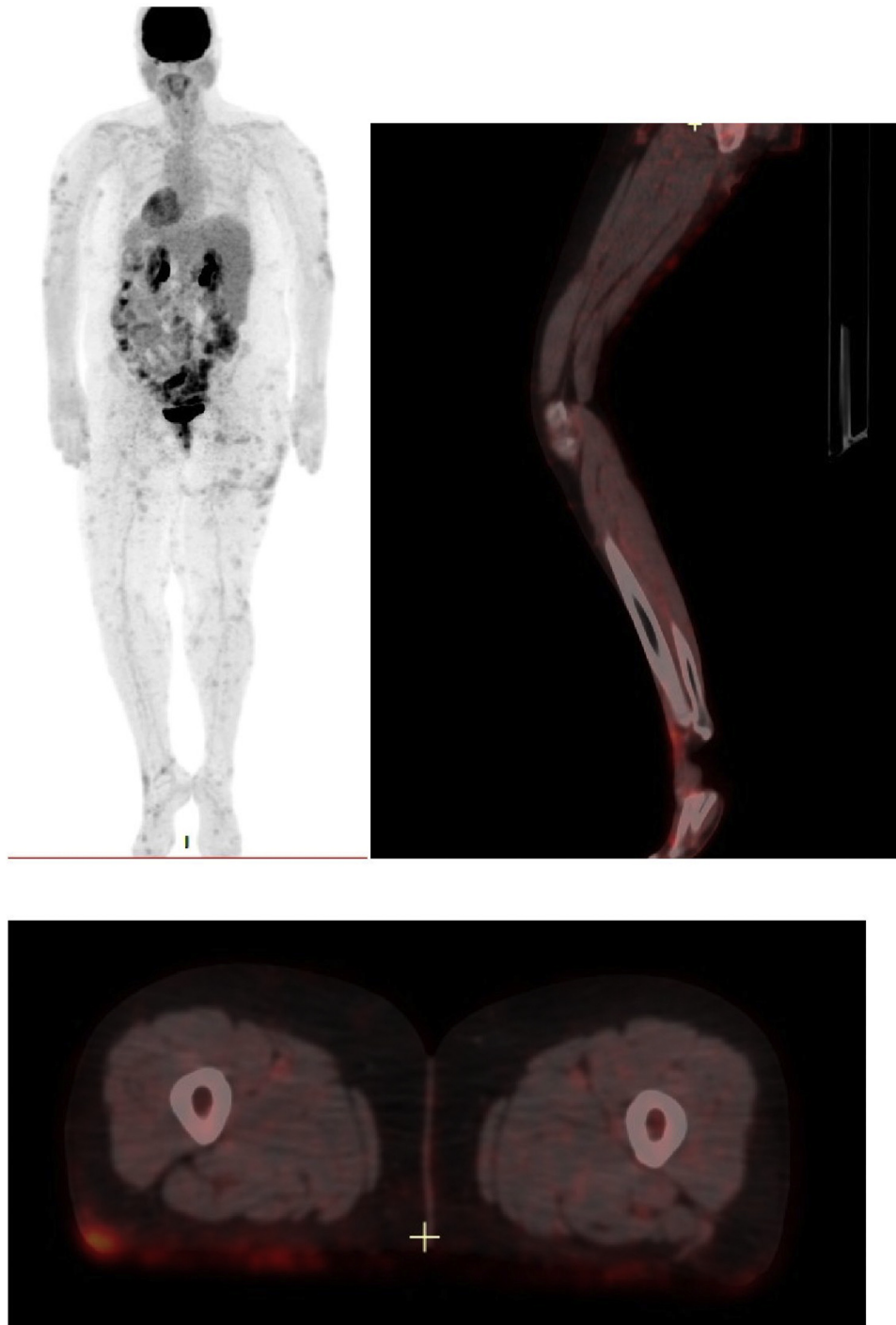


Fig 2. PET scan shows multiple skin lesions.

hormone, transthoracic echocardiography, and gastrointestinal endoscopy with biopsies.

The patient was hospitalized, and PET scan was performed to rule out atypical polymyalgia rheumatica. The analysis found hypermetabolism of multiple disseminated skin thickenings over the trunk

(with posterior predominance), upper limbs, and lower limbs (Fig 2) with no lesions in the stomach or bowel.

A skin biopsy of the right thigh showed a rather dense inflammatory infiltrate consisting of lymphohistiocytes associated with numerous neutrophils with

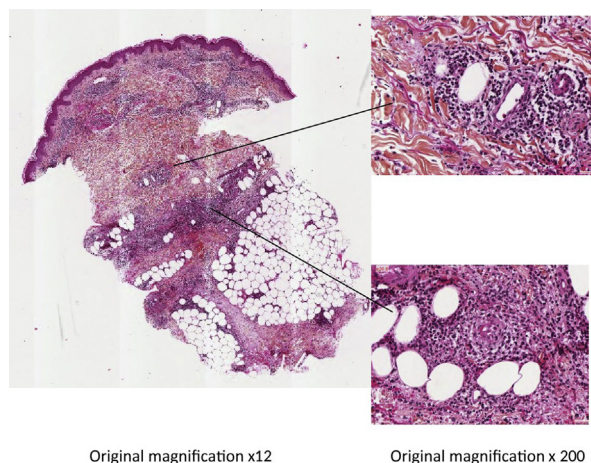


Fig 3. Skin biopsy shows dermo-hypodermic inflammatory infiltrate with lympho-histiocytes associated with numerous neutrophils and sometimes leukocytoclastic vasculitis.

fragmented and pycnotic nuclei. The infiltrate topography was interstitial and around thick-walled capillaries and remodeled by fibrinoid necrosis (Fig 3).

A positive small intestine bacterial overgrowth test (hydrogen-methane breath test with lactulose as the substrate) confirmed bacterial overgrowth. All these elements led to the diagnosis of BADAS caused by the bypass surgery the patient received 8 years earlier.

DISCUSSION

BADAS is a noninfectious neutrophilic dermatosis that often clinically presents with rash, fever, and arthralgia. This syndrome occurs in multiple conditions, including intestinal surgery and inflammatory bowel diseases, but 1 case was also reported in a patient with appendicitis.⁴ The syndrome can occur in up to 20% of patients who have undergone jejunioileal bypass surgery.³ Except for 1 case of fulminant BADAS with necrotizing fasciitis-like disease,⁵ the syndrome is generally benign. The most common explanation for BADAS is a bacterial overgrowth in the bowel loop (caused by bypass or inflammatory disease), which leads to inflammation, immune response to bacterial antigens, and, therefore, release of immune complexes consisting of antibodies to *Escherichia coli* and *Bacteroides fragilis*,⁶ in the blood leading to the systemic manifestations. Peptidoglycan, the polymer responsible for bacterial wall rigidity, seems to be responsible for this disease. Indeed, in animal models, similar skin lesions were induced after administration of purified peptidoglycan.⁷ A similar clinical presentation of BADAS was in a 29-year-old patient with a small intestine bacterial overgrowth,⁸ which is another argument implying a bacterial overgrowth in the pathophysiology.

Diagnosis may be difficult because no key test is available. Blood tests find a nonspecific

inflammatory syndrome, skin biopsies find lesions similar to those of Sweet syndrome, and symptoms are nonspecific. The small intestine bacterial overgrowth test remains the reference test to confirm the bacterial overgrowth. We report for the first time the use of PET imaging in BADAS. In our case, PET scan was first performed to eliminate other diagnoses but it also provided a characterization of the skin lesions and may help in the choice of lesion for the skin biopsy. PET should not be used for every suspected case of BADAS, but it can be helpful when the diagnosis is uncertain. Treatment for BADAS lacks consensus, but because the pathophysiology implies both an infectious and inflammatory mechanism, the most common treatment consists of antibiotics such as tetracycline or metronidazole and nonsteroidal anti-inflammatory drugs with sometimes cyclosporin or mycophenolate mofetil⁹ to reduce the steroid dose. The treatment efficiency is good, even if new flares can occur. Finally, for BADAS caused by bypass surgery, restoration of normal bowel anatomy, with another surgical operation eliminating the bowel loop, has also been curative in many cases.¹⁰

CONCLUSION

With the increasing incidence of obesity (which has led to more frequent bariatric surgery) and inflammatory bowel diseases, BADAS will probably be an emerging disease. Commonly, it causes arthralgia and skin eruption without vital involvement. Its pathophysiology implies a bacterial overgrowth with an immune response. Thus, the most common treatment, despite no clear recommendations, consists of both antibiotic and anti-inflammatory drugs and in some cases, reversal of the bypass.

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