

CASE REPORT

An unusual cause of reactive arthritis with urticarial: A case report

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ABSTRACT

Reactive arthritis (ReA) is a syndrome of arthritis and tenosynovitis with defined extra-articular manifestations following certain infections. Despite being recognized a long time ago, debates still surrounds its definition. It is still unclear if the spectrum of the disease should include arthritis induced by other than the classical organisms. Here, we present an unusual cause of ReA. A young healthy female patient presented with acute polyarthritis and acute urticaria after 2 weeks of diarrheal illness. She was found to have blastocystis in the stool microscopy. Extensive evaluation ruled out other causes of her arthritis. She received metronidazole with a short course of NSAIDs and steroids with complete resolution of her skin and joint symptoms. She was followed for six months with no recurrence of arthritis or urticaria. Blastocystis sp. is a parasite that is prevalent in developing countries. It has been linked to isolated ReA or isolated urticaria among a few other case reports. This is the very first case to have blastocystis induced ReA that coexisted with acute urticaria. Upon review of the literature, we found that blastocystis induced ReA affects mainly young and middle-aged females such as in our case. The arthritis is usually settled with the parasite eradication. Finally, urticaria might be a distinguishing feature for blastocystis induced ReA that requires specific antimicrobial therapy.

Keywords: reactive arthritis, blastocystis homogeneous, urticaria

BACKGROUND

Reactive arthritis (ReA) was first quoted in the 1770s when Stoll noticed a triad of arthritis, urethritis, and conjunctivitis following dysentery. We have since learnt much about the disease nevertheless, debate still surrounds the disease definition. It was proposed

that the definition of "reactive arthritis" should be used only for specific scenarios where the microbes involved are Chlamydia, Salmonella, Shigella, Campylobacter, and Yersinia. In contrast, the term "infection-related arthritis" is used for arthritides related to or associated with other infections.¹

More and more cases have been reported with a syndrome of arthritis and tenosynovitis with variable extra-articular manifestations following gastrointestinal-related infections other than the classical organisms.² The similarity between these cases and the cases caused by the "classical organisms" may indicate that the ReA spectrum is broader than we think. This is particularly important in developing countries where infectious diseases are much more prevalent. Here, we describe a case of a young lady with ReA that coexisted with urticaria as sequelae from blastocystis infection. Extensive workup excluded other causes of arthritis.

CASE REPORT

An 18-year-old female patient, who was fit and well, presented to the hospital with a three day history of urticarial skin rash that was followed with arthritis and tenosynovitis. Her presentation started with urticaria that failed to improve with antihistamine use. Two days later, she started to have multiple joint pain affecting knees, elbows, shoulders, and ankles in addition to small joints of the right hand. There was no history of preceding insect bite, recent vaccination or medication use. She could not link her symptoms to any particular food. There was no history to suggest any connective tissue disease. The patient had on and off bouts of diarrhea with 5 kg weight loss over five months before her presentation. Two weeks before presenting to the hospital, she had an episode of watery diarrhea with mild abdominal cramps. On examination, there were scattered wheels of urticarial rash. She had tenosynovitis of both the achilles tendons and arthritis with redness, hotness, and detectable effusion mainly in her left wrist and both ankles. In addition to the tenderness of the knees and some of her MCPs.

Her complete blood count, liver and kidney function tests were normal except for mild microcytic anemia. Her inflammatory markers were high with ESR of 55 mm/1st hour and CRP of 35 mg/L. Autoimmune workup, including rheumatoid factor, antinuclear antibody, and anti-cytoplasmic antibody were negative. Her HLA B27 was negative as well as her serology for brucella, parvovirus, hepatitis B, hepatitis C and HIV.

PCR for adenovirus, CMV, EBV was also negative. Urine analysis was normal. Her iron profile showed evidence of iron deficiency anemia with hemoglobin of 9.1 gm/dL. Her stool microscopy showed blastocystis cysts. The patient was started on oral metronidazole 750 mg three times per day for ten days. Her symptoms were controlled by naproxen and prednisolone 30 mg daily, which were tapered off after two weeks with complete resolution of her joint and skin symptoms. Upper and lower endoscopies were done to rule out inflammatory bowel disease (IBD) and were unremarkable except for mild nodular gastritis. Biopsies from the duodenum, ileum and colon showed normal histological appearance. The patient's diarrhea later improved, and she gained 4 kg after five months of follow up with normalization of the inflammatory markers.

DISCUSSION

Blastocystis sp. is an enteric parasite that is particularly endemic in developing countries. It has a fecal-oral route of transmission and causes non-specific gastrointestinal symptoms such as diarrhea, abdominal pain, flatulence, nausea, vomiting, constipation, weight loss and fatigue.³ There is growing evidence of a link between blastocystis infection and urticarial.⁴ In one study, the prevalence of blastocystis infection among patients with acute or chronic urticaria was found to be 60% compared to 8% in healthy controls.⁵

Blastocystis was rarely encountered as the causative organism for ReA. However, among a few reported cases, the disease was found to be self-limited, affecting mainly females with improvement after parasite eradication⁶⁻⁸ (Table 1). Rarely, the parasite can cause invasive infection and septic arthritis in immunocompromised patients.⁹ Our case is the very first to have blastocystis induced ReA that coexisted with urticaria. A few cutaneous manifestations are associated with ReA including erythema nodosum, keratoderma blenorrhagica and circinate balanitis.¹⁰ The presence of urticaria might be a feature that might distinguish ReA induced by blastocystis from other infectious causes.

CONCLUSION

This report shed light on an unusual cause of ReA and urticaria. It is important to consider organisms other than the classical, as a trigger for ReA, especially in the developing countries. Here we report the first case of ReA with urticaria caused by blastocystis sp.

Table 1. Cases of ReA triggered by Blastocystis Spp.

Case	Year	Age	Sex	Gastroenteritis in relation to arthritis	The pattern of joints involved	Urticaria	Uveitis	Treatment	Prognosis
Our case	2019	18	F	Chronic with an episode of diarrhea two weeks before arthritis	Asymmetrical polyarthritis with back involvement	Yes	No	Metronidazole with short course of NSAIDs and steroids	Lasted for 2 weeks with complete resolution and no recurrence with 6 months follow up
Tejera et al. ⁶	2012	45	F	Acute episode 10 days prior to arthritis	Knee monoarthritis	No	Yes	Metronidazole/ Tinidazole with short course of NSAIDs	Complete resolution
Krüger et al. ⁷	1994	46	F	Chronic gastroenteritis	Oligoarthritis	No	NA	NSAIDs and steroids failed to control the arthritis, however, improved with Metronidazole.	Complete resolution. No follow is available
Lakhanpal et al. ⁸	1991	46	F	3 weeks prior to arthritis	Asymmetrical polyarthritis predominately lower limb with back involvement	No	No	Metronidazole and short course of NSAIDs	Lasted less than a month and had no recurrence with follow for around one year.

F: Female; NA: Not Available; NSAIDs: Non-Steroidal Anti-Inflammatory Drugs;

the patient had a recurrence of ocular involvement and had a repeat stool test which was positive for blastocystis later receiving a second course of tinidazole and gad topical treatment.

Disclosure statement

None of the authors have any conflict of interest to disclose.

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Ethics and consent

The present study was approved by the Ethics Committee of Hamad Medical Corporation (protocol number: MRC-04-19-234). Written informed consent was obtained from the patient according to the Declaration of Helsinki.

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