



## Case report

# Large abscess formed in the abdominal wall by *Mycobacterium avium* complex: A case of unmasking immune reconstitution inflammatory syndrome



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## ARTICLE INFO

## Article history:

Received 7 July 2019

Received in revised form 23 July 2019

Accepted 23 July 2019

## Keywords:

Immune reconstitution inflammatory syndrome

*Mycobacterium avium* complex

Acquired immunodeficiency syndrome

Abscess

## ABSTRACT

Immune reconstitution inflammatory syndrome (IRIS) is a clinical entity with a broad presentation that is complicated in patients with acquired immunodeficiency syndrome after initiating antiretroviral therapy. A 51-year-old Japanese man was diagnosed with disseminated *Mycobacterium avium* complex (MAC) infection presenting as unmasking IRIS, which formed a large abscess in the patient's abdominal wall. MAC-IRIS commonly involves the lymph nodes, bone marrow, and gastrointestinal tract. To our knowledge, this is the first case report of an abdominal wall abscess caused by MAC-IRIS.

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## Introduction

Paradoxical worsening of treated infections or unmasking of previously subclinical infections after introducing antiretroviral therapy (ART) has been recognized as immune reconstitution inflammatory syndrome (IRIS). IRIS typically involves infectious diseases, including *Mycobacterium avium* complex (MAC), tuberculosis, and progressive multifocal leukoencephalopathy. The development of an abdominal wall abscess due to MAC-IRIS has not been previously reported [1,2].

## Case report

A 51-year-old Japanese man was diagnosed with *pneumocystis jiroveci* pneumonia (PJP) with human immunodeficiency virus (HIV) infection in a regional hospital. His CD4 cell count was 15 cells/ $\mu$ L and HIV RNA count was 150,000 copies/mL. The PJP was successfully treated with administering 960 mg trimethoprim/

sulfamethoxazole for 3 weeks. An ART regimen comprising tenofovir disoproxil fumarate, emtricitabine, and efavirenz was initiated. Unfortunately, MAC prophylaxis was not given.

On the 39<sup>th</sup> day of the ART treatment, 40 mg of prednisolone was given without the evaluation for the origin of a persistent fever. The patient became afebrile after initiating corticosteroid therapy which was tapered and then discontinued over 3 months. Subsequently, 3 months later, bilateral neck lymph nodes swelling appeared. Cefazolin was administered without culture evaluation. The therapy was not effective. Lymph nodes swelling persisted, and an abscess had formed. No intervention was performed for the abscess. The patient subsequently became febrile 4 months later and experienced abdominal fullness, and was referred to our institution.

His CD4 cell count was 22 cells/ $\mu$ L and HIV RNA count was 20 copies/mL. CT revealed a large mass in the abdominal wall and multiple intra-abdominal lymph node swellings (Fig. 1). The fine-needle aspiration examination revealed 10 bacteria per field by acid-fast bacteria stain, however, were culture negative. *Mycobacterium intracellulare* was positive by polymerase chain reaction (PCR) examination on the abscess fluid. The abdominal rectus muscle was open and the abscess was incised and drained. Furthermore, CT revealed destruction of the lumbar vertebrae and

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**Fig. 1.** Abdominal CT revealing a large abscess in the abdominal wall. The abscess was located under the abdominal rectus muscle and was separated from intra-abdominal lymph node swellings.

the intervertebral disk as well as the surrounding abscess formation, which was compatible with pyogenic spondylitis. He was diagnosed with disseminated MAC infection and was administered 800 mg of clarithromycin, 875 mg of ethambutol, and 450 mg of rifabutin each daily and ART was continued.

Nine months later, the patient complained of lumbago while on MAC therapy. His CD4 cell count was 80 cells/ $\mu$ L and HIV RNA count was 89 copies/mL. CT revealed a recurrence of the abscess in the abdominal wall. He further developed arthritis in the lumbar, shoulder, and sternoclavicular joints, as detected by gallium scintigraphy (Fig. 2). The abscess was incised and drained. The acid-fast bacteria stain revealed 5 bacteria per field. *Mycobacterium intracellulare* was positive by PCR examination on the abscess fluid. MAC therapy and ART were continued, and his back pain was reduced gradually.

Two years later, his back pain improved. No recurrence of the abdominal abscess was revealed by CT. His CD4 cell count was 74 cells/ $\mu$ L and HIV RNA count was 78 copies/mL. However, his creatinine level had increased to 1.49 mg/dL, which suggested that his renal function had worsened. Therefore, MAC therapy was switched to 600 mg of azithromycin and 750 mg of ethambutol. Presently, the patient's condition is stable.

## Discussion

The incidence of disseminated MAC infection presenting as unmasking IRIS is 3.5% among patients who are initiated with ART with a base-line CD4 cell count of <100 cells/ $\mu$ L [3]. MAC-IRIS commonly involves the lymph nodes, bone marrow, and gastrointestinal tract [4–6]. In this case, MAC-IRIS could have developed on the 39<sup>th</sup> day of the ART treatment, when the fever continued for a week. In a prospective study the patients developed IRIS in a median of 48 days (29–99 days) [7]. Because the patient was



**Fig. 2.** Nine months after the diagnosis of disseminated MAC infection, left sternoclavicular joint was swollen.

diagnosed and treated late, MAC infection was disseminated and finally formed an abscess in the abdominal wall. Early diagnosis is important improve the outcome. Abscesses rarely develop in the abdominal wall. To our knowledge, this is the first case report of an abdominal wall abscess caused by MAC-IRIS.

MAC-IRIS is commonly observed in patients with low CD4 cell counts [8]. It is often self-limited, with favorable outcomes in most cases when MAC is suitably treated. However, despite advances in ART, MAC-IRIS remains a therapeutic challenge in some cases, especially in intra-abdominal diseases [9]. It has been suggested that the involvement of the abdominal rather than that of the peripheral lymph nodes has a poor outcome [1]. This could be because the burden of mycobacterial organisms in lymph node tissues is higher in these cases. Therefore, surgical resection, including drainage, may be necessary in addition to anti-MAC medical management for such cases. In the present case, resection of the abscess was not performed at the first visit, which may explain the requirement of a second drainage in nine months after the initial drainage.

The therapeutic response and outcome of MAC-IRIS is influenced by an ART-associated CD4 cell count increase [3]. In our case, the CD4 cell count was 22 cells/ $\mu$ L at the time of MAC-IRIS diagnosis. The CD4 counts did not increase significantly despite continuation of ART. This could be the reason for the recurrence of the abscess after 9 months when the CD4 cell count increased to 80 cells/ $\mu$ L. The median CD4 cell count at MAC-IRIS diagnosis has been reported to be 120 cells/ $\mu$ L [3]. The MAC therapy has been continued because the patient's CD4 cell count is still not sufficient high (78 cells/ $\mu$ L in April 2018). The median CD4 cell count at the completion of MAC therapy is reported to be 230 cells/ $\mu$ L [3]. In contrast to that in the case of tuberculosis, it is difficult to eradicate MAC only using antimicrobial therapy. Additionally, immune reconstitution is necessary to cure MAC infection.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request".

## CRedit authorship contribution statement

**Narika Yanagisawa:** Writing - original draft. **Seisho Takeuchi:** Conceptualization, Writing - review & editing. **Miho Nakamura:** Visualization. **Yukitaka Yoshida:** Resources. **Katsuji Teruya:** Writing - review & editing, Supervision. **Kiyonori Takada:** Writing - review & editing, Project administration.

## Declaration of Competing Interest

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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