〈Case Report〉

# *Mycobacterium heckeshornense*-induced deep abscess in the gluteus maximus muscle: a case report and review of the literature

**Hirotoshi Kikuchi[1,*], Kurumi Asako[1], Hajime Kono [1], Miwa Asahara[2], Takashi Tanaka[2], Go Kamosida[3], Takane Ueda[3], Shigeru Nagakawa[3], Tsuneyuki Ubagai[3], Yuko Kazumi[4] and Yasuo Ono[1, 3]**

[1] Department of Internal Medicine, Teikyo University School of Medicine, Tokyo, Japan
[2] Department of Central Clinical Laboratory, Teikyo University Hospital, Tokyo, Japan
[3] Department of Microbiology and Immunology, Teikyo University School of Medicine, Tokyo, Japan
[4] The Research Institute of Tuberculosis, Japan Anti-Tuberculosis Association, Tokyo, Japan

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*Mycobacterium heckeshornense* is rarely isolated from a clinical sample. We report a case of a patient with polymyositis in whom a deep abscess formed in the gluteus maximus muscle and in which *M. heckeshornense* was identified by DNA sequencing. Combination therapy of levofloxacin and clarithromycin for 2 years improved inflammatory findings and 5 years have passed without recurrence of the gluteal abscess. Identification of the bacterial strain in nontuberculous mycobacterium (NTM) infection is important to determine the treatment plan and accumulation of data on drug sensitivity is required to establish a therapeutic strategy for rare pathogen such as *M. heckeshornense*.

## Introduction

The incidence of nontuberculous mycobacterium (NTM) infection has shown a tendency to increase in Japan. The rates for individual bacterial strains are 60% for *Mycobacterium avium*, 25% for *M. intracellulare*, and 8% for *M. kansasii*, while other strains including *M. abscessus* are

*Corresponding auther: Tel: +81–3–3964–1211, Fax: +81–3–3964–4707, E-mail: hiro-k@med.teikyo-u.ac.jp

rare[1]. In contrast, the incidence of NTM infection in Europe is highest for *M. avium complex* (MAC), followed by *M. xenopi*[2,3].

Herein, we report a case of a patient with polymyositis in whom a deep abscess formed in the gluteus maximus muscle and in which *M. heckeshornense* is isolated.

## Case Report

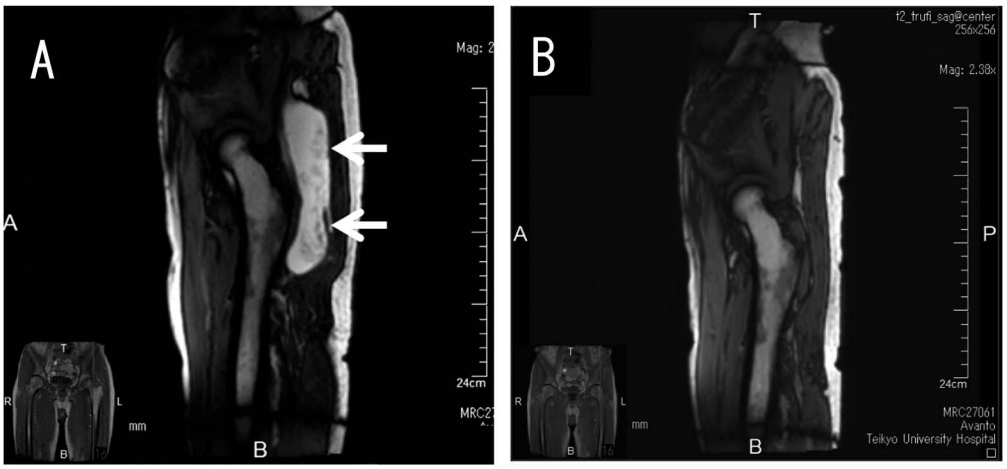
A 62-year-old man developed myalgia at the proximal muscle 10 years ago. An elevated blood creatine kinase (CK) level and positive anti-Jo-1 antibody were found and he was diagnosed with polymyositis based on the results of muscle biopsy. Since the disease redeveloped when the dose of adrenal cortical steroids was decreased, maintenance treatment was provided with prednisolone (12 mg/day), cyclosporine (150 mg/day) and methotrexate (10 mg/week). The patient became aware of an uncomfortable feeling in his left buttock 4 months ago and swelling in the same region 2 months ago.

At his first visit to our hospital, he had a temperature of 36.0℃ with no myalgia, arthralgia, or obvious trauma. Swelling was confirmed from the left buttock to the external side of the left femoral region. A blood test showed a white blood cell count of 15,800/$\mu$L (stab cells 2.0%, segment cells 94.0%, and lymphocytes 4.0%), a C-reactive protein level of 1.90 mg/dL, and an erythrocyte sedimentation rate of 40 mm/h. The levels of CK (69 IU/L), IgG (1,270 mg/dL), IgA (163 mg/dL), IgM (142 mg/dL), antinuclear antibody (titer $<$40), and anti-Jo-1 antibody (142.5 IU/L, positive) were similar to those in a previous examination, suggesting stable polymyositis.

Magnetic resonance imaging of the area from the buttock to the femoral region suggested fluid accumulation in a deep area to the side of the left femoral muscle (Fig. 1). Puncture was performed under ultrasound guidance. A standard plate count of bacteria was negative in purulent aspirate however the concentrating method for tubercle bacillus was 1＋. Chest X-ray and chest computed tomography performed in the same period gave no clear findings other than pulmonary fibrosis associated with polymyositis, and acid fast bacterium was negative in gastric fluid and blood culture. Puncture was performed twice for the abscess and growth of NTM was found in both aspirates on the 15th day. In a DNA-DNA hybridization (DDH) test (DDH Mycobacteria, Kyokuto Pharmaceutical Industrial Co., Ltd, Japan), the bacterium was identified as *M. xenopi*.

*M. xenopi* is rare in Japan and the number of reports on infection of tissues other than the lungs is small [4]. In addition, a drug susceptibility test for the detected bacterium showed sensitivity to ethambutol (Table 1), which is inconsistent with *M. xenopi*. Furthermore, DNA sequencing of 16S rRNA and *rpoB gene* of the bacterium suggested concordance rates with *M. heckeshornense* of 100% and 99%, respectively, while those with *M. xenopi* were 95%. Thus, the NTM was identified as *M. heckeshornense*. After drainage of the abscess, the patient was treated with concomitant 500 mg/day levofloxacin and 400 mg/day clarithromycin for 2 years, and there has been no redevel-

**Figure 1.**




A. Magnetic resonance imaging (sagittal view) of the area from the buttock to the femoral region. Arrow: deep abscess in the gluteus maximus muscle. B. MRI findings after 10 months from treatment are shown. Abscess has disappeared.

**Table 1. Results drug susceptibility testing in our case**

| Antibiotics | | | MIC (mg/L) |
|---|---|---|---|
| Streptomycin | (SM) | Susceptible | |
| Isoniazid | (INH) 0.2 | Resistant | |
| Isoniazid | (INH) 1.0 | Susceptible | |
| Rifampicin | (RFP) | Susceptible | |
| Ethambutol | (EB) | Susceptible | |
| Kanamycin | (KM) | Susceptible | |
| Enviomycin | (EVM) | Susceptible | |
| Ethionamide | (ETH) | Susceptible | |
| Cycloserine | (CS) | Susceptible | |
| Para-aminosalicylate | (PAS) | Susceptible | |
| Levofloxacin | (LVFX) | Susceptible | 0.004 |
| Clarithromycin | (CAM) | Susceptible | <0.016 |

Drug susceptibility tests were determined using a Vite Spectrum-SR kit. (Kyokuto Pharmaceutical Industrial Co., Ltd, Japan). There are no criteria of CLSI (Clinical and Laboratory Standards Institute) in *M. heckeshornense*.

opment of the condition for 5 years without administration of these antibacterial agents.

## Discussion

There are several methods for identification of NTM bacterial strains. In 2011, Morimoto *et al.* suggested that *M. heckeshornense*, which cannot be identified using the DDH kit, may be incorrectly identified as *M. xenopi*[5]. In addition, two case reports showed that a NTM identified as *M. xenopi* by the DDH kit was found to be *M. heckeshornense*, a slow acid fast bacterium [6].

**Table 2. Summary of documented *Mycobacterium heckeshornense* case reports**

| Case no. / author / year | Age (years) / gender | Site of infection | Predisposing condition / complications | Target of identification | Treatment for NTM | | Susceptibilities | Outcome | Reference |
|---|---|---|---|---|---|---|---|---|---|
| | | | | | antibacterial agents | Others | | | |
| 1. Roth *et al*. (2000) | 30 /F | pulmonary (bilateral cavitary) | non | 16S rRNA sequencing +16S-23S spacer | INH + RFP + EB + protinamid + CPFX | surgical resection | S: SM, EB, CPFX, CAM R: INH, RFP | failed | [7] |
| 2. van Hest *et al*. (2004) | 43 /M | pulmonary | pneumothorax AMI | 16S rRNA sequencing | INH + RFP + EB + PZA | surgery | S: RFP, SM, CPFX, CAM I: INH, EB R: AMI | favorable | [9] |
| 3. van Hest *et al*. (2004) | 73 /M | pulmonary | COPD | 16S rRNA sequencing | INH + RFP + CAM + LVFX | | ND | ND | [9] |
| 4. Godreuil *et al*. (2006) | 86 /F | tenosynovitis (hand) | non | 16S rRNA sequencing + *hsp65 gene* sequencing | no treatment | surgery | S: RFP, EB, SM R: INH | favorable | [10] |
| 5. Kazumi *et al*. (2006) | 51 /F | pulmonary | obsolete TB | 16S rRNA sequencing + *robB gene* sequencing | KM + EB + RFP | | S: SM, RFP R: INH, EB | failed | [6] |
| 6. Kazumi *et al*. (2006) | 72 /M | pulmonary | pneumoconiosis | 16S rRNA sequencing + *rpoB gene* sequencing | no treatment | | S: SM, RFP, EB R: INH | favorable | [6] |
| 7. Jauréguy *et al.* (2007) | 65 /F | pulmonary | lobectomy following a traffic accident | reverse hybridization line probe | CAM + MFLX + EB | | S: RFP, AMK, CAM, EB, CPFX | recurrence | [11] |
| 8. Elyousfi *et al*. (2009) | 51 /M | spondylodiskitis, abscesses | Etanercept treatment for RA | 16S rRNA sequencing | CAM + MFLX + RFP | irrigation and | ND | favorable | [12] |
| 9. McBride *et al*. (2009) | 84 /F | axillary lymphadenitis | Squamous cell carcinoma of the | 16S rRNA sequencing + *hsp65 gene* sequencing | no treatment | surgery | ND | favorable | [13] |
| 10. Ahmed *et al*. (2010) | 40 /M | disseminated with bacteremia | HIV lymphoma | 16S rRNA sequencing | INH + CAM + MFLX + rifabutin | | S: RFP, EB, SM, CPFX, CAM, AMK I: INH | favorable | [14] |
| 11. Chan *et al.* (2011) | 76 /M | peritonitis | peritoneal dialysis | 16S rRNA sequencing + *hsp65 gene* sequencing | no treatment | drainage | ND | favorable | [15] |
| 12. Morimoto *et al.* (2011) | 47 /M | pulmonary | COPD | 16S rRNA sequencing + *robB gene* sequencing + *hsp65 gene* sequencing | ND | | S: INH, EB, RFP, CS, LVFX, CAM | favorable | [5] |
| 13. Carpenter *et al*. (2015) | 45 /M | Spinal osteomyelitis | AIDS | 16S rRNA sequencing | INH + RFP + EB + PZA | | S: RFP, EB, CAM R: PZA | favorable | [16] |
| 14, Kurosaki *et al*. (2018) | 39 /W | pulmonary | non | 16S rRNA sequencing + *rob* B gene sequencing + *hsp* 65 gene | RFP + EB + CAM | surgery | S: RFP, EB, CAM | favorable | [17] |
| 15. Yokoyama *et al.* (2018) | 40 /M | pulmonary | Behçet's disease | MALDI-TOF MAS | INH+RFP+EB+ sitafloxacin | | S: SM, EB, KM, RFP, LVFX, CAM | favorable | [18] |
| 16. Douiri *et al.* (2018) | 48 /M | lumber spondylodiscitis | HIV | 16S rRNA sequencing | RFP + EB + CAM+ | | S: RFP, CAM, moxifloxacin | favorable | [19] |
| 17. Present case | 62 /M | deep abscess in the gluteus maximus | polymyositis | 16S rRNA sequencing + *rpoB gene* sequencing | LVFX + CAM | drainage | See Table 1 | favorable | |

F, female; M, male; AMI, acute myocardial infarction; COPD, chronic obstructive pulmonary disease; TB, tuberculosis; RA, rheumatoid arthritis; HIV, human immunodeficiency virus; AIDS, acquired immunodeficiency syndrome; NTM, nontuberculous mycobacteria; MALDI-TOF MAS, matrix-assisted laser desoraption ionization-time of flight mass spectrometry; INH, isoniazid; RFP, rifampicin; EB, ethambutol; CPFX, ciprofloxacin; CAM, clarithromycin;LVFX, levofloxacin; KM, kanamycin; MFLX, moxifloxacin; PZA, pyrazinamide; SM, streptomycin; AMK, amikacin; R, resistant; I, intermediate; S, susceptible; ND, not described.

These reports indicate a problem with the method used to identify bacterial strains.

At first, we used a DDH kit, with which 18 bacterial strains can be identified at one time by hybridization using DNA of a type strain and DNA of a suspected strain. However, the NTM was sensitive to ethambutol (Table 1) and the infection site was not in the lungs, both of which differ from the typical characteristics of *M. xenopi*. Therefore, we performed DNA sequencing for 16S rRNA and *rpoB gene* of the bacterium and identified the strain as *M. heckeshornense*.

*M. heckeshornense* was first reported by Roth *et al.*[7] and has been confirmed in only 17 cases (Table 2)[5～7, 9～19], including our case, because DNA sequencing is required for identification. In recent years, Yokoyama *et al.*[19] diagnosed *M. heckeshornense* using matrix-assisted laser desorption ionization-time of flight mass spectrometry (MALDI-TOF MS). They had also confirmed in the DNA sequence and had reported that MALDI-TOF MS was useful in the diagnosis of *M. heckeshornense*.

A half of these cases (47.1%) had infection at sites other than the lungs. The outcomes were favorable in most cases (improvement: 81.2%, redevelopment: 18.8%) and improvement occurred without administration of antibacterial agents in some cases. Pulmonary infection with *M. xenopi* is often unresponsive to drug treatment alone, and thus surgery is required in many cases[8], however the pathogenicity of *M. heckeshornense* is unclear. van Hest *et al.* suggested that *M. heckeshornense* detected from lung tissues with normal immunity was pathogenic[9], but a therapeutic strategy has not been established because of the small number of reported cases.

In our case, multiple immunosuppressants were used to control the disease activity of polymyositis. Initially we were considering treatment with three drugs such as rifampicin, levofloxacin and clarithromycin, but because the patient refused to increase the amount of steroids associated with rifampicin administration, we treated with two agents with reference to the case report of *M. xenopi*. Since the possibility that the combination of levofloxacin and clarithromycin induces drug resistance of *M. heckeshornense* cannot be denied either, studies on the resistance mechanism of *M. heckeshornense* by clarithromycin are desired.

We conclude that identification of the bacterial strain in NTM infection is important to determine the treatment plan. Accumulation of data on drug sensitivity is required to establish a therapeutic strategy for rare pathogen such as *M. heckeshornense*.

**Conflict of interests**

We declare that we have no conflict of interest.

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