CASE REPORT

Cutaneous melioidosis in a healthy Danish man after travelling to South-East Asia

Jacob Bodilsen,¹ Henrik Langgaard,² Hans Linde Nielsen³¹Department of Infectious Diseases, Aalborg University Hospital, Aalborg, Denmark²Department of Orthopaedics, Hjørring Hospital, Hjørring, Denmark³Department of Clinical Microbiology, Aalborg University Hospital, Aalborg, Denmark

Correspondence to

Dr Jacob Bodilsen, jacob.bodilsen@rn.dk

Accepted 30 December 2014

SUMMARY

A healthy Danish man presented with infected prepatellar bursitis 8 months after being involved in a car accident in Malaysia resulting in exposure of a laceration of his knee to stagnant water. Tissue samples grew *Burkholderia pseudomallei* and diagnostic work up revealed no secondary foci. The patient was successfully treated with surgical debridement and 3 months of oral trimethoprim-sulfamethoxazole. At 6 months follow-up the patient was without relapse.

BACKGROUND

Burkholderia pseudomallei is a Gram-negative bacillus that can wreak havoc in humans. It is particularly endemic in South East Asia and Northern Australia, but it may also be encountered in Central and South America, certain parts of Africa and the Indian subcontinent.¹ However, exposed European travellers are rarely infected. Disease caused by *B. pseudomallei* is called melioidosis and can include multiple abscess formation, pneumonia, genitourinary infections, septic arthritis, osteomyelitis, parotitis and cutaneous ulcers. Studies suggest a potential latency from exposure to symptomatic disease^{1–4} and long-term antibiotic treatment is needed, usually including an initial intravenous phase.⁵ *B. pseudomallei* may therefore pose a particular challenge in travellers returning to non-endemic countries.

To the best of our knowledge, only two cases of primary cutaneous melioidosis in healthy travellers returning to Europe have been reported. They both had a short incubation period of less than 5 weeks and were treated with a lead-in phase of intravenous antibiotic therapy followed by long-term oral consolidation therapy.^{6,7}

We report a case of a healthy Danish traveller returning with cutaneous melioidosis 8 months after relevant exposure in South-East Asia, who was successfully treated with surgical debridement and an oral antibiotic regimen without a lead-in intravenous phase.

CASE PRESENTATION

A 23-year-old man was admitted to the hospital in January 2014 with infected prepatellar bursitis on his right knee. He was a previously healthy Caucasian student without any comorbidities and he used no medication. Eight months prior to admission the patient had travelled to South-East Asia for 3 months, where he had been implicated in a car accident near Kuantan, Malaysia. He was thrown from his vehicle and landed in a ditch of

stagnant water. He was admitted to the local hospital and observed for a concussion and treated with an unknown antibiotic for a laceration of his right knee for 3 days. After 2 weeks, he was admitted to another hospital for surgical revision followed by secondary suture a few days later (figure 1). Antibiotic treatment with ciprofloxacin 250 mg twice daily was also initiated. The wound healed well and he was discharged after 14 days of treatment, and returned to Denmark.

Although the patient had continuous discomfort in his right knee during the next 8 months, he remained free of other signs of infection. However, in late December 2013 he once again developed pain, redness and swelling with a spontaneous perforation of a cutaneous abscess. As the wound progressed to a size of 7×2.5 cm, he was admitted to the orthopaedic department at a local hospital. At admission the patient was afebrile and without tender or enlarged lymph nodes in his inguina. C reactive protein level was 46 mg/L and white cell count was 11.8×10⁹/L. X-ray of the knee revealed soft tissue swelling without involvement of the joint or bones. The patient underwent surgical debridement and a 5 mm fragment of glass was removed from the wound. Tissue samples were sent for culture and the wound was treated with negative pressure wound therapy as well as peroperative and postoperative dicloxacillin. The patient was discharged after 2 days of admission.

By cultivation of the tissue specimens on a 5% horse blood agar plate (SSI Diagnostica, Denmark), Gram-negative, aerobic, motile, rod-shaped bacteria were isolated in monoculture. Preliminary identification based on MALDI-TOF Mass Spectrometry (Bruker Daltonik GmbH, Germany) identified

Figure 1 The wound after secondary suture at the second admission in Malaysia.

CrossMark

To cite: Bodilsen J, Langgaard H, Nielsen HL. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-207340

B. pseudomallei (log-score >2.5). Further analysis of the isolate was performed at the national reference class III laboratory (SSI, Copenhagen) and 16S rRNA sequencing confirmed the identification. Minimum inhibitory concentrations (MICs) were determined and the isolate was susceptible to ceftazidime (MIC 2.0 mg/L), meropenem (MIC 2.0 mg/L) and trimethoprim-sulfamethoxazole (TMP-SMX) (MIC 0.25 mg/L).

The patient was admitted to the Department of Infectious Diseases, Aalborg University Hospital for further evaluation and treatment. A CT of the thorax, abdomen and pelvis revealed no other foci of infection. He was treated with TMP-SMX 800/160 mg twice daily and discharged 5 days later for outpatient follow-up. Negative pressure wound therapy was continued for a total of 17 days and TMP-SMX was continued for 3 months. The patient tolerated the antibiotic treatment well and the wound healed without scarring. At follow-up 6 months later, the patient was without signs of relapse.

DISCUSSION

Cutaneous melioidosis occurs as either a primary or secondary infection with the latter being a presentation of bacteraemic seeding. The recommended treatment of cutaneous melioidosis without disseminated disease includes intravenous ceftazidime for 10 days followed by 3 months of high-dose TMP-SMX.⁵

In a large prospective observational study of 540 cases of melioidosis from Northern Australia, 10% of patients presented with primary cutaneous melioidosis and 2% of patients with secondary cutaneous melioidosis.^{4 5} Primary cutaneous melioidosis was associated with chronic infection (duration of symptoms for more than 2 months), age under 15 years and an occupational exposure to *B. pseudomallei* compared to other forms of melioidosis. It was not associated with diabetes mellitus, alcohol abuse, chronic lung disease or bacteraemia. Only 7% of patients with primary cutaneous melioidosis had disseminated disease and relapses were rare. In that study, nine patients were cured exclusively with an oral antibiotic regimen and one patient even recovered without therapy. In another recent randomised controlled non-inferiority trial from Thailand comparing TMP-SMX and doxycycline combination therapy versus TMP-SMX alone for the maintenance phase of melioidosis treatment, 40 patients were treated solely with oral antibiotics with only one relapse.⁸ Twenty-nine of these patients had cutaneous melioidosis and none had sepsis at presentation (Direk Limmathurotsakul, personal communication). In contrast to these seemingly benign presentations of cutaneous melioidosis, other case reports describe more serious infections including necrotising fasciitis, secondary abscess formation and sepsis.^{9–11}

The patient in our case was infected through a laceration of his knee exposed to stagnant water in an endemic country. The persisting fragment of glass in his wound probably facilitated a prolonged cutaneous infection ultimately manifesting itself as ruptured prepatellar bursitis 8 months after exposure. Besides the exposure the patient had no risk factors for melioidosis.

We are aware of only two other reports of cutaneous melioidosis in healthy returning travellers.^{6 7} Both patients had an incubation period of less than 5 weeks, localised disease without dissemination and were treated with initial intravenous ceftazidime for 10 days followed by oral TMP-SMX for 3 and 5 months, respectively, without relapses. Other reports of

cutaneous melioidosis are in elderly returning travellers with comorbidities,¹² with one case seemingly occurring 62 years after exposure;³ or from endemic areas such as Singapore, Taiwan and China.^{9 10 13 14} Some of these cases were successfully treated with oral amoxicillin/clavulanate and/or doxycycline, although one case of relapse was also reported.³

In summary, our patient had an insidious and delayed onset of cutaneous melioidosis, and he was successfully treated with surgical debridement and oral TMP-SMX. Omission of initial intravenous antibiotics may be a treatment option in uncomplicated cases, but there is a need for further studies to address this issue—preferably from large prospective studies in endemic countries.

Learning points

- ▶ Melioidosis may have an insidious and delayed onset of disease.
- ▶ Cutaneous melioidosis should be considered as a differential diagnosis in returning travellers with relevant exposures—even when they are without risk factors for melioidosis.
- ▶ In uncomplicated cases of cutaneous melioidosis, a lead-in phase of intravenous antibiotics may be omitted.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Wiersinga WJ, Currie BJ, Peacock SJ. Melioidosis. *N Engl J Med* 2012;367:1035–44.
- 2 Cheng AC, Currie BJ. Melioidosis: epidemiology, pathophysiology, and management. *Clin Microbiol Rev* 2005;18:383–416.
- 3 Ngaay V, Lemeshev Y, Sadkowski L, et al. Cutaneous melioidosis in a man who was taken as a prisoner of War by the Japanese during World War II. *J Clin Microbiol* 2005;43:970–2.
- 4 Currie BJ, Ward L, Cheng AC. The epidemiology and clinical spectrum of melioidosis: 540 cases from the 20 year Darwin prospective study. *PLoS Negl Trop Dis* 2010;4:e900.
- 5 Gibney KB, Cheng AC, Currie BJ. Cutaneous melioidosis in the tropical top end of Australia: a prospective study and review of the literature. *Clin Infect Dis* 2008;47:603–9.
- 6 Meckenstock R, Therby A, Marque-Juillet S, et al. Cutaneous melioidosis in adolescent returning from Guadeloupe. *Emerg Infect Dis* 2012;18:359–60.
- 7 Svensson E, Welinder-Olsson C, Claesson BA, et al. Cutaneous melioidosis in a Swedish tourist after the tsunami in 2004. *Scand J Infect Dis* 2006;38:71–4.
- 8 Chetchotisakd P, Chierakul W, Chaowagul W, et al. Trimethoprim-sulfamethoxazole versus trimethoprim-sulfamethoxazole plus doxycycline as oral eradication treatment for melioidosis (MERTH): a multicentre, double-blind, non-inferiority, randomised controlled trial. *Lancet* 2014;383:807–14.
- 9 Wang YS, Wong C-H, Kurup A. Cutaneous melioidosis and necrotizing fasciitis caused by *Burkholderia pseudomallei*. *Emerg Infect Dis* 2003;9:1484–5.
- 10 Teo L, Tay Y-K, Mancor KJF. Cutaneous melioidosis. *J Eur Acad Dermatol Venerol* 2006;20:1322–4.
- 11 Apisarnthanarak A, Anthonont P, Kiratisin P, et al. Photo quiz. A Thai woman with fever and skin lesions. *Clin Infect Dis* 2005;40:988–9, 1053–4.
- 12 Ezzedine K, Heenen M, Malvy D. Imported cutaneous melioidosis in traveler, Belgium. *Emerg Infect Dis* 2007;13:946–7.
- 13 Tran D, Tan HH. Cutaneous melioidosis. *Clin Exp Dermatol* 2002;27:280–2.
- 14 Tzeng WT, Hsieh CS. Recurrent cutaneous melioidosis treated with surgery and antibiotics. *J Plast Reconstr Aesthet Surg* 2009;62:280–1.

Copyright 2015 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow