

World Journal of *Clinical Cases*

World J Clin Cases 2021 May 26; 9(15): 3487-3795



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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Ji-Hong Lin; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

May 26, 2021

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INSTRUCTIONS TO AUTHORS

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<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>

Brucellosis of unknown origin with haemophagocytic syndrome: A case report

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Author contributions: Tian LH and Xiao PP contributed equally; Dong ZG and Chen XY managed the patient in the hospital; Tian LH collected the patient's clinical data; Huang LJ collected the laboratory data; Xiao PP drafted and revised the manuscript; All authors approved the final manuscript.

Supported by Xiamen Medical and Health Guidance Project, No. 3502Z20199137.

Informed consent statement:

Written informed consent was obtained from the patient for the inclusion of his clinical details for publication.

Conflict-of-interest statement: The authors declare that they have no conflicts of interest.

CARE Checklist (2016) statement:

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was

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Abstract

BACKGROUND

Brucellosis is a contagious bacterial disease caused by *Brucella* species, which is a leading zoonotic disease worldwide. Most patients with brucellosis have a clear infection source; however, our case had a rare presentation of secondary haemophagocytic lymphohistiocytosis without any epidemiological history.

CASE SUMMARY

A 50-year-old man was admitted to our hospital with a fever of unknown origin. After laboratory examinations, such as blood culture and bone marrow biopsy, the patient was diagnosed with brucellosis and secondary haemophagocytic lymphohistiocytosis. After antibiotic therapy, the patient was afebrile, and his haemogram recovered to normal, after which he was discharged.

CONCLUSION

Brucellosis cannot be excluded in patients with clinically unexplained fever, even in those without epidemiologic history.

Key Words: *Brucella*; Brucellosis; Haemophagocytic syndrome; Haemophagocytic lymphohistiocytosis; Fever; Case report

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Core Tip: Most patients of brucellosis present with a clear infection source. However,

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Manuscript source: Unsolicited manuscript

Specialty type: Infectious diseases

Country/Territory of origin: China

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): 0
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

Received: October 25, 2020

Peer-review started: October 25, 2020

First decision: January 17, 2021

Revised: January 27, 2021

Accepted: March 15, 2021

Article in press: March 15, 2021

Published online: May 26, 2021

P-Reviewer: Olt S

S-Editor: Zhang L

L-Editor: Filipodia

P-Editor: Zhang YL



our patient showed a rare case presentation of haemophagocytic lymphohistiocytosis with no clear infection source and unremarkable medical history. Our findings suggest that brucellosis cannot be excluded in patients with clinically unexplained fever, even in those without epidemiologic history. To prevent timely exacerbation of the disease, before obtaining the aetiology test results, the administered antibiotics should cover rare pathogens, such as *Brucella*.

Citation: Tian LH, Dong ZG, Chen XY, Huang LJ, Xiao PP. Brucellosis of unknown origin with haemophagocytic syndrome: A case report. *World J Clin Cases* 2021; 9(15): 3649-3654

URL: <https://www.wjgnet.com/2307-8960/full/v9/i15/3649.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v9.i15.3649>

INTRODUCTION

Brucellosis is a zoonotic infectious disease caused by *Brucella* species[1]. Brucellosis has various clinical manifestations, the most common being fever, followed by weakness, hyperhidrosis, myalgias, and arthralgias[2,3]. Most patients come into contact with infected animals or ingest infected meat or unpasteurised milk. However, unknown origin brucellosis is relatively rare. Here, we report a case of brucellosis with secondary haemophagocytic lymphohistiocytosis without epidemiologic history.

CASE PRESENTATION

Chief complaints

A 50-year-old man was admitted to our hospital with a history of fever.

History of present illness

The patient's symptoms were fever and weakness for 6 d and diarrhoea for half a day. His body temperature reached 39.2 °C; he had no cough or other symptoms.

History of past illness

The patient had no remarkable medical history.

Personal and family history

The patient had unremarkable personal and family history.

Physical examination

There were no remarkable findings on physical examination except the patient's temperature was 39.5 °C.

Laboratory examinations

Laboratory analyses were conducted for blood, blood culture, biochemical tests, coagulation, and levels of serum C-reactive protein, serum procalcitonin, and ferritin. Laboratory data showed pancytopenia with a white blood cell count of 2.67×10^9 cells/L, haemoglobin levels of 14.7 g/dL, and a platelet count of 83×10^9 cells/L (Table 1). The blood culture on the sixth day of incubation grew *Brucella melitensis* (Figure 1). The level of soluble CD25 was 3256 U/mL, and abdominal ultrasonography revealed no splenomegaly or hepatomegaly. High resolution computed tomography of the chest showed no abnormalities.

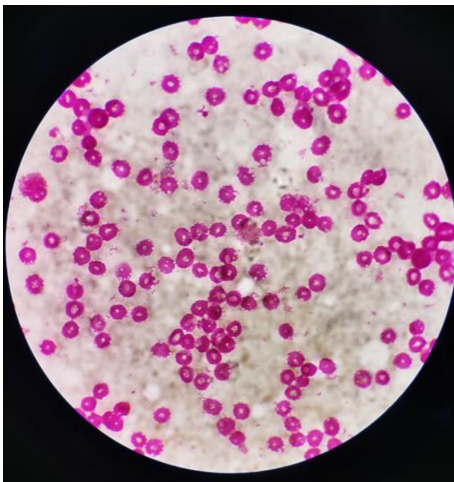
Imaging examinations

Bone marrow biopsy showed slight hypocellularity with an increase in macrophages exhibiting haemophagocytosis (Figure 2). *Brucella* was detected by mass spectrometry (Figure 3).

Table 1 Laboratory results for a 50-year-old man with brucellosis and secondary haemophagocytic syndrome

Test	Day 1	Day 4	Day 7	Day 10	Reference range
WBC, 10 ⁹ /L	2.67	2.35	3.92	3.98	3.5–9.5
HGB, g/L	147	139	143	145	130–175
PLT, 10 ⁹ /L	83	57	99	181	125–350
CRP, mg/L	30.76	18.25	4.87	0.79	0–10
PCT, ng/mL	0.385	-	-	-	3.5–9.5
Ferritin, ng/mL	825.7	> 1500	1401.2	895	23.9–336.2
AST, U/L	84.1	98.1	98.3	102.5	15–40
ALT, U/L	64.9	70.9	75.5	95.3	9–50
APTT, s	36.4	42.1	36.3	36.2	25.1–36.5
PT, s	11.3	11.8	11.8	11.8	9.4–12.5
Fib, g/L	3.17	2.69	3.17	2.73	2.0–4.8
FDP-D-dimer, ng/mL	1560	4190	696	284	0–222

ALT: Alanine aminotransferase; APTT: Activated partial thromboplastin time; AST: Glutamic-pyruvic transaminase; CRP: C-reactive protein; FDP: Fibrinogen degradation products; HGB: Haemoglobin; PCT: Procalcitonin; PT: Prothrombin time; WBC: White blood cell.

**Figure 1** *Brucella* in blood culture. Gram staining, 100 ×.

FINAL DIAGNOSIS

The final diagnosis of the presented case was brucellosis with haemophagocytic lymphohistiocytosis.

TREATMENT

Treatment for the infection and conventional supportive therapy were administered after admission. After oral doxycycline (100 mg/dose, twice a day) and intravenous cefoperazone/sulbactam (3000 mg/dose, twice a day) for 4 d, the fever disappeared, and the body temperature was normal (Figure 4).

OUTCOME AND FOLLOW-UP

The laboratory data of day 10 showed recovery. The patient was subsequently

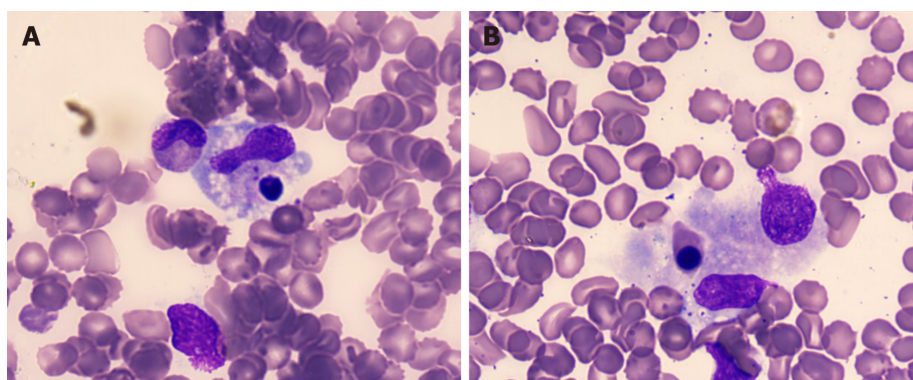


Figure 2 Haemophagocytosis in bone marrow aspiration and biopsy specimen, and histiocytes phagocytizing lymphocytes, platelets, and red cells. A and B: Wright staining, 1000 ×.

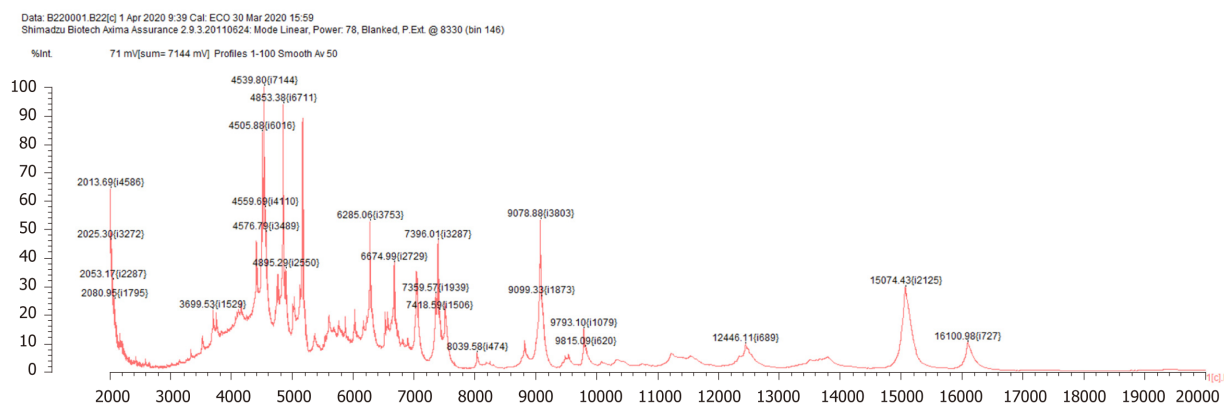


Figure 3 Mass spectrometry results.

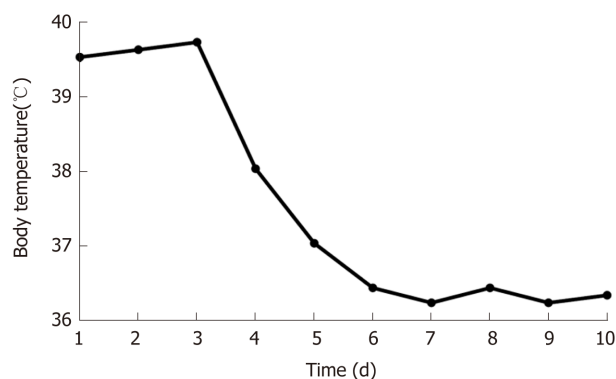


Figure 4 Body temperature changes during treatment.

discharged from our hospital.

DISCUSSION

Brucellosis is a zoonotic disease caused by bacteria of the *Brucella* species[4,5]. Diagnostic criteria include the epidemiologic history, clinical manifestations, and *Brucella* detection[6]. In this case, we identified *Brucella* by blood culture and mass spectrometry. This was a case of brucellosis with secondary haemophagocytic lymphohistiocytosis. After treatment with anti-brucella drugs, the haemogram became normal.

Generally, brucellosis develops after exposure to infected animals or contaminated products such as milk[7-9]. It should be noted that this patient lacked any history of exposure to these predefined epidemiologic factors. Interestingly, more and more unexplained infected cases have occurred. Recently, two cases of infective endocarditis in injection drug users without zoonotic exposure have been reported[10]. Zange *et al*[11] reported a patient with brucellosis who had no travel history, no exposure to unpasteurised dairy products, no animal contact, and no insect bites. The patient's ingested meat samples also showed negative results in polymerase chain reaction testing and microbiological cultures for *Brucella* species. Hence, from these reports, it is recommended to pay attention to unexplained infected individuals in whom an accurate diagnosis may have been missed. The patient in this present case denied any exposure to animals or contaminated products. Before the detection of *Brucella*, the absence of epidemiologic factors contributed to misdiagnosis, especially before the results of aetiological examination. Thus, in all unexplained, infectious febrile patients admitted to hospitals, it is necessary to use broad-spectrum antibiotics. Moreover, it is also warranted to consider rare pathogenic bacteria, such as *Brucella*.

Other concerns in this case report were the indicators of inflammation. *Brucella* is a gram-negative bacterium, and the detection of C-reactive protein, procalcitonin, and ferritin contribute to the severity of the inflammation. However, we noticed that C-reactive protein and ferritin levels showed a typical rising at the time of onset, although procalcitonin levels were below normal during hospitalisation. Obviously, procalcitonin levels did not correlate with the severity of illness. Incidentally, changes in procalcitonin levels may not be reflective of the severity of brucellosis.

CONCLUSION

Brucellosis cannot be excluded in patients with clinically unexplained fever, even in those without epidemiologic history. Before obtaining the aetiology test results, the administered antibiotics should cover rare pathogens, such as *Brucella*, which could prevent the timely exacerbation of the disease.

ACKNOWLEDGEMENTS

We thank our patient who agreed to the publication of this case report and provided information.

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