# **CASE REPORT**

# **Open Access**



# Pyogenic sacroiliitis caused by *Salmonella schwarzengrund* in a young healthy woman: a case report and literature review

Yuki Tokuyama<sup>1</sup>, Hiroyuki Yamada<sup>1,2</sup>, Ken Shinozuka<sup>1</sup>, Tomoyuki Yunoki<sup>1</sup> and Shigeru Ohtsuru<sup>1\*</sup>

# Abstract

**Background** Salmonella species are a leading cause of diarrheal diseases worldwide. Recent epidemiological studies have shown that Salmonella schwarzengrund (S. schwarzengrund) is highly prevalent in various regions. Herein, we report that S. schwarzengrund caused sacroiliac joint (SIJ) infection with septic shock in a young woman, although she was immunocompetent.

**Case presentation** A 20-year-old woman presented with left hip pain, accompanied by vasopressor-requiring hypotension. Her imaging examinations showed fluid collection in her SIJ and a small abscess in the left iliac muscle. Later, the blood and aspiration fluid culture and genetic analysis revealed the presence of S. schwarzengrund. We diagnosed sacroiliac joint (SIJ) infection with septic shock caused by S. schwarzengrund. Her condition improved after performing several interventional radiology (IVR) procedures for SIJ abscesses and providing appropriate antibiotic treatment. Finally, she was discharged without any sequelae. Screening tests and genetic analysis about her immuno-deficiency did not indicate a congenital disorder.

**Conclusion** These clinical courses indicate that S. schwarzengrund could cause the fatal SIJ infection irrespective of the host immunocompetence. Considering the recent increase in the diagnostic rate of S. schwarzengrund, this case emphasized the need to be more cautious about Salmonella species infection.

Keywords Non-typhoidal Salmonella, Pyogenic sacroiliitis, Psoas abscess

# Background

*Salmonella* species infection is a major global threat to public health [1, 2]. The most common symptom is gastroenteritis, which is self-limiting [3]. The microorganism does not frequently cause infection at different sites and severe sepsis[3]; however, it could be lethal among immunocompromised hosts such as patients with human

Shigeru Ohtsuru

ohtsuru@kuhp.kyoto-u.ac.jp

immunodeficiency virus (HIV) infection and malignancy and those receiving corticosteroids [4–7]. That is, *Salmonella* species is a common cause of bloodstream infection among individuals living in low-resource areas and may be associated with a high case fatality ratio [8]. Even in developed countries, *Salmonella* infection-related mortality is reported annually [9]. Moreover, recent studies have shown that the diagnostic rate of *Salmonella schwarzengrund* (*S. schwarzengrund*), a specific type of *Salmonella* species, is increasing in both animals and food products worldwide [10–12]. This can then lead to the local outbreak of *Salmonella* infection [13, 14].

Pyogenic sacroiliitis is a relatively rare disease [15]. A recent study showed that it accounts for only 1-2% of all septic arthritis cases [16]. Further, it is associated with



© The Author(s) 2023. **Open Access** This article is licensed under a Creative Commons Attribution 4.0 International License, which permits use, sharing, adaptation, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons licence, and indicate if changes were made. The images or other third party material in this article are included in the article's Creative Commons licence, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons licence and your intended use is not permitted by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this licence, visit http://creativecommons.org/licenses/by/4.0/. The Creative Commons Public Domain Dedication waiver (http://creativecommons.org/publicdomain/zero/1.0/) applies to the data made available in this article, unless otherwise stated in a credit line to the data.

<sup>\*</sup>Correspondence:

<sup>&</sup>lt;sup>1</sup> Department of Primary Care and Emergency Medicine, Kyoto University, Kyoto, Japan

<sup>&</sup>lt;sup>2</sup> Department of Nephrology, Graduate School of Medicine, Kyoto University, Kyoto, Japan

low clinical suspicion, vague clinical image, and poorly defined symptom localization [17]. Hence, its diagnosis can be challenging. The main causative pathogens are *Staphylococcus aureus* and *Pseudomonas aeruginosa* because they frequently cause bacteremia [18, 19]. To the best of our knowledge, there are only few case reports on sacroiliitis caused by *S. schwarzengrund*.

Herein, we present a young healthy female patient with *S. schwarzengrund*-related pyogenic sacroiliitis who developed septic shock and review other reported cases.

# **Case presentation**

A 20-year-old woman with a history of epilepsy became aware of left hip pain radiating down to the back of her leg for 3 days. The pain progressed gradually and became so severe that she could not move. Thus, she was admitted to the local hospital and received intravenous

**Fig. 1** Pelvic computed tomography image upon admission. The arrowhead indicates fluid collection at the left sacroiliac joint

antibiotics, cefazoline 6 g/day for her possible infection. However, her hip pain worsened, and she developed fever the following day. A pelvic computed tomography scan showed a fluid collection at the left sacroiliac joint (SIJ) and a small abscess in the left iliac muscle (Fig. 1). The next day, her hemodynamic parameters also deteriorated despite the antibiotic treatment, and consciousness disturbance developed. She was transferred to the emergency department of our hospital.

Upon hospital arrival, the patient's vital signs were as follows: heart rate, 122 beats/min; blood pressure, 95/34 mmHg (norepinephrine 0.16 mcg/kg/min, dobutamine 4.0 mcg/kg/min); body temperature, 36.4 °C; and oxygen saturation while on oxygen therapy at 2 L/min via nasal cannula, 98%. Further, the following are the laboratory test results: C-reactive protein level, 16.8 mg/dL and arterial blood gas lactate level, 4.4 mmol/L (Table 1).

Based on the examination results and clinical symptoms, the patient was diagnosed with septic shock caused by SIJ infection. Ultrasonography-guided abscess aspiration was performed to drain the joint fluid and identify the bacterial species. Later, the blood and aspiration fluid culture and genetic analysis revealed the presence of *S. schwarzengrund*.

Broad spectrum antibiotics (meropenem 3 g/day and vancomycin 2 g/day), vasopressors, and oxygen therapy were administered initially. The patient's hemodynamic and respiratory status gradually improved. After obtaining the culture results, antibiotic treatment was changed to levofloxacin. Ten days after the first drainage, the fever pattern and inflammation markers such as C-reactive protein and erythrocyte sedimentation rate significantly improved.

However, after the first drainage tube removal, the patient exhibited persistent fever and inflammation

 Table 1
 Blood test findings upon hospital admission

23
1.44
264
1005
403
1.1
2739
100 <
16.8
1.76
40.0 <



again, and hip magnetic resonance imaging revealed a growing abscess (Figs. 2 and 3). Thus, we performed the second drainage from days 26 to 35. The patient was discharged on day 38 with oral ampicillin treatment for eight more weeks. Upon discharge, she had neither any symptoms nor sequelae (Fig. 3).

To validate the entry route of *S. schwarzengrund*, we interviewed the patient in detail. Except for keeping one dog and two cats, she did not have any specific medical history, such as intravenous drug usage, recent overseas travel, diagnosis of sexually transmitted diseases, or consumption of suspicious food. She did not complain of any preceding gastrointestinal symptoms. The screening

test results for immunodeficiency diseases, including HIV infection and autoimmune disorders, were negative. Genetic analysis of congenital immunodeficiency also revealed no significant findings. These results indicated that she did not have any immune system disorders.

## **Discussion and conclusion**

Salmonella species present various kinds of clinical symptoms in human beings. Its common symptom is gastroenteritis, which is self-limiting. Moreover, Salmonella causes extraintestinal infections in different organ systems, such as the urinary tract, lung, and

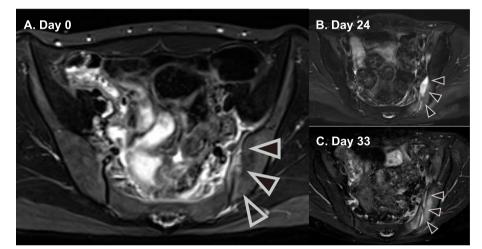


Fig. 2 Pelvic magnetic resonance image (A) upon admission and days (B) 24 and (C) 33. The arrowhead indicates fluid collection at the left sacroiliac joint

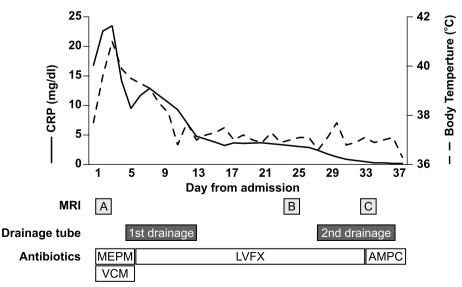


Fig. 3 Summary of clinical course

Author	Publication	Age/sex	Immunodeficiency	Culture	Treatment	Outcome
Tokuyama et al.	2023	20/F	None	Blood and synovial	Drainage	Improved
Shanahan et al.	1985	19/M	None	Blood	Surgical	Improved
Horgan et al.	1983	18/M	None	Stool and synovial	Surgical	Improved

Table 2 Previous cases of Salmonella schwarzengrund-related sacroiliitis

central nervous system [3, 20]. Occasionally, this infection can be fatal in immunocompromised hosts.

Our case showed that *S. schwarzengrund* could cause SIJ infection, which is an atypical extraintestinal infection. Additionally, it caused septic shock requiring vasopressors although the patient was young and healthy. These clinical courses indicated *S. schwarzengrund* infection could be a major concern even among immunocompetent patients.

To the best of our knowledge, only two cases of pyogenic sacroiliitis caused by *S. schwarzengrund* have been reported [21, 22]. Generally, extraintestinal *Salmonella* infection occurs in immunocompromised patients, such as elderly individuals and patients with HIV infection [23, 24]. However, as shown in Table 2, *S. schwarzengrund* caused extraintestinal focal infection even in young and healthy individuals. The background characteristics of our patient are consistent with those of patients in previously published reports. Considering the recent outbreak of *Salmonella* infection, it is crucial to be more cautious about the epidemiological status of *S. schwarzengrund* [13, 14].

Another highlight of this case report is the appropriate application of the interventional radiology (IVR) technique. Compared with surgery, the IVR approach is a non-invasive and cost-effective procedure [25, 26]. Nonetheless, it enables adequate drainage of abscesses, similar to our case. Now, SIJ has become a percutaneously accessible site through the development of highresolution CT and sonography, unlike at the time when the previous reports were published [21, 22]. Therefore, it should be highly desirable to consider its application moving forward.

This study also has certain limitations. First, the removal timing of the drainage tube might be disputable. Second, we could not identify the entry route of *S. schwarzengrund* except via pet food or exposure.

In conclusion, our case report showed that *S. schwarzengrund* could cause SIJ infection, which is fatal regardless of the host's immunocompetency. Moreover, it emphasized that people should be vigilant and aware of *Salmonella* infections.

# Abbreviations

AMPC	Amoxicillin
HIV	Human immunodeficiency virus
IVR	Interventional radiology
LVFX	Levofloxacin
MEPM	Meropenem
SIJ	Sacroiliac joint
VCM	Vancomycin

...

#### Acknowledgements

We are grateful to Michio Tanaka (Department of Clinical Laboratory Medicine, Kyoto University) for his technical support.

#### Authors' contributions

YT and HY wrote the manuscript and treated the patient. KS organized the ultrasound-guided procedure. TY provided technical and academic support. SO supervised the whole study. The author(s) read and approved the final manuscript.

#### Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

## Availability of data and materials

The datasets from this study are available from the corresponding author on request.

## Declarations

Ethics approval and consent to participate None.

#### **Consent for publication**

Written informed consent was obtained from the patient for publication of this study and accompanying images. The document for informed consent is available from the corresponding author on request.

## **Competing interests**

The authors declare no competing interests.

Received: 22 September 2022 Accepted: 8 March 2023 Published online: 20 March 2023

## References

- Marchello CS, Birkhold M, Crump JA, Vacc-iNTS consortium collaborators. Complications and mortality of non-typhoidal salmonella invasive disease: a global systematic review and meta-analysis. Lancet Infect Dis. 2022;22(5):692–705. https://doi.org/10.1016/S1473-3099(21)00615-0. (PMID 35114140).
- GBD 2017 Non-typhoidal Salmonella invasive disease collaborators. The global burden of non-typhoidal salmonella invasive disease: a systematic analysis for the Global Burden of Disease Study 2017. Lancet Infect Dis.

2019;19(12):1312–24. https://doi.org/10.1016/S1473-3099(19)30418-9. PMID 31562022.

- Eng S-K, Pusparajah P, Ab Mutalib N-S, Ser H-L, Chan K-G, Lee L-H. Salmonella: a review on pathogenesis, epidemiology and antibiotic resistance. Front Life Sci. 2015;8(3):284–93. https://doi.org/10.1080/21553769.2015. 1051243.
- Kariuki S, Onsare RS. Epidemiology and genomics of invasive nontyphoidal salmonella infections in Kenya. Clin Infect Dis. 2015;61(Suppl 4):S317–24. https://doi.org/10.1093/cid/civ711. (PMID26449947).
- Phu Huong Lan N, Le Thi Phuong T, Nguyen Huu H, Thuy L, Mather AE, Park SE, et al. Invasive non-typhoidal Salmonella infections in Asia: clinical observations, disease outcome and dominant serovars from an infectious disease hospital in Vietnam. PLOS Negl Trop Dis. 2016;10(8):e0004857. https://doi.org/10.1371/journal.pntd.0004857. (PMID 27513951).
- Gruenewald R, Blum S, Chan J. Relationship between human immunodeficiency virus infection and salmonellosis in 20- to 59-year-old residents of New York City. Clin Infect Dis. 1994;18(3):358–63. https://doi.org/10. 1093/clinids/18.3.358. (PMID8011816).
- Graham SM. Nontyphoidal salmonellosis in Africa. Curr Opin Infect Dis. 2010;23(5):409–14. https://doi.org/10.1097/QCO.0b013e32833dd25d. (PMID20736739).
- Crump JA, Sjölund-Karlsson M, Gordon MA, Parry CM. Epidemiology, clinical presentation, laboratory diagnosis, antimicrobial resistance, and antimicrobial management of invasive salmonella infections. Clin Microbiol Rev. 2015;28(4):901–37. https://doi.org/10.1128/CMR.00002-15. (PMID26180063).
- Scallan E, Hoekstra RM, Angulo FJ, Tauxe RV, Widdowson MA, Roy SL, et al. Foodborne illness acquired in the United States-major pathogens. Emerg Infect Dis. 2011;17(1):7–15. https://doi.org/10.3201/eid1701.p11101. (PMID21192848).
- Duc VM, Shin J, Nagamatsu Y, Fuhiwara A, Toyofuku H, Obi T, et al. Increased Salmonella Schwarzengrund prevalence and antimicrobial susceptibility of Salmonella enterica isolated from broiler chickens in Kagoshima Prefecture in Japan between 2013 and 2016. J Vet Med Sci. 2020;82(5):585–9. https://doi.org/10.1292/jvms.20-0096. (PMID32213751).
- Silva KC, Fontes LC, Moreno AM, Astolfi-Ferreira CS, Ferreira AJ, Lincopan N. Emergence of extended-spectrum-β-lactamase CTX-M-2-producing Salmonella enterica serovars Schwarzengrund and Agona in poultry farms. Antimicrob Agents Chemother. 2013;57(7):3458–9. https://doi.org/ 10.1128/AAC.05992-11. (PMID23629722).
- Aarestrup FM, Hendriksen RS, Lockett J, Gay K, Teates K, McDermott PF, et al. International spread of multidrug-resistant Salmonella Schwarzengrund in food products. Emerg Infect Dis. 2007;13(5):726–31. https://doi. org/10.3201/eid1305.061489. (PMID17553251).
- Tamber S, Dougherty B, Nguy K. Salmonella enterica serovars associated with bacteremia in Canada, 2006–2019. Can Commun Dis Rep. 2021;47(56):259–68. https://doi.org/10.14745/ccdr.v47i56a03. (PMID34220350).
- Du X, Jiang X, Ye Y, Guo B, Wang W, Ding J, et al. Next generation sequencing for the investigation of an outbreak of Salmonella Schwarzengrund in Nanjing. China Int J Biol Macromol. 2018;107(1):393–6. https://doi.org/10. 1016/j.ijbiomac.2017.09.005. (PMID 28888545).
- Kucera T, Brtkova J, Sponer P, Ryskova L, Popper E, Frank M, et al. Pyogenic sacroiliitis: diagnosis, management and clinical outcome. Skeletal Radiol. 2015;44(1):63–71. https://doi.org/10.1007/s00256-014-1999-y. (PMID25231169).
- Hodgson BF. Pyogenic sacroiliac joint infection. Clin Orthop Relat Res. 1989;246(246):146–9. https://doi.org/10.1097/00003086-198909000-00023. (PMID2766603).
- Barnes M, Bush C, Jones J. Pyogenic sacroiliitis: A rare complication of inflammatory bowel disease. Am J Emerg Med. 2019;37(7):1395.e1-2. https://doi.org/10.1016/j.ajem.2019.04.017. (PMID31005399).
- Cosma S, Borella F, Carosso A, Ingala A, Fassio F, Robba T, et al. Osteomyelitis of the pubic symphysis caused by methicillin-resistant Staphylococcus aureus after vaginal delivery: a case report and literature review. BMC Infect Dis. 2019;19(1):952. https://doi.org/10.1186/s12879-019-4595-x. (PMID31703612).
- Ross JJ, Hu LT. Septic arthritis of the pubic symphysis: review of 100 cases. Med (Baltim). 2003;82(5):340–5. https://doi.org/10.1097/01.md.00000 91180.93122.1c. (PMID14530783).

- Ramos JM, Aguado JM, García-Corbeira P, Alés JM, Soriano F. Clinical spectrum of urinary tract infections due on nontyphoidal Salmonella species. Clin Infect Dis. 1996;23(2):388–90. https://doi.org/10.1093/clinids/23.2. 388. (PMID8842280).
- Horgan JG, Walker M, Newman JH, Watt I. Scintigraphy in the diagnosis and management of septic sacro-iliitis. Clin Radiol. 1983;34(3):337–46. https://doi.org/10.1016/s0009-9260(83)80357-2. (PMID6839659).
- Shanahan MD, Ackroyd CE. Pyogenic infection of the sacro-iliac joint. A report of 11 cases. J Bone Joint Surg Br. 1985;67(4):605–8. https://doi.org/ 10.1302/0301-620X.67B4.4030859. (PMID 4030859).
- Keddy KH, Sooka A, Musekiwa A, Smith AM, Ismail H, Tau NP, et al. Clinical and microbiological features of salmonella meningitis in a South African population, 2003–2013. Clin Infect Dis. 2015;61(Suppl 4):S272–82. https:// doi.org/10.1093/cid/civ685. (PMID26449942).
- Chen PL, Chang CM, Wu CJ, Ko NY, Lee NY, Lee HC, et al. Extraintestinal focal infections in adults with nontyphoid Salmonella bacteraemia: predisposing factors and clinical outcome. J Intern Med. 2007;261(1):91–100. https://doi.org/10.1111/j.1365-2796.2006.01748.x. (PMID17222172).
- Maudgil DD. Cost effectiveness and the role of the National Institute of Health and Care Excellence (NICE) in interventional radiology. Clin Radiol. 2021;76(3):185–92. https://doi.org/10.1016/j.crad.2020.09.017. (PMID33081990).
- Doherty MG. Value of interventional radiology: past, present, and future. Semin Interv Radiol. 2019;36(1):26–8. https://doi.org/10.1055/s-0039-1679951. (PMID30936613).

# **Publisher's Note**

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

## Ready to submit your research? Choose BMC and benefit from:

- fast, convenient online submission
- thorough peer review by experienced researchers in your field
- rapid publication on acceptance
- support for research data, including large and complex data types
- gold Open Access which fosters wider collaboration and increased citations
- maximum visibility for your research: over 100M website views per year

## At BMC, research is always in progress.

Learn more biomedcentral.com/submissions

