CASE REPORT

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# Kushiyaki-related Streptococcus suis meningitis with ventriculitis: A case report

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#### 1 **INTRODUCTION**

This report describes Streptococcus suis meningitis with ventriculitis in a 66-year-old kushiyaki chef, who presented with fever and meningeal irritation signs. Cerebrospinal fluid testing revealed increased cell counts and protein levels, and the presence of gram-positive cocci. Kushiyaki chefs are at high risk of this infection, and prophylaxis should be considered.

Streptococcus suis, a gram-positive, facultative anaerobic bacterium, is an emerging zoonotic pathogen.<sup>1</sup> Streptococcus suis is endemic in the upper respiratory tract of pigs, particularly in the tonsils and nasal cavity, but it can also be found in the genitalia and gastrointestinal tract.<sup>2,3</sup> Although multiple serotypes can infect pigs, S. suis serotype 2 is the most common human pathogen.<sup>2,3</sup> As of 2012, 1584 human cases of S. suis serotype 2 have been reported.<sup>4</sup> Streptococcus suis infections are more prevalent in South-East Asia owing to the high consumption of pork and frequent small-scale pig rearing.<sup>5-7</sup> People with occupations that involve contact with pigs or pork, such as slaughterhouse workers and butchers, are at a particularly high risk of S. suis infection.<sup>8</sup>

Abstract

Streptococcus suis causes a zoonotic disease that commonly manifests as meningitis. People handle pork or its derivatives are at a high risk of infection. Handwashing and donning personal protective equipment are the practical preventive measures.

**KEYWORD** Streptococcus suis, infectious disease, ventriculitis

> The characteristic clinical manifestation of S. suis infection in human beings is meningitis.<sup>4,9</sup> Irreversible hearing loss and vestibular dysfunction are well-known complications of S. suis meningitis,<sup>4,10</sup> and its causes, treatments, and predictive scores have been investigated.<sup>11</sup> However, the underlying causes of less frequent complications, such as ventriculitis, are poorly understood.<sup>4,10</sup> Herein, we report a case of meningitis with pyogenic ventriculitis associated with S. suis infection.

#### 2 **CASE HISTORY**

A 66-year-old Japanese man presented to the hospital with a 4-day history of chills, fever, nausea, vomiting, and a medical history of type 2 diabetes mellitus. The patient was a kushiyaki (skewered foods) chef and handled pork while cooking without gloves or other personal protective equipment. He had no travel history. We obtained informed consent from the patient for the publication of this case report. Ethics approval was not required per the relevant review board.

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# 2.1 | Examination and investigations

Upon admission, he was disoriented; his vital signs were as follows: body temperature, 38.1°C; heart rate, 100/min; blood pressure, 168/98 mm Hg; and respiratory rate, 16/ min. He had signs of nuchal rigidity. He had no anisocoria, intact light reflexes, and no pain on sinus tapping. Other physical examination findings were normal. Laboratory findings on admission were as follows (Table 1): white blood cells, 16 200/µL; hemoglobin, 17.3 g/dL; and platelets,  $4.6 \times 10^4/\mu$ L. Liver enzyme levels were elevated: aspartate aminotransferase, 123 IU/L and alanine aminotransferase, 114 IU/L. Renal function was also exacerbated: blood urea nitrogen, 28.1 mg/dL and creatinine, 1.23 mg/dL. The C-reactive protein level and erythrocyte sedimentation rate were 24.1 mg/L and 17 mm/h, respectively. Anti-HIV antibodies were absent, and the HbA1c level was 7.3%.

Head computed tomography showed no evidence of brain herniation. We identified positive signs of meningeal irritation and conducted a fluid test. Cerebrospinal fluid (CSF) had a cloudy appearance, and analysis revealed a pressure of 24 cm CSF, a cell count of 496 cells/µL (84% neutrophils and 16% lymphocytes), protein levels of 417 mg/dL, and sugar levels of 1 mg/dL. Gram staining on the CSF revealed grampositive cocci.

# 2.2 | Differential diagnosis and treatment

These findings led us to diagnose bacterial meningitis caused by gram-positive bacteria. Ceftriaxone (2.0 g in 12-hour intervals), vancomycin (1.0 g in 8-hour intervals), ampicillin (2.0 g in 4-hour intervals), and dexamethasone (9.9 mg in 6-hour intervals) were administered. On day 2, the fever subsided, but the patient experienced a generalized seizure. Levetiracetam 1000 mg was administered to control the seizure. Head diffusion-weighted magnetic resonance imaging (MRI) showed a high signal in the bilateral lateral ventricular triangles, indicating ventriculitis (Figure 1), and *S. suis* serotype 2 was identified in CSF culture by agglutination with antisera. A careful physical examination revealed two small scars in the patient's left hand (Figure 2). A family member mentioned that the patient often accidentally pointed the skewer at his hand when cooking pork on a spit.

# 2.3 | Outcome and follow-up

Altogether, our findings suggested that *S. suis* infected the patient through the scars in his hands, causing meningitis, ventriculitis, and inner ear dysfunction. The antibiotic treatment was changed from ceftriaxone, vancomycin, and ampicillin

#### TABLE 1 Laboratory data of the patient

Variable	Reference range	Value on admission
Red blood cell (per µL)	3 860 000-4 920 000	4 910 000
Hemoglobin (g/dL)	11.6-14.8	17.3
Hematocrit (%)	35.1-44.4	47.8
White blood cell (per $\mu$ L)	3300-8600	16 200
Differential blood count (per µL)		
Neutrophils (%)	0.0-2.5	39.0
Lymphocytes (%)	16.5-49.5	43.8
Monocytes (%)	2.0-10.0	14.1
Eosinophil (%)	0.0-8.5	2.8
Basophil (%)	0.0-2.5	0.3
Platelets (per µL)	158 000-348 000	46 000
Sodium (mmol/L)	135-145	138
Potassium (mmol/L)	3.6-4.8	3.4
Chloride (mmol/L)	101-108	100
Urea nitrogen (mg/dL)	8.0-20.0	28.1
Creatinine (mg/dL)	0.46-0.79	1.23
Alanine aminotransferase (U/L)	13-30	114
Aspartate aminotransferase (U/L)	7-23	123
Alkaline phosphatase (U/L)	106-322	223
Creatine kinase (U/L)	41-153	140
Lactate dehydrogenase (U/L)	124-222	264
C-reactive protein (mg/ dL)	0.0-0.1	24.1
Erythrocyte sedimentation rate (mm/h)	1-7	17
Glycated hemoglobin (%)	4.6-6.2	7.3

to penicillin G (150 000 units/kg/day in 4-hour intervals) according to antimicrobial susceptibility results (Table 2).

Despite dexamethasone treatment for 4 days, the patient had a persistent hearing impairment. Hearing tests revealed a decrease of 110 dB on the right and left sides. Auditory brainstem response assessment showed a value of 100 dB without a bilateral response. On day 6, contrast MRI showed right cochlea enhancement, suggesting right labyrinthitis. We concluded that the patient had sensorineural hearing loss owing to inner ear damage. Therefore, corticosteroid therapy was administered. Intravenous methylprednisolone (250 mg/ day) was initiated; the dose was tapered to 125 mg/day on day 8, 60 mg/day on day 10, and 40 mg/day on day 12.

**FIGURE 1** (A) Diffusion-weighted magnetic resonance images showing high signals in the bilateral ventricles (arrow) and (B) apparent diffusion coefficient maps showing low signals





TABLE 2 Antimicrobial susceptibility test results



**FIGURE 2** A photograph of the patient's hand. A small scar can be seen on the left hand, which may have been accidentally caused by a skewer (arrow)

On day 6, the patient's general condition improved, and CSF examination revealed improved pressure (16 cm CSF). Laboratory values also improved as follows: cell count, 44 cells/ $\mu$ L; protein level, 84 mg/dL; and plasma glucose level, 180 mg/dL. On day 20, MRI showed improvement in ventriculitis, and penicillin G was administered for 22 days. After treatment completion, the patient showed no signs of recurrent meningitis and was discharged on day 40. Inner ear dysfunction did not improve, and cochlear implant surgery

		MIC
	MIC (µg/mL)	value
Penicillin G	≤0.03	S
Ampicillin	≤0.12	S
Cefazoline	≤0.25	S
Cefotiam	0.5	S
Cefotaxime	≤0.06	S
Ceftriaxone	≤0.06	S
Imipenem hydrate/cilastatin	≤0.12	S
Erythromycin	>8	R
Clindamycin	>4	R
Minocycline	>8	R
Levofloxacin	1	S
Vancomycin	$\leq 1$	S
Teicoplanin	<1	S

*Note:* The antimicrobial susceptibility of *S suis* strains was determined by Clinical and Laboratory Standards Institute (CLSI).<sup>25</sup>

Abbreviations: MIC, minimum inhibitory concentration; R, resistance; S, sensitive.

was performed 4 months after disease onset. The patient gradually improved and recovered without relapse.

# **3** | **DISCUSSION**

We reported a rare case of pyogenic ventriculitis associated with *S. suis* infection. People who handle pork or its derivative products are at a high risk of *S. suis* infection <sup>1</sup> since the route of entry of the organism into human beings is via small skin wounds.<sup>12</sup> However, recent studies have shown that only 20% of patients reported having skin wounds when handling pork.<sup>13</sup> Kushiyaki is a traditional Japanese dish in WILEY\_Clinical Case Reports

which bite-sized pieces of chicken or pork are skewered and grilled over an open fire. In this case, the patient was a kushiyaki chef who handled pork with his bare hands. Since there were small scars on his hands, probably linked to cooking kushiyaki, we considered that to be the route of his infection. Kushiyaki chefs may be at a higher risk of infection than people engaging in other activities involving pigs. As there is no *S. suis* vaccine for human beings,<sup>14</sup> handwashing and wearing personal protection equipment are the most practical preventive measures.

Several patients with S. suis meningitis successfully recovered following a treatment scheme similar to that used for pneumococcal meningitis.<sup>15</sup> However, other patients with S. suis meningitis showed relapse 2 weeks post-treatment completion and required prolonged therapy (4-6 weeks).<sup>16</sup> Dejace et al suggested that treatment should be tailored to clinical and laboratory findings.<sup>17</sup> A recent review found that the antimicrobial treatment duration ranged from 7 to 42 days in cases of meningitis, spondylitis, and endocarditis.<sup>1</sup> In this case, S. suis infection only led to meningitis and pyogenic ventriculitis. Ventriculitis is a rare condition secondary to meningitis<sup>18</sup> that can cause persistent infection and treatment failure.<sup>19,20</sup> Additionally, if meningitis is associated with ventriculitis, prolonged antibiotic treatment might be required. Only one case of S. suis meningitis associated with ventriculitis has been reported so far; the patient required longer antibiotic treatment than the usual 24-day course for bacterial meningitis.<sup>21</sup> However, few studies have reported ventriculitis in cases involving long-term antimicrobial treatment for S. suis meningitis, suggesting that ventriculitis might be an underlying cause for disease recurrence.<sup>22</sup> Furthermore, although our patient was treated for a longer period because of the presence of ventriculitis, the recommended treatment duration for S. suis-associated ventriculitis has not been determined<sup>23</sup>; thus, further studies are needed. As ventriculitis is most frequently detected based on diffusion-weighted MRI findings,<sup>24</sup> MRI should be performed in cases of suspected ventriculitis, especially in cases of S. suis meningitis involving a poor response to treatment.

# 4 | CONCLUSION

We reported a rare case of pyogenic ventriculitis associated with *S. suis* infection. The patient's occupation as a kushiyaki chef may be linked to a high infection risk because it involves handling pork. Therefore, preventive measures should be taken to avoid infection. Additionally, MRI should be performed for patients with *S. suis* ventriculitis to ensure suitable and effective treatment. Since the appropriate treatment duration has not yet been determined, further reports of *S. suis* ventriculitis are needed.

# CONFLICT OF INTEREST

None declared.

## AUTHOR CONTRIBUTIONS

All authors critically revised the report, commented on drafts of the manuscript, and approved the final report.

## ETHICAL APPROVAL

We have obtained informed consent from the patient for publication of this case report.

### DATA AVAILABILITY STATEMENT

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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