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Case Report

The first report of human meningitis and pyogenic ventriculitis caused by *Streptococcus suis*: A case reportTomonobu Yanase ^{a,*}, Daiichi Morii ^{a,b}, Satoshi Kamio ^c, Ayako Nishimura ^c, Eri Fukao ^c, Yuri Inose ^c, Yutaka Honma ^c, Nobuo Kitahara ^d, Takayuki Yokozawa ^e, Bin Chang ^f, Toshimi Oda ^a^a Department of Infectious Disease, Showa General Hospital, 8-1-1 Hanakoganei, Kodaira, Tokyo 187-8510, Japan^b Department of Infectious Control and Prevention, Graduate School of Medicine, Osaka University, 2-15 Yamadaoka, Suita, Osaka 565-0871, Japan^c Department of Neurology, Showa General Hospital, 8-1-1 Hanakoganei, Kodaira, Tokyo 187-8510, Japan^d Department of Otorhinolaryngology, Showa General Hospital, 8-1-1 Hanakoganei, Kodaira, Tokyo 187-8510, Japan^e Department of Clinical Laboratory, Showa General Hospital, 8-1-1 Hanakoganei, Kodaira, Tokyo 187-8510, Japan^f Department of Bacteriology I, National Institute of Infectious Diseases, 1-23-1 Toyama, Shinjuku, Tokyo 162-8640, Japan

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ABSTRACT

Streptococcus suis, a gram-positive facultative anaerobe commonly found in pigs, is an emerging zoonotic pathogen. Herein, we describe a case of a 45-year-old male Japanese meat wholesaler with *S. suis* meningitis and pyogenic ventriculitis. *S. suis* was isolated from his blood and cerebrospinal fluid culture, and sequence type (ST) and serotype were confirmed to be ST1 and serotype 2, respectively, by multi-locus sequence typing and the Quellung reaction. Magnetic resonance imaging (MRI) revealed right labyrinthitis and pyogenic ventriculitis. The patient was treated with ceftriaxone and ampicillin for 24 days; the treatment was deemed successful based on negative blood cultures on day 4. However, the patient experienced hearing loss and a vestibular nerve disorder. *S. suis* is a rare pathogen in Japan but can cause severe infection and sequelae. To the best of our knowledge, this is the first report of a human case of pyogenic ventriculitis caused by *S. suis*. Our findings suggest that *S. suis* infection should be considered when hearing impairment is present in a patient with bacterial infection and that MRI can help detect ventriculitis, which can necessitate a prolonged treatment duration.

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1. Introduction

Streptococcus suis, a gram-positive facultative anaerobe, is an emerging zoonotic pathogen [1]. *S. suis* can be a both commensal and invasive bacterium in pigs. The natural habitat of *S. suis* is the upper respiratory, genital and gastrointestinal tract of pigs [2], and it can be transmitted to humans by close contact with pigs or pork [3]. Most human *S. suis* cases have been reported in South-East Asia [4–7], which is characterized by a high density of pigs and the consumption of uncooked or lightly cooked pig products [8]. Since the first human case was described in Denmark in 1968 [9], more than 900 cases of human *S. suis* infection have been reported worldwide [10].

Meningitis is the most common clinical presentation of an *S. suis* infection in humans [4]. Potentially irreversible hearing and vestibular function loss are striking complications of *S. suis* meningitis [11].

To the best of our knowledge, this is the first case of pyogenic ventriculitis associated with an *S. suis* infection in a human.

2. Case report

A 45-year-old Japanese man, with no remarkable medical history, was admitted to our hospital with a diagnosis of bacterial meningitis. He worked as a meat wholesaler, dealing with pork, beef, and chicken.

A week before this admission, he developed painful erythematous swelling on the back of his right hand. Three days later, he developed a slight fever. The next day, he developed a high-grade

* Corresponding author.

E-mail address: tomo.nov.20.1990@gmail.com (T. Yanase).

fever, headache, tinnitus, and hearing loss. His symptoms worsened the next day and he visited a local doctor. The levels of inflammatory markers were above normal (leukocytes 19700 cells/ μ L, C-reactive protein 26.5 mg/dL) and therefore he was transferred to our local tertiary hospital.

On arrival, the patient had a body temperature of 38.5 °C, pulse rate of 104 beats/min, blood pressure of 163/103 mmHg, and a respiratory rate of 24 breaths/min. He was alert and oriented. He showed bilateral deafness and meningeal signs. His neurological signs were otherwise normal. An abraded wound was observed on the back of his right hand.

Head computed tomography (CT) scan revealed no abnormalities. The result of the cerebrospinal fluid (CSF) was shown in Table 1. Gram staining of the CSF revealed gram-positive diplococci. After obtaining two sets of blood and CSF cultures, we empirically administered ceftriaxone (2.0 g at 12-h intervals), ampicillin (2.0 g at 4-h intervals), and vancomycin (1.0 g at 8-h intervals), in addition to dexamethasone (0.15 mg/kg at 6-h intervals).

On day 4, bacterial growth was detected in both the blood and CSF cultures, which were sent to the National Institute of Infectious Disease for confirmation of *S. suis* serotype 2 by using the Quellung reaction with pneumococcal antisera (Staten Serum Institut, Copenhagen, Denmark). Multilocus sequence typing (MLST) was performed as described by King et al. [12]. Allelic numbers and sequence type (ST) of the strain were assigned using the pneumococcal MLST website (<http://ssuis.mlst.net/>) for designations. The strain isolated from our patient was identified as ST1 (Table 2). Based on the antimicrobial susceptibility results (Table 3), we discontinued vancomycin. Dexamethasone was finished after a total of 4 days of administration.

Diffusion-weighted magnetic resonance imaging (MRI) on day 5 revealed high signal intensity in the ventricles (Fig. 1), which suggested pyogenic ventriculitis. No abnormal signals were noted in the cerebral parenchyma. On day 10, contrast-enhanced MRI revealed enhancement in the right cochlea, indicating right labyrinthitis (Fig. 2). The high signal intensity in the ventricles decreased. The whole-spine MRI, the whole-body contrast-enhanced CT scan, and transthoracic echocardiography performed on day 3, 19, and 21, respectively, showed no other focus of infection.

The patient's fever and headache resolved on day 5. The repeated lumbar puncture on day 10 revealed resolving CSF, with negative Gram staining and culture study results. We continued ceftriaxone and ampicillin for 3 weeks after the first negative blood culture which was obtained on day 4 (from day 1 to day 24). On the last day of antibiotic treatment, we performed brain MRI to confirm that the pyogenic ventriculitis was resolved. The third lumbar puncture and contrast-enhanced MRI conducted on day 31 and 33, respectively, revealed no abnormal results. He was discharged in stable condition on day 36.

Table 2
Sequence type.

Strain	ST	<i>aroA</i>	<i>cpn60</i>	<i>dpr</i>	<i>gki</i>	<i>mutS</i>	<i>recA</i>	<i>thrA</i>
Sample 1	1	1	1	1	1	1	1	1

Despite complete recovery from the meningitis and pyogenic ventriculitis, the patient's hearing and vestibular function loss persisted (Fig. 3); this impairment showed no improvement on day 11, we started the patient on adjunctive corticosteroid treatment in accordance with sudden deafness as shown in Fig. 4. On discharge, audiometry still showed right profound (83.8 dB) and left moderate (47.5 dB) hearing impairment; however, the patient improved to the point of being able to work without a hearing aid at 3 weeks after discharge.

3. Discussion

Streptococcosis is a major health problem in pigs worldwide. Workers in the swine and pork industry or people who consume raw or undercooked pork are at increased risk of this infection [13]. The transmission from bacteria to humans is thought to occur through skin wounds or inhalation [3]. Our patient was a meat wholesaler dealing with pork, beef, and chicken. His work included transporting packed meat and cutting bones, intestinal tracts, hearts, and livers. He often worked without gloves and sustained hand injuries. It was reasonable to consider his hand wound was a bacterial entry gate. Since no human *S. suis* vaccine is available, the most practical prophylaxis strategy is preventive measures such as wearing gloves and a mask, and washing hands.

S. suis is classically classified into 35 serotypes (1–34 and 1/2) [14]. The distribution of serotypes among clinical cases differs geographically, but serotype 2 is the most frequently isolated strain from both in pig and human infection cases. Additionally, bacterial isolates have been characterized using MLST, which is based on seven housekeeping genes [12]. In the present case, the isolate was ST1 serotype 2, which is associated with most cases worldwide [15]. Since the patient's company imported pork from a variety of countries, we were unable to determine the exact origin of the causative organism in this case.

Meningitis is the most frequently (50–60%) described clinical presentation of *S. suis* infection [16]. *S. suis* meningitis is characterized by a high rate (53.0–55.3% [10,16]) of hearing loss and vestibular dysfunction, compared with 14% in bacterial meningitis [17]. The hearing loss is usually sensorineural, occurring in the high-frequency range [18]. Hemorrhagic labyrinthitis has been suggested as a cause of this hearing impairment [19]. Our patient developed bilateral hearing impairment, even though contrast-enhanced MRI revealed no abnormality in his left cochlea or

Table 1
Cerebrospinal fluid.

Variable	Reference range	On admission	Day 10	Day 31
Opening pressure (cm of water)	<20	27	7	16
Color	Colorless	Light white	Colorless	Colorless
Turbidity	Clear	Cloudy	Clear	Clear
Xanthochromia	None	None	None	None
Cell count (/3 μ L)	<15	15520	44	14
Differential count (%)				
Neutrophils		95	20	1
Lymphocytes		4	80	91
Monocytes and others		1	0	8
Protein (mg/dL)	5–45	459	46	51
Glucose (mg/dL)	45–80	27	47	50

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