CASE REPORT

Endophthalmitis with bilateral deafness from disseminated *Streptococcus suis* infection

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SUMMARY

Streptococcus suis is a Gram-positive cocci bacterium that are found mainly in pigs and can be transmitted to human through pigs or pork exposure. The disease is mainly found among occupations involving swine contact in western countries whereas in Asia the disease is usually contracted through raw pork consumption. In this case report, we present a case of a middle-aged Thai man who acquired the infection from raw pork consumption. He presented with endogenous endophthalmitis with infective spondylodiscitis, sepsis and meningitis and later developed blindness of the right eye and permanent bilateral hearing loss disseminated from *S. suis* infection. Our report suggests that *S. suis* infection be considered as a causative factor in patient presenting with established clinical symptoms and predisposing factors. Cultural habit of eating raw pork should be taken into account especially in Asian countries.

BACKGROUND

Streptococcus suis is an emerging zoonosis that is mainly found as normal flora in pigs.[1] The pathogen can be transmitted to human through exposure of pigs or consumption of raw pork[2] causing a number of serious infections.[3] The most common clinical manifestations found were meningitis (68%), followed by sepsis (25.0%) and arthritis (12.9%) whereas infective endocarditis (12.4%), endopthalmitis (4.6%) and spondylodiscitis (3.7%) were uncommon.[4] Sensorineural hearing loss (SNHL) (39%) was the most common complication from *S. suis* meningitis in which the disease is usually irreversible despite effective treatment.[4 5]

The disease is usually found prevalent among farmers, butchers and abattoir workers involving swine contact in western countries.[6 7] However, this was not always the case in Asian countries where pig-related occupation infection is less than half of the reported cases.[8 9] According to a recent systematic review and meta-analysis, raw pork consumption, exposure to pigs or raw pork, pig-related occupation and male sex are the significant risk factors of the infection.[10] This suggests that traditional food habit involving raw pork consumption probably plays an essential role in the disease infection in the Asian region.

Since the first *S. suis* infection in human related to meningitis and sepsis was reported in Denmark in 1968,[11] the disease has caused wide spread infection globally with more than 1,500 reported cases as of 2012.[12] In Thailand, *S. suis* infection is an important health problem with more than 500 cases reported up to 2017[10] and possibly the second causative agent of adult streptococcal meningitis.[13]

We therefore present a patient with endogenous endophthalmitis which is an uncommon clinical presentation, who later developed blindness of the right eye and permanent bilateral SNHL disseminated from *S. suis* infection.

CASE PRESENTATION

A 48-year-old educated, Thai male was admitted to Chiang Mai University Hospital (CMUH) with endogenous endopthalmitis infecting his right eye. He is a degree graduate and works as an airport security administration officer. He consumes two to three bottles of beer daily and often consumes raw fermented pork or *'naem'* (sour pork). He has a medical history of gout and spondylodiscitis. He refused taking any other medications except analgesics for his back pain. He did not travel to anywhere within the region and had no pig farming exposure prior to admission at the hospital.

Upon detailed consultation, the patient narrated that he often experienced lack of sleep due to work stress, fatigue and dizziness for the past few months before admission. Six days before admission, the patient reported to have consumed a northern-Thai style raw pork dish called *'larb dib'* at *Songkran* (the Thai new year) party at his office. The pork and ingredients were bought from wet markets in Sankampaeng and Saraphi Districts, Chiang Mai Province and cooked by his colleagues. According to the patient, he was the only one who fell sick after the party. Two days later, he developed a low-grade fever, neck, joint and waist pain and blurred vision at his right eye along with a decrease in hearing ability at his right ear. The symptoms got worse and he visited a local hospital where he underwent an X-ray for waist and neck bone. He received an unknown intravenous antibiotic for joint pain, then later progressed to have nausea, vomiting, hearing loss and redness of his right eye. He was then referred to CMUH.

At admission, the body temperature was 37.5°C with 24 hours peak temperature of 37.8°C, pulse rate of 90 beats per minute and a blood pressure of 130/80 mm Hg. His respiratory rate was 20 breaths per minute. A physical examination identified chemosis and periorbital swelling at the right eye and C-spine tenderness.

Figure 1 MRI of whole spine.

INVESTIGATIONS

A routine laboratory and microbiological culture were ordered. An MRI of the whole spine was done to rule out infective spondylodiscitis suggested spondylodiscitis at C4/5 level and left side of L5/S2 (figure 1). There was left paracentral disc extrusion with associated disc bulging and osteophyte causing mild spinal cord compression.

Assessment of visual acuity (VA) revealed hand movement (HM) at right VA and a VA of 6/16 on the left. There were marked injected conjunctiva, whitish, large keratic precipitate, and hypopyon uveitis in the right eye. The relative afferent pupil defect (RAPD) test showed a positive reverse RAPD. The left eye was normal. The patient was still alert and oriented despite the presence of febrile illness and neck stiffness.

The laboratory results showed no outstanding value except an elevated neutrophil (83.6%; normal range, 40%–75%), and erythrocyte sedimentation rate (65 mm/hour; normal range, 0–15 mm/hour), and a transient increase of alanine aminotransferase (65 U/L; normal range, 0–41 U/L), blood urea nitrogen (21 mg/dL; normal range, 6–20 mg/dL) and hyponatraemia (128 mmol/L; normal range, 136–145 mmol/L). Ultrasound abdomen was performed to rule out liver abscess. A serologic anti-HIV antibody testing was negative. Immunoglobulin and IgG subset analysis were not performed. A lumbar puncture was carried out 3 days after admission showing a turbid colour of the cerebrospinal fluid (CSF), an increased CSF protein level (88 mg/dL; normal range, 15–45 mg/dL) and decreased glucose level (15 mg/dL; normal range, 45–80 mg/dL). The CSF analysis showed white blood cell count 0.131×10^9/L, red blood cell count 100×10^{12}/L, polymorphonuclear 41%, mononuclear 60%. The Gram staining was negative.

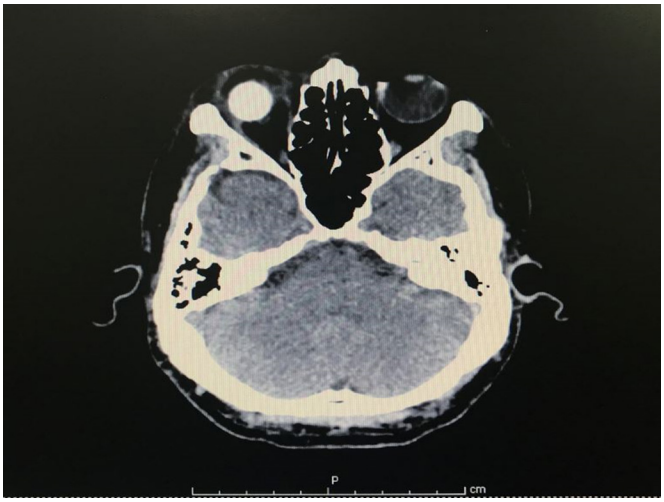

Figure 2 CT brain with contrast media (CM)

DIFFERENTIAL DIAGNOSIS

Three bottles of blood samples and vitreous from the right eye positively revealed *S. suis* infection after 3 days collections. The Gram strain result of vitreous at the right eye showed moderate quantity of Gram-positive coccobacilli. The minimum inhibitory concentrations were 0.094 µg/mL for both penicillin and ceftriaxone. The repeated microbiological culture was negative 2 days upon treatment initiation. Two days after admission, the right eye globe pathology report showed acute endophthalmitis with perforated cornea and vitreous haemorrhage. The audiogram was done at 5-day post admission suggesting irreversible bilateral SNHL disseminated from *S. suis* infection. The tympanic membrane was intact and external auditory canal remained normal.

TREATMENT

Fortified antibiotic ophthalmic solutions (ceftazidime 50 mg/mL at right eye at 1-hour interval and vancomycin 50 mg/mL at right eye at 1-hour interval) and intravenous ceftriaxone (2.0 g at 12-hour interval) and vancomycin (1.0 g at 8-hour interval) were empirically given to treat the infection. Intravenous hydration was administered to treat hypovolaemic hyponatraemia. Ceftriaxone treatment was continued after the microbiological culture confirmed of *S. suis* infection.

The right vitreous tapping, enucleation was performed. Conjunctival peritomy was done. Tenon was undermined and cut separately from sclera. The residual was cleaned and sutured by vicryl stitches. The optic nerve was cut to around 2 mm under the globe. Mull no. 16 and conformer were placed to fit in orbit. One day later, the patient developed a low-grade fever with a temperature of 38.2°C, and experienced a significant loss of hearing in both ears starting from left to right. The CT brain showed no evidence of brain abscess nor leptomeningeal enhancement. There was heterogeneous enhancing lesion adjacent to anterior aspect of right enucleation material and swelling of right extraocular muscles and right optic nerve which might be due to infectious process or postoperative change (figure 2). The condition remained stable without any complications.

After 15 day of admission, the patient recovered from *S. suis* meningitis, septicaemia and spondylodiscitis but suffered from loss of hearing and right eye vision. Ceftriaxone 2.0 g at 12-hour interval until 4-week treatment completion and levofloxacin 500 mg 1.5 tablets per oral for 2 months were prescribed for home medication. In addition, the patient was referred to a nearby local hospital for intravenous ceftriaxone injection.

OUTCOME AND FOLLOW-UP

At 3-month follow-up medical examination, the CT scanning of the temporal bone revealed mild to moderate degree of labyrinthitis ossificans involving both vestibules and all semicircular canals of both ears (figure 3). The basal turn of cochlear was more severe on the left side. Both facial and vestibulocochlear nerve were within normal limit. The videonystagmography at 4 months was done showing bilateral death labyrinths. The right cochlear implant was done at 7 months. A lead electrode was

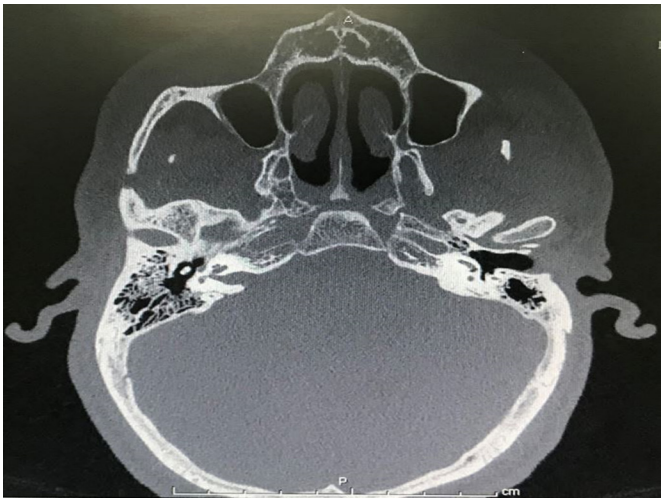

Figure 3 CT scan of temporal bone.

fully inserted into the scala tympani. Impedance/neural response imaging responses were detected. The patient was discharged in a stable condition after a 4-day admission.

DISCUSSION

S. suis is usually an occupational related disease affecting farmers, abattoir workers and butchers in western countries.[6 7] The first case of endopthalmitis secondary to *S. suis* infection was reported in 1978 by McLendon *et al*[14] in which a Caucasian man from the UK who worked at a pork pie factory developing deafness at the right ear and bilateral endophthalmitis was described.[14]

In our case, the patient was an office worker without any occupational exposure to pigs or raw pork. The disease transmission was presumably from raw pork consumption or '*Larb Dib*' he ate 6 days predated the admission. '*Larb*' is a northern Thai dish commonly eaten in raw form in northern Thailand. This emphasises the significant role of raw pork eating in *S. suis* infection. His physical weakness from sleep deprivation and underlying conditions were probably contributing factors resulting in a more susceptible condition to infection. His medical history of regular alcohol drinking was not surprising as raw pork is usually consumed together with alcohol drinks among Thai people especially northern Thai men. Consistently, a considerable large number of alcohol consumption was also noted in studies from Thailand.[13 15 16]

The patient established acute endopthalmitis and infective spondylodiscities in addition to meningitis and septicaemia which are common clinical manifestations of *S. suis*. He developed the symptom shortly after consuming raw pork together with back and joint pain implicating infective spondylodiscites. However, the cause of his illness was not identified until nearly a week after the onset when fortified antibiotic ophthalmic solutions were given. The urgent enucleation due to endopthalmitis with perforated cornea at his right eye was performed and, right blindness resulted.

According to a systematic review, the disease transmission from skin abrasion is believed to be the main route of pathogen entry, however this was noted only in some studies.[10] The presence of fever prior to the ocular symptom suggested that endopthalmitis could be disseminated from *S. suis* septicaemia in this patient.

The property of steroids in reducing inflammatory reactions at subarachnoid space which is a major process causing brain injury and neuronal dysfunction in acute bacterial meningitis has provided the rational of adjunctive corticosteroids use.[17 18] However, the benefit of steroids in reducing hearing loss has been established in paediatric meningitis but has yet to be clearly confirmed among adult population.[19] As *S.suis* infection is mainly found in adults and its effects in this population remains uncertain, using adjunctive dexamethasone may not have reversed the sequelae. In this patient, dexamethasone was not administered. The diagnosis was delayed in the primary setting and permanent SNHL as a consequence of *S. suis* meningitis seemed to be existed before admission. In our views, it still warrants the use of steroids to benefit the patient from its anti-inflammatory property in order to reduce inflammation. A comprehensive counselling session to emphasise on the risk of acquiring *S.suis* infection from raw or partially cooked pork including sour pork or '*Naem*' in raw form should have been mandated.

The high fatality rate in the China outbreak in 2005 involving 215 patients and 38 deaths has emphasised the importance of this zoonosis infection.[20] In this outbreak, the main cause of the widespread of infection was due to backyard slaughtering of sick pigs whereas the deep-rooted behaviour of raw pork consumption seemed to be the predominating cause of the infection in Thailand. Apart from laboratory confirmation, the virulence factors of *S.suis* isolates from the outbreak including *mrp, sly* and *ef* were also related to strains in Europe which are considered to be more virulent than strains identified in North America.[21]

Regardless of the rare incidence of endopthalmitis, physicians should be more aware of the clinical symptoms especially in patient with suggestive clinical presentations and predisposing risk factors of *S. suis* infection. In addition, *S.suis* could also be the causative pathogen causing septicaemia among recent travellers to the Southeast Asia region or endemic areas presenting with fever. A detailed patient interview and history taking to indicate the timelines, relevant exposures and all relevant information are essential for a clear clinical picture. In the absence of available *S. suis* vaccine, early and adequate treatment is crucial to alleviate the disease progress and clinical complications as well as long term neurological sequelae particularly deafness which usually occurs among most *S. suis* meningitis survivors.

Learning points

- *Streptococcus suis* is not uncommon in Asia population where there is an association between traditional eating habits and raw pork consumption, whereas farmers and abattoirs workers in western countries are certainly at high risk of *S. suis* infections.
- *S. suis* infection should be considered in patients presenting with established clinical symptoms and predisposing factors.
- Early detection and appropriate treatment are important to improve symptom outcomes and complications.
- Public health awareness programme is essential especially to those areas where traditional cultural habit of raw pork consumption is still practiced.
- There is limited data concerning *S. suis* epidemiology and molecular characteristics. Further research should be carried out to understand the disease epidemiology, clinical manifestations and treatment regimens.

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