

Case Report

Secondary pyogenic ventriculitis following erector spinae muscle abscess caused by *Streptococcus intermedius*

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Abstract: Background: Pyogenic ventriculitis is a rare and severe cerebral infection. It may result from the rupture of brain abscess, neurosurgical procedures, hematogenous spread and so on. Contiguously diffusion through spinal dura from erector spinae muscle abscess is rare. Moreover, pyogenic ventriculitis caused by *Streptococcus intermedius* is rare. We reported a patient with pyogenic ventriculitis secondary to erector spinae muscle abscess owing to *Streptococcus intermedius* infection. Case presentation: A 61-year-old male with diabetes mellitus had been initially suffered from backache, then presented with fatigue and postural tremor when standing. On the second day of admission, he suffered from retention of urine and disturbance of consciousness. Magnetic resonance imaging of the brain showed pyogenic ventriculitis. And enhanced computed tomography of the lumbar vertebra revealed abscess in the right erector spinae muscle, which helped us identified the pathogen as *Streptococcus intermedius* through the pus drained from the lesions. After antibiotic treatment for two and a half months, association with drainage of the abscess for one month, the cerebrospinal fluid tended to normal. Ventriculo-peritoneal shunt operation was performed at last as the complication of hydrocephalus. The outcome of the patient was relatively good. Conclusion: *S. intermedius* has an affinity for invading the brain. Poor glycemic control with sustained hyperglycemia is the risk factor for abscess formation. Pyogenic ventriculitis can be caused by contiguously diffusion through spinal dura from the erector spinae muscle abscess.

Keywords: Pyogenic ventriculitis, *streptococcus intermedius*, abscess, erector spinae muscle, magnetic resonance imaging

Introduction

Pyogenic ventriculitis (also called pyocephalus or ventricular empyema) is a rare and serious intracranial infection characterized by the presence of suppurative fluid in the cerebral ventricles. The clinical manifestation is usually characterized by headache, vomiting, impaired consciousness with fever and meningeal signs. Although a life threatening condition requiring the earliest diagnosis, symptoms and signs of pyogenic ventriculitis may sometimes be subtle and aspecific [1]. The possible mechanisms of infection of the ventricular system include intraventricular rupture of cerebral abscess, hematogenous spread to the subependyma of the choroids plexus, complication of meningitis and direct implantation of pathogens following head

injury and neurosurgical procedures, e.g, ventricular drain insertion [2, 3]. The exact incidence of pyogenic ventriculitis is unknown.

We here in present an unusual case with pyogenic ventriculitis and erector spinae muscle abscess due to *Streptococcus intermedius* infection.

Case presentation

A 61-year-old male with a past medical history of diabetes mellitus had been suffered from backache followed by pain of the right lower abdomen and testis after one week, with low fever about 38°C. He agreed to sign written informed consents conforming to the tenets of the Declaration of Helsinki prior to the study. He

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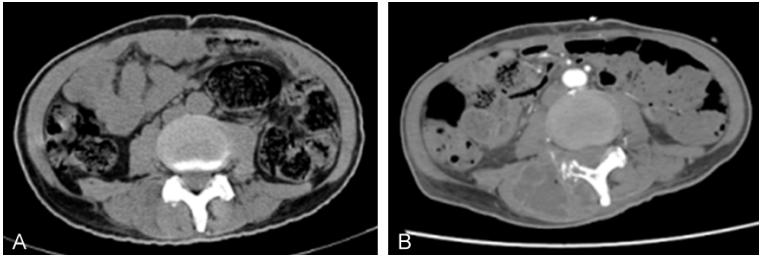


Figure 1. Plain CT and contrast-enhanced CT of lumbar vertebra before admission and on day 5. A: Lumbar vertebra plain CT scan before admission showed the right erector spinae muscle swelling, muscle gap blurred. B: Lumbar vertebra contrast enhancement CT scan on hospital day 5 showed hypodense lesions in the right erector spinae muscle with ring-like enhancement and bony destruction of right vertebral facet joint.

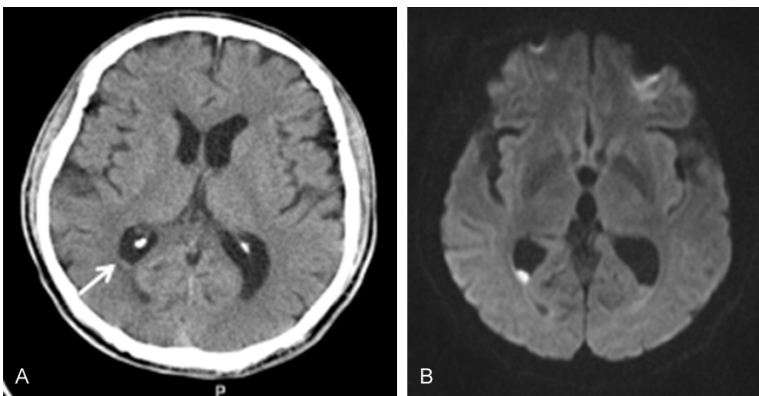


Figure 2. Plain CT and magnetic resonance imaging of the brain on days 3 and 4. A: Brain plain CT scan on hospital day 3 showed less obvious intraventricular isodense lesions (arrow) in bilateral occipital horns which had been lost. B: The diffusion-weighted imaging (b value of 800) clearly indicated hyperintense signals in right occipital horn of lateral ventricles and slightly hyperintensity in the left.

sought treatment at the urinary outpatient clinic. Urinary ultrasound and a computed tomography (CT) scan of the lumbar vertebra and lower abdomen were performed. CT scan showed lumbar intervertebral disc bulging slightly at level of L4-L5 and L5-S1 (**Figure 1A**). He was diagnosed as prolapse of lumbar intervertebral disc and treated with nonsteroidal anti-inflammatory drugs, and was required to rest at home. After ten days, he showed no improvement but suffered from fatigue and mild postural tremor when standing. He sought for treatment again. As the elevated blood glucose was 25 mmol/L, he was admitted to the department of endocrinology for the purposes of further examination and treatment. The patient had lost approximately six kilogram during the past one month. He didn't have a contact history of tuberculosis.

Physical examination showed a fleshless man and responsive slowly. Temperature was 36.5°C, and blood pressure, pulse and respiratory rate, as well pulse oximetry, were normal. The lips were dry and the limbs were slightly strengthless. The rest of the physical examination was normal. Blood tests revealed an elevated white blood cell count (WBC) of 16590/ μ L with a differential count of 93.5% neutrophils. The C-reactive protein (CRP) was high at 148.38 mg/L. Blood culture was negative. Other laboratory examination showed decreased sodium (127.3 mmol/L) and albumin (29.7 g/L), increased glycosylated hemoglobin (13.1%). Test of purified protein derivative of tuberculin and T cell detection of tuberculosis infection were negative.

After admission, the insulin was used to control the blood glucose, and empirical piperacillin/tazobactam and levofloxacin were commenced for infection. On the second day of admission, he suffered

from retention of urine and then became confusion. He could no longer eat a meal, change his clothes, or talk with others. His level of consciousness sharply deteriorated thereafter. An urgent CT scan of the brain was performed on hospital day 3, and showed no obvious abnormalities (**Figure 2A**). So in consultation with the Department of Neurology, according to stiffness of his neck and a positive Kernig's sign, he was diagnosed as suspected encephalitis or meningitis. Then he was transferred to our Neurology unit. Empirical ceftriaxone and acyclovir were commenced for suspected encephalitis and meningitis. Lumbar puncture was performed on the fourth day of admission, but failed. Magnetic resonance (MR) imaging of the brain was performed at the same day, and showed hyperintense lesion on diffusion-weighted image (DWI) with isointense on T2-

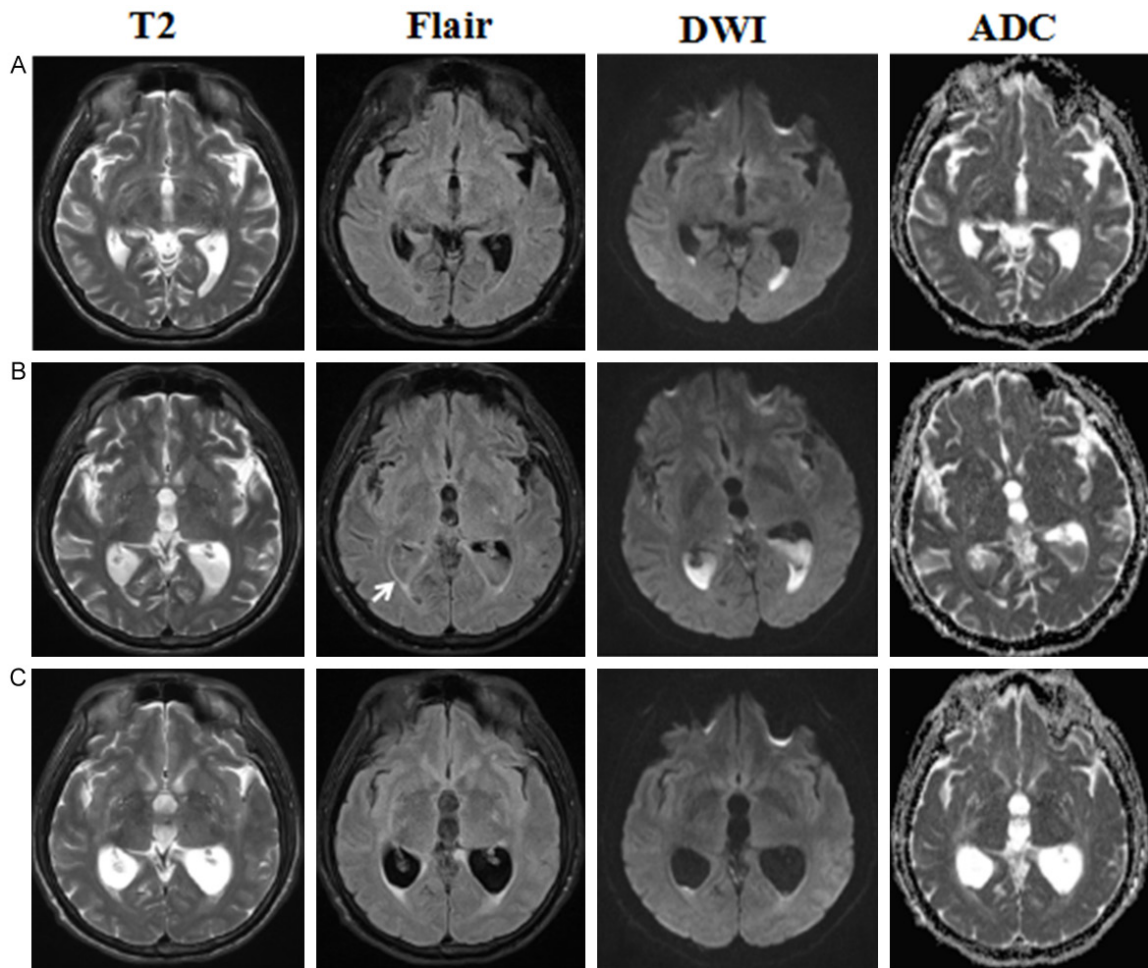


Figure 3. Brain magnetic resonance imaging performed on hospital day 4 (A), day 8 (B), day 39 (C). Intraventricular debris was shown in the occipital horn of the lateral ventricles. The debris was irregular with hyperintense on DWI, isointense on T2WI and hypointense on ADC maps (A, B). Periventricular hyperintense signal was also noted on FLAIR images (arrow). The ventricles enlarged gradually (A-C).

weighted images (T2WI) and hypointense on apparent diffusion coefficient (ADC) at bilateral occipital horns of the lateral ventricles, and periventricular hyperintensity on fluid-attenuated inversion recovery (Flair), which strongly supported infection (**Figure 3A**). Come back to read the cranial CT again, less obvious intraventricular isodense lesions were found in bilateral occipital horns compared with MR imaging (**Figure 2B**). Because the failure of lumbar puncture, a CT scan of the lumbar vertebra was performed again on hospital day 5, and showed a surprising finding. There were hypodense lesions in the right erector spinae muscle at levels of L1 to S1 with ring-like enhancement, and bony destruction were found of right facet joint at level of L4 to L5 (**Figure 1B**). As the unbelievable fact, we checked the back of the patient

again, but didn't find local signs of red, hot, and obvious swell. We read the CT scan of lumbar vertebra before admission once again and found swelling of the right erector spinae muscle. For clarifying the nature of the lesions, an ultrasound-guided aspiration was performed on the sixth day of hospitalization, and revealed large amount of coffee-cream like material. The material was detected for smear and culture with pleocytosis (neutrophils 95%) but no organisms. From the fifth day of hospitalization, the patient suffered from daily fever. According to the purulent material and daily fever, the antibiotic regimen was changed to meropenem 1.0 g three times daily. On the seventh day of admission, repeated contrast-enhanced MR showed increased lesions of bilateral lateral ventricles (**Figure 3B**). At the same day, an

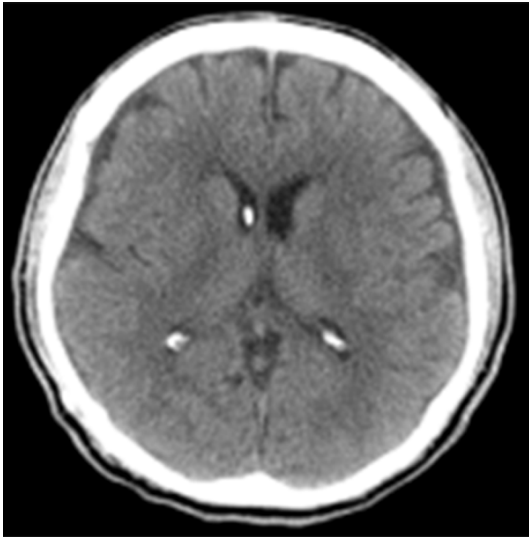


Figure 4. Brain plain CT scan performed one month after operation. Ventriculo-peritoneal shunt was placed and the hydrocephalus was relieved.

ultrasound-guided aspiration of the muscle lesions was performed again and a drainage catheter was left in place. The material extracted from the abscess was smeared immediately and found Gram positive cocci in chain (**Figure 5**). Considering meropenem was not sufficient to control the inflammation, the antibiotic regimen was changed into linezolid associated with meropenem. Five days later, *Streptococcus intermedius* was isolated in the pus culture. It proved to be sensitive to penicillin, cefotaxime, levofloxacin and vancomycin. According to the sensitivity reports and the blood-brain barrier permeability of drugs, the antimicrobial therapy was later readjusted to ceftriaxone and linezolid. On the twelfth day of drainage, cerebrospinal fluid (CSF) like liquid was seen in the drainage pack. Microscopy showed pleocytosis ($300 \times 10^6/l$, polymorphs). Protein content was 2550 mg/l. Three days later, the fluid decreased gradually, and the drainage catheter was removed on day 26. At the same time, the patient was free from fever, conscious awareness, but cognitive function aggravated obviously accompanied by delusion of persecution. Repeated MR of the brain showed gradually decreased dense material but increased hydrocephalus (**Figure 3C**). Intravenous administration of ceftriaxone was terminated on day 38. Two months later, repeated lumbar puncture showed decreased leukocytes count in the CSF to $40 \times 10^6/l$, and protein content was normal. The patient was treated by ventriculo-peritone-

al shunt operation for hydrocephalus. After the operation, oral linezolid was continued for half a month. The patient improved remarkably and can walk alone, with mild to moderate cognitive impairment. CT scan that performed one month after operation did not show any evidence of hydrocephalus (**Figure 4**).

Discussion

Pyogenic ventriculitis is a serious, life-threatening infectious disease of the central nervous system. The clinical manifestations of pyogenic ventriculitis are often aspecific, since consciousness impairment may hide other symptoms, and neurological focal deficits are hardly found when there are no abscesses or other lesions involving the brain [1]. Our patient mainly characterized by motor symptoms, urinary retention and consciousness impairment, but with no evidence of fever and headache. The nonsteroidal anti-inflammatory drugs maybe was responsible for the absence of fever and pain. The most important feature of this case was the discovery of the abscess in the erector spinae muscle, which played an important role in the diagnosis. The pus aspirated from the abscess helped us to identify the pathogenic agent. We believed that the development of pyogenic ventriculitis owed to contiguously spread through the spinal dura, rather than hematogenous spread, because the blood culture was negative. Moreover, the clinical features of this case pointed out that as to signs of paravertebral cold abscess, the diagnosis of bacteriogenic abscess should be considered too, although destruction of intervening intervertebral disc and a paravertebral or psoas abscess were the typical form of spinal tuberculosis [4]. The cause of lumbar puncture failure in our case maybe was due to destruction of lumbar structure or the turbid of cerebrospinal fluid.

In the present case, neuroimaging played a critical role in the diagnosis especially magnetic resonance imaging. The DWI sequences clearly indicated the presence of purulent material within the ventricles. On DWI, hyperintense signals were found in both lateral ventricles, with corresponding decreased signal intensities in the apparent diffusion coefficient maps. Pus in the ventricles was a viscous fluid that consisted of inflammatory cells, cellular debris, microorganisms, and macromolecules. High vis-

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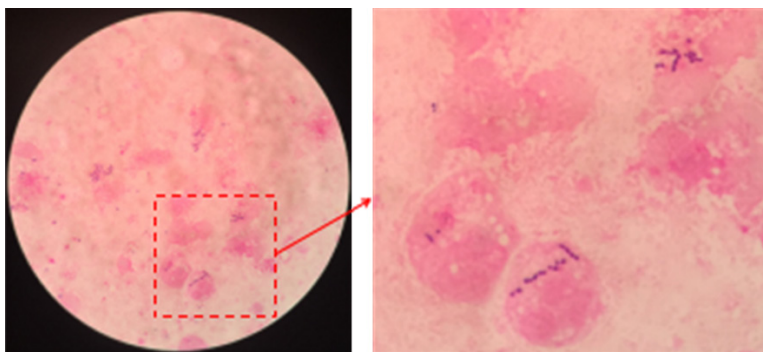


Figure 5. Light microscopy (1000 × magnification) of gram stained pus sample showed gram positive cocci in chain (arrow).

cosity, hypercellularity, and binding of water to macromolecules have been suggested to be responsible for restricted water diffusion [5]. Relative to cerebrospinal fluid, the debris was irregular and slightly hyperintense on T1-weighted images, slightly hypointense on T2WI, and hyperintense on Flair images. Other findings may include hydrocephalus, periventricular hyperintensity and ependymal enhancement [6, 7]. In our case, gradually enlargement of ventricular and periventricular hyperintensity were obvious too. Besides, another advantage of DWI was its high sensitivity for cortical and white matter cytotoxic edema, i.e., acute ischemic infarction, likely due to septic emboli and/or vasculitis. Lummel *et al.* [5] recently described the MR imaging features of 75 patients with acute bacterial meningitis, and revealed that signs of pyogenic ventriculitis were present in 41 cases (54.7%), with ischemic infarctions in 13 cases (17.3%). These MR imaging signs serve to distinguish intraventricular hemorrhage from pyogenic ventriculitis and play a crucial role in the diagnosis of pyogenic ventriculitis. However, in patients who are unstable to undergo MR scans, CT scans can still be informative. The irregular debris morphology is highly specific for purulent material. The ventricular debris sinks towards the occipital horns as the patient supine in the scanner. The surface of the viscous debris is often round or irregular, in contrast to the straight level of acute intraventricular blood. Moreover, we can see that CSF was isolated at the tip of the right occipital horn, by the overlying debris. Lo *et al.* [8] reported a case like this and described it as the 'lodge sign'. Therefore, neuroimaging is a useful noninvasive method for diagnosis as well as follow-up of pyogenic ventriculitis.

The pathogen of pyogenic ventriculitis varies according to geographic regions, the age of the patients, predisposing factors, and the status of immune system. Among immunocompetent adults, *S. pneumoniae* and *Neisseria meningitidis* are the two most common causes of acute bacterial meningitis [9]. In association with nosocomial infection or immunosuppression, the commonest organisms are *Staphylococcus aureus*,

other Gram-positive cocci, *Enterobacter* and *Klebsiella* species [8]. In our case *Streptococcus intermedius* was cultured. They are part of normal flora of the oral cavity and gastrointestinal tract and possess the ability to cause abscesses, especially for the brain and liver. This characteristic can be explained by the virulence factors produced by *S. intermedius*. It produces the enzymes α -N-acetylneuraminidase (sialidase) and hyaluronidase, which destroy host tissues and, presumably, convert them into small nutrients to be utilized in bacterial growth. Moreover, *S. intermedius* specifically secretes a human-specific cytolysin, known as intermedilysin, which triggers the formation of deep-seated infection [10]. In the present case, poor glycemic control with sustained hyperglycemia predisposed the patient to such severe infection.

As to the treatment, there are no guidelines for management of pyogenic ventriculitis. Woehrl *et al.* [11] suggested that systemic intravenous antibiotic therapy seems sufficient to treat pneumococcal meningitis-associated pyogenic ventriculitis. Marinelli *et al.* [1] reported a patient underwent a number of surgical procedures and eventually developed pyogenic ventriculitis. Intravenous vancomycin for 7 weeks and then replaced by oral linezolid were as the antimicrobial management which lead to a complete recovery. Akuzawa *et al.* [12] presented a rare adult case of a left occipital lobe brain abscess that likely resulted from the hematogenous spread of an *Escherichia coli* right renal cyst infection followed by formation of pyogenic ventriculitis. The patient recovered after a 6-week course of meropenem. But Savardekar *et al.* [13] suggested that once the

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diagnosis of pyogenic ventriculitis secondary to brain abscess had been established, it was imperative to start simultaneous intrathecal and intravenous antibiotics, and frontal external ventricular drain should be placed and ventricular CSF drainage should be instituted. In conclusion, a neurosurgical intervention plus an antibiotic regimen is warranted for most patients. However, conservative management with antibiotics may be appropriate in patients too unwell for surgery and with significant comorbidity. The treatment needs to be highly individualized in accordance with the patient's clinical condition, the etiology of pyogenic ventriculitis, the CSF reports, the brain imaging findings, the complications and the pathogens. The patient in this case, ultimately required two and a half months of antibiotic therapy, association with drainage of the erector spinae muscle abscess for one month. Ventriculo-peritoneal shunt operation was performed as the complication of hydrocephalus. The outcome of the patient was relatively good with a sequela of mild to moderate cognitive impairment.

Conclusion

The clinical manifestations of pyogenic ventriculitis are often aspecific, with extremely unfavorable outcome. So early diagnose is important. Neuroimaging has a critical role in the diagnosis. Increased diffusion-weighted magnetic resonance imaging and decreased apparent diffusion coefficient maps are sensitive in pyogenic ventriculitis diagnosis, and are noninvasive methods rather than cerebrospinal fluid sampling. Fluid-attenuated inversion recovery sequences are specialized to detect periventricular signal abnormality. Moreover, *S. intermedius*, found in normal intestinal flora, has an affinity for invading the brain [14-16]. This was the first case report of pyogenic ventriculitis secondary to erector spinae muscle abscess infected by *S. intermedius*. So as to patients with pyogenic ventriculitis and diabetes mellitus, deep infection as a source should be considered. Pyogenic ventriculitis can be caused by contiguously diffusion through spinal dura.

Disclosure of conflict of interest

None.

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