Rare disease

A 51-year-old man with intramedullary spinal cord abscess having a patent foramen ovale

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The authors report a case of a 51-year-old man with intramedullary spinal cord abscess (ISCA) having a patent foramen ovale (PFO). He developed fever and tetraplegia after a recent dental treatment. MRI showed ISCA with longitudinal swelling from the upper cervical to the lumbar spinal cord. Cerebrospinal fluid (CSF) analysis indicated bacterial meningitis, and the culture of CSF revealed *Streptococcus viridans*. Transoesophageal echocardiography revealed the existence of a PFO. We suspected another possibility other than systemic bacteraemia, that paradoxical bacteric embolisation through PFO after the dental treatment caused ISCA. While several reports of brain abscess with PFO are available, this is the first report of ISCA with PFO.

BACKGROUND

Intramedullary spinal cord abscess (ISCA) is a rare infectious disease of the central nervous system (CNS). Only about 120 cases of ISCA have been reported since the first description by Hart in 1830.^{1,2} It usually occurs secondary to bacteraemia from pulmonary, cardiac or urinary infection.³ In addition, the presence of right-to-left shunts such as patent foramen ovale (PFO), atrial septal defect (ASD), or pulmonary arteriovenous malformation (PAVM) would allow pathogens to bypass pulmonary capillary filtering and enter arterial circulation more easily.⁴ Here, we report a case of ISCA with PFO that occurred after a dental procedure.

CASE PRESENTATION

A 51-year-old man visited a dentist and received treatment for his decayed teeth 1 month before. Two weeks later, he had abdominal distention and constipation and visited his family doctor. More a week later, he ran a fever and developed weakness in the lower limbs and ischuria and was admitted to a local hospital. His condition exacerbated gradually, and he developed paraplegia. Thus, he was admitted to our Kasai City hospital for further examination.

At admission, he complained of weakness and numbness in the trunk and all extremities. Physical examination revealed that his body temperature was 37.3°C. His consciousness had deteriorated mildly with manic mood. Meningeal signs were positive, and Horner syndrome was observed in his right eye. He had severe muscle weakness, especially in both his legs, with complete paraplegia. Sensation of all the modalities below the level of C5 was lost. He also had bladder and bowel disturbance.

INVESTIGATIONS

Laboratory studies revealed increased white blood cell (WBC) count of 14 400/mm³ with 85% polymorphonuclear cells and serum C reactive protein level of 11.4 mg/dl.

Cerebrospinal fluid (CSF) analysis indicated bacterial meningitis with pleocytosis of 2667/mm³ (85% polymorphonuclear cells), elevated protein content of 400 mg/dl and decreased glucose level of 11 mg/dl. CSF culture revealed growth of *Streptococcus viridans*. T2-weighted MRI showed extensive swelling at almost the entire length of the spinal cord and in the upper cervical to lumbar segments. Gadolinium-enhanced T1-weighted MRI showed a rim-enhanced lesion extending from Th2 to Th7 vertebral level (figure 1). ISCA with bacterial meningitis was diagnosed.

Chest and abdominal CT and Gallium scintigraphy showed no other obvious focus of infection. Transthoracic echocardiography was normal. While transoesophageal echocardiography showed no evidence of vegetation, a bubble test revealed a PFO with a right-to-left shunt on Valsalva manoeuvre.

DIFFERENTIAL DIAGNOSIS

Malignant lymphoma, neoplastic infiltration or transverse myelitis which occurs by multiple sclerosis, neuromyelitis optica, tuberculosis, sarcoidosis, viral myelitis may show similar features as ISCA in MRI.

TREATMENT

We immediately started intravenous antibiotics treatment of meropenem (3 g/day) and vancomycin (2 g/day) as well as a steroid-pulse therapy (for 3 days of methylprednisolone 1 g/day) and intravenous immunoglobulin (for 3 days of 5 g/day). Despite improvement in his febrile pattern and laboratory data (WBC count, serum C reactive protein level, CSF cell count, etc), he developed dysphagia and intractable hiccup and could not turn his head 8 days after admission. His consciousness level and respiratory condition remained stable. MRI showed that the spinal cord abscess had extended up to the medulla oblongata

Figure 1 MRI at admission. (A) T2-weighted sagittal image. (B) T1-weighted sagittal image with contrast enhancement.

(figure 2.) We successfully treated his condition by changing the antibiotics (ampicillin 4 g/day) with an additional series of steroid-pulse therapy.

OUTCOME AND FOLLOW-UP

Antibiotics treatment was continued for 4 weeks in all, and the serum C reactive protein level decreased to 0.4 mg/dl. He was able to walk with parallel bars and was transferred to a rehabilitation centre 3 months after admission.

DISCUSSION

The cause of ISCA in children is the contiguous spread of infection from skin lesions such as dermal sinus tract. ISCA is thought to occur in adults via the haematogenous spread of pathogens from a cardiopulmonary or urinary source. However, in 65% of ISCA cases, the primary infectious

focus remained unknown.³ In this case, there was no other focus responsible for the development of ISCA except for poor oral hygiene and a history of dental procedure. Bacteria can enter the blood stream from even very mild manipulations of the mouth. The oral cavity harbours more than 500 bacterial species. Just 1 mg of dental plaque may contain more than 10^{11} microorganisms. Bacteraemia was observed in 100% of the patients after dental extraction, in 70% after dental scaling and in 20% after endodontic treatment.⁵

ISCA and brain abscess usually occur secondary to bacteraemia from infection focus. Several reports have demonstrated the relationship between brain abscess and right-to-left shunts, including PFO (table 1).^{5–15} But, there had been no report that demonstrated relationship between ISCA and PFO from the direct observation of right-to-left shunt. The presence of right-to-left shunts

Figure 2 MRI at 8 days after admission. (A) T2-weighted sagittal image. (B) T1-weighted sagittal image with contrast enhancement. (C, D) T1-weighted axial image with contrast enhancement at the line.

Table 1 The cases of the abscess with PFO

Year	Author	Age (years)/sex	Place of abscess	Anamnesis	Pathogens
2001	Kawamata <i>et al</i> ¹¹	36/M	Brain	Periodontitis	Could not be detected
		63/M	Brain	Alveolar abscess	<i>Streptococcus</i>
2003	Friedlander <i>et al</i> ¹⁰	58/F	Brain	Dental procedure	<i>Streptococcus milleri</i> <i>Haemophilus aphrophilus</i>
2003	Listernick ¹⁴	2/F	Brain	Vomiting, >48 h	<i>Haemophilus parainfluenzae</i>
2006	Doepp <i>et al</i> ⁹	67/F	Brain	Perirectal abscess	<i>Escherichia coli</i>
2006	Khouzam <i>et al</i> ⁴	43/M	Brain	Caries, gingivitis	<i>Streptococcus viridans</i>
2006	LaBarbera <i>et al</i> ¹³	63/F	Brain	Dental procedure	<i>Streptococcus intermedius</i>
		43/F	Brain	Migraine	<i>Streptococcus intermedius</i>
2007	Stathopoulos <i>et al</i> ⁶	53/M	Brain	COPD Dental procedure	<i>Staphylococcus haemolyticus</i>
2008	Keith-Rokosh <i>et al</i> ¹²	51/M	Brain	A heavy drinker	MRSA
2008	Wolf and Curtis ¹⁵	12/M	Brain	Dental procedure	<i>Haemophilus aphrophilus</i>
2008	Chen <i>et al</i> ⁸	51/M	Brain	Verruca on PFO	<i>Staphylococcus aureus</i>
2010	Ariyaratnam <i>et al</i> ⁷	53/M	Brain	Dental procedure	<i>Aggregatibacter paraphrophilus</i>

COPD, chronic obstructive pulmonary disease; F, female; M, male; MRSA, methicillin-resistant *Staphylococcus aureus*; PFO, patent foramen ovale.

such as PFO, ASD or PAVM would allow pathogens to bypass these pulmonary defences and enter the arterial circulation more easily.⁴ Although PFO is a cryptic condition, it might be a cause of right-to-left shunting under increased pressure of the right atrium at straining for stool or Valsalva manoeuvre.⁶ Thus, PFO is a possible route for the passage of emboli from the venous into the arterial circulation, and the association between PFO and cryptogenic ischaemic stroke has been well known. Patients with PFO have a high risk of recurrence of embolic disease.^{4 16} Brain abscess and ISCA may have similar aetiological features and abscess development process. That is, the presence of PFO also indicates a high risk of ISCA not only of brain abscess.¹⁶ Hoche *et al*¹⁷ demonstrated that the introduction of only microorganisms into the circulation of experimental animals did not result in the development of ISCA. However, if both microorganisms and thrombi were introduced, ISCA developed.³ Although

even bacteraemia alone could cause ISCA, PFO would increase the risk of ISCA by becoming the cause of paradoxical bacterial embolism.

In ISCA, early diagnosis and treatment are essential for neurological improvement. Advances in modern diagnostic imaging can be major help in making a conclusive diagnosis. Analysis of the CSF is helpful to diagnose ISCA that accompanies bacterial meningitis. However, 25–40% of the abscesses in CNS are microbiologically sterile even when appropriate culture techniques are used under best settings.¹⁸ Fortunately, we were able to detect *S viridans* from the CSF culture. *S viridans* exist in the human oral cavity as one of the most frequent pathogens of the normal flora. This group of bacteria is one of the most common constituents of the flora found in empyemas, bacteraemia or endocarditis and is particularly associated with abscess formation. Many literatures showed the importance of immediate administration of antibiotics over a sufficiently

long period for good prognosis.^{3 18} If no obvious focus or risk factor of infection is found, unsanitary oral cavity can be a focus of infection. The presence of a right-to-left shunt and a history of paradoxical embolism would indicate high risk of recurrence. In such cases, the right-to-left shunt must be closed.

The mortality (4%) for recent cases of ISCA was significantly lower than that (90%) reported in the 'preantibiotic' era.¹ Despite the improvement in survival rates, neurological deficits persisted in the majority (60%) of patients who survived.¹ The lack of information about this emerging infectious condition can lead to misdiagnosis and the delay of appropriate treatment, resulting in a miserable prognosis. The possibility of ISCA should always be considered in patients with transverse myelitis.

Learning points

- ▶ The possibility of ISCA should always be considered in patients with transverse myelitis
- ▶ Prompt diagnosis and appropriate treatment will result in better prognosis.

Competing interests None.

Patient consent Obtained.

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