

Unusual presentation of more common disease/injury

A neurosurgical presentation of patent foramen ovale with atrial septal aneurysm

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Summary

We describe a case of cerebral abscess in a 53-year-old lady with a background of congenital heart defect. She has an atrial septal defect with atrial septal aneurysm, which remained undiagnosed until this clinical presentation. She presented with a short history of right-sided hemiplegia and neuroimaging revealed a heterogeneous lesion in the left frontoparietal region. Neuronavigation-guided left frontoparietal craniotomy was performed to debulk the lesion and preoperatively frank pus was drained, which grew *Streptococcus constellatus*. She was successfully treated with antibiotics for 6 weeks and her clinical condition improved. We believe that the patients' previous dental extraction has possibly resulted in a paradoxical embolism through the atrial septal defect bypassing the pulmonary circulation. The MRI scan picture was misleading, as it was initially thought to be a high-grade brain tumour.

BACKGROUND

Patent foramen ovale (PFO) is a congenital cardiac defect that often persists into adulthood.¹ Atrial septal aneurysm (ASA) is frequently found in association with PFO and involves a redundant and hypermobile interatrial septal tissue in the region of the fossa ovalis. Although usually entirely asymptomatic, PFO and ASA can have unusual

clinical manifestations including cryptogenic ischaemic stroke.^{2 3} Cerebral abscess is a rare complication, thought to occur by shunting of bacteria from the right to the left circulation, bypassing the filtering capacity of the pulmonary capillary bed. Cerebral abscess is a very unusual presentation of an undiagnosed PFO. To date, pathogens reported mainly come from the oral flora. We describe



Figure 1 Plain CT scan of brain.

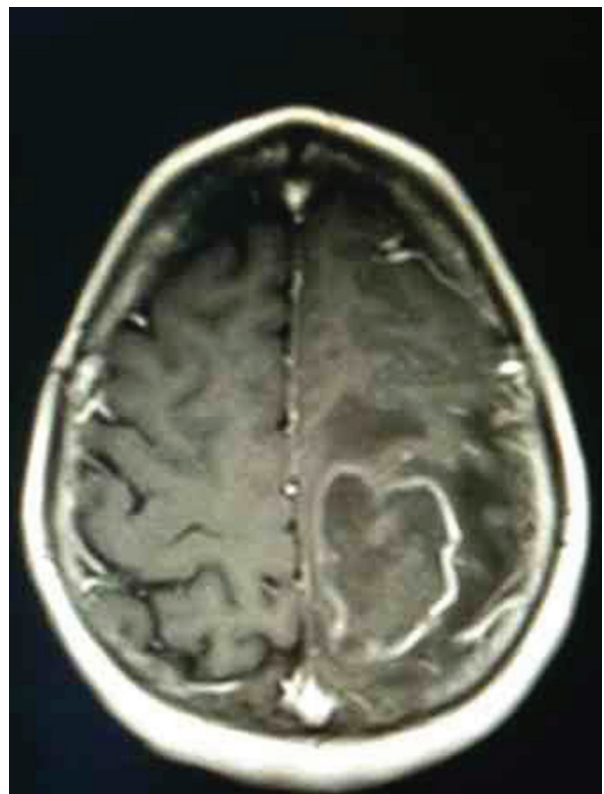


Figure 2 Postcontrast T1-weighted MRI brain.

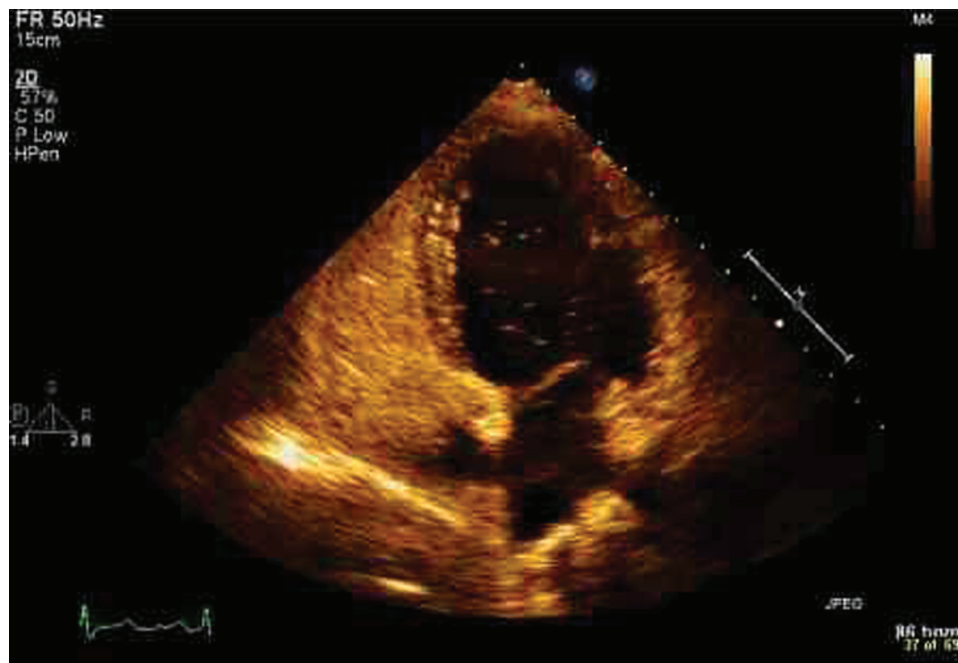


Figure 3 2D echocardiography demonstrating the PFO + ASA (arrow).

the first documented case of a latent PFO+ASA presenting with a *Streptococcus constellatus* brain abscess.

CASE PRESENTATION

A 53-year-old right-handed, non-smoking female presented to us with a 2-week history of weakness and 'clumsiness' of the right upper and lower limbs (RUL; RLL). She also complained of paraesthesia of the RUL and RLL and intermittent 'shaking' of the right hand. She had a mild headache for 1 day prior to admission with no vomiting or visual disturbances. On this initial presentation, she was fully orientated in time, place and person with a Glasgow Coma Score (GCS) of 15/15. She was afebrile and her motor power was 5/5 in all limbs with normal sensory examination. Cranial nerve examination revealed a mild right upper motor neuron facial paralysis. Babinski sign was positive in the right side. Her heart sounds were normal with no audible murmur. The medical history included epilepsy (seizure-free for 1 year), migraine, osteoporosis and asthma. She had a dental extraction 3 weeks prior to admission following loose upper incisors. Her regular medications were levateracitam 750 mg twice daily (by mouth), fluticasone/salmeterol inhaler 250 mg twice daily and glucosamine 400 mg twice daily (by mouth) and calcium supplements. She had a strong family history of breast carcinoma.

INVESTIGATIONST

Haematological investigations showed a normal white cell count and C reactive protein. CT brain pre and postcontrast revealed a heterogeneous area of increased attenuation in the left parietal lobe about 3 cm in diameter, with haemorrhage inside it (figure 1). There was enhancement in the periphery of the mass after contrast injection, and extensive surrounding oedema with local effacement of the sulci and of the left lateral ventricle suggestive of a high-grade brain neoplasm.

A mammogram and CT of thorax abdomen and pelvis were performed which failed to reveal any source of metastatic neoplastic disease. She was referred to neurosurgical service for further management. At this stage, she developed dense right hemiplegia with worsening of facial weakness. MRI brain pre and postcontrast (figure 2) was performed to evaluate the lesion further. Diffusion weighted imaging (DWI) with apparent diffusion coefficient (ADC) map MRI brain was suggestive of haemorrhagic lesion than cerebral abscess.

DIFFERENTIAL DIAGNOSIS

- Primary high-grade Glioma
- Cerebral abscess

TREATMENT

With this background, the patient was offered surgical excision of the lesion. She underwent a left parietal craniotomy and debulking of the space-occupying lesion. Peroperatively, 10 ml of frank purulent material and clotted blood were evacuated. These samples were sent for pathology and microbiology. Histopathology revealed acute inflammatory response and gram staining of samples showed gram-positive cocci.

OUTCOME AND FOLLOW-UP

Pending culture and sensitivity, she was commenced empirically on ceftriaxone 2 g intravenous twice daily, flucloxacillin 1 g intravenous four times daily and metronidazole 500 mg intravenous thrice daily. Three days later, full cultures revealed growth of *S. constellatus* sensitive to flucloxacillin. She responded well to antibiotics and dexamethasone and her right limb weakness gradually improved to 3/5. As a part of diagnostic investigation she had a trans-thoracic bubble echo study at rest and with provocative manoeuvres (Valsalva and cough). This was positive for a PFO with

aneurysmal atrial septum (figure 3). Postoperative CT and MRI brain scan (figure 4) showed satisfactory drainage of the abscess. She has completed her 6 weeks course of antibiotics and her weakness has improved considerably. She is currently mobilising independently and undergoing rehabilitation.

DISCUSSION

The foramen ovale is a flap-like structure that exists in foetal life allowing communication between the left and right atria. This allows oxygenated blood in the foetal circulation coming from the umbilical vein to bypass the undeveloped pulmonary circulation. The foramen ovale should close shortly after birth due to the acute increase in left atrial pressure. In some people, this mechanism fails and the foramen ovale remains open, persisting even into adulthood. Prevalence of PFO in the general population is 25–30%.^{4–5} ASA involves a redundant and hypermobile interatrial septal tissue in the region of the fossa ovalis. It is a common finding on transthoracic echo^{6–8} and is frequently associated with an intracardiac shunt.⁹

The main focus in the literature to date regarding PFO and ASA has been on their role in paradoxical embolism in the setting of cryptogenic ischaemic stroke. It has been well established that PFO with ASA is a risk factor for stroke, and indeed stroke patients with both abnormalities are at higher risk of recurrent ischaemic events.¹⁰ Theoretically, clots may originate from the venous circulation,¹¹ from within the PFO or ASA⁶ or from the atria in the setting of paroxysmal arrhythmia.¹² They bypass the normal filtration mechanisms of the pulmonary capillary bed and gain access to the arterial circulation. Emerging data also reveal an increased prevalence of migraine in patients with PFO and ASA, possibly due to vasoactive migraine triggers entering the cerebral circulation via the atrial septal defect.¹³ Interestingly, this particular patient had a long history of migraine since her teenage years. There is no data, however, on infective embolism in the setting of PFO and ASA.

We postulate a similar mechanism of paradoxical embolism of infected emboli to the brain in this case, with pathogens entering the bloodstream during the dental extraction, possibly seeding at the site of the ASA and then accessing the cerebral arterial circulation via the PFO. It is debatable whether or not the ASA was incidental or contributory to the seeding of infection. The pathogen isolated here, *S. constellatus*, is one of the *Streptococcus milleri* group. These non-haemolytic viridians streptococci are normally found in the flora of the oral cavity and gastrointestinal tract and are known for their propensity for abscess formation.¹⁴ *S. constellatus* has long been established as a cause of brain abscess.^{15–17} However, there is no evidence from literature to suggest pathogenesis of cerebral abscess by this pathogen in the setting of intracardiac shunting.

Another unusual feature of this case was the radiological imaging, which was quite atypical for brain abscess. The degree of haemorrhage within the lesion and the heterogeneity of the surrounding ring enhancement initially suggested malignancy, either metastasis or a high-grade glioma. This highlights the difficulty often encountered in differentiating tumour from abscess with conventional MRI techniques. The use of DWI and ADC mapping have been

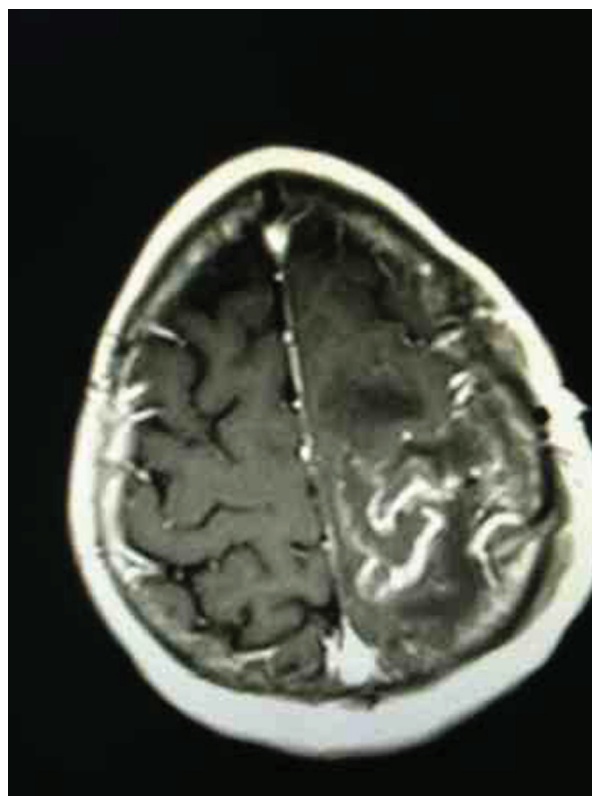


Figure 4 Postop MRI T1-weighted brain.

examined as aids in distinguishing between the two.^{18–19} These MRI techniques depend on the molecular motion of water. Pyogenic abscess should give increased signal on DWI and low ADC values due to restricted diffusion within the purulent centre of the abscess. This case is an exception to this generalisation, highlighting the necessity of tissue diagnosis in ambiguous cases. MR spectroscopy (MRS) may be beneficial in such circumstance to differentiate tumour from abscess based on the biochemical component present in the abnormal brain tissue.

Learning points

- ▶ Pyogenic brain abscess can occur from an otherwise undiagnosed congenital cardiac condition (PFO+ASA) in a healthy individual.
- ▶ Obtaining a detailed history from the patient is essential as there is a chance of bias from the radiological imaging.
- ▶ Neuroimaging, particularly MRI brain with DWI/ADC map is very helpful in differentiating abscess from tumour.
- ▶ Appropriate antibiotic treatment usually results in successful clinical recovery after primary treatment, drainage of the abscess, in the case described.
- ▶ Tissue biopsy may be needed in case of doubt regarding the clinical diagnosis.

Competing interests None.

Patient consent Obtained.

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