CASE REPORT

Sinus venosus atrial septal defect presenting with brain abscesses in a 77-year-old immunocompetent patient

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SUMMARY

We present a case of 77-year-old immunocompetent patient who initially presented with vertigo, nausea and vomiting, with symptoms later progressing to headaches and increased lethargy. Brain MRI revealed ring-enhancing lesions typical of abscesses in the right cerebellum. Transoesophageal echocardiogram (TEE) was performed to look for the source of his abscesses, and uncovered a sinus venosus type atrial septal defect. Cardiac CT was carried out; the patient was found to have a sinus venosus atrial septal defect with partial anomalous pulmonary venous return. Moreover, dental examination showed multiple dental caries with poor oral hygiene. The patient was started on intravenous empiric antibiotics and steroids. Subsequent brain imaging showed almost complete resolution of the abscesses. The patient's symptoms started to improve, and he was eventually sent to an intensive rehabilitation centre with future plans to surgically correct his congenital heart disease to prevent further complications.

BACKGROUND

Congenital heart disease (CHD) when discovered in adulthood is an interesting entity. Detailed history taking, physical examination and appropriate imaging can usually reveal the anomaly. A common form of CHD is atrial septal defect (ASD). What is rare about our case is the age of presentation and that ASD was the source of the brain abscesses.

CASE PRESENTATION

A 77-year-old African-American man with medical history only significant for hypertension, presented to our emergency department, with a 3-day history of nausea, vomiting and vertigo. He was noted to be hypoxic to 88% on room air, and had distended jugular veins, but otherwise physical examination, EKG and laboratory work up were all within normal limits. A CT pulmonary embolism study with venous run-off was performed in the emergency department due to the reportedly new hypoxia and normal chest X-ray, which showed no alveolar infiltrates, pulmonary embolism or deep vein thrombosis; however, it did reveal prominent pulmonary arteries. The patient was admitted for intractable symptoms. During day 2 of hospitalisation, he developed a low grade fever and an abnormal cerebellar examination including abnormal finger to nose and heel to shin tests, and

progressive lethargy. Empiric broad spectrum antibiotics were given prior to non-contrast brain CT, which did not show any abnormalities. A lumbar puncture was then performed to evaluate for meningitis. Cerebrospinal fluid analysis was remarkable for a white cell count of 12 000 with neutrophilic predominance. Gram stain and cultures, however, remained negative. Brain MRI was also performed to better evaluate the posterior fossa. MRI showed multiple ring-enhancing lesions involving the right cerebellum with surrounding vasogenic oedema, the largest measuring 1.2×2.0 cm. The patient was maintained on empiric vancomycin and meropenem. Steroids were also started for the oedema. Given the close vicinity of the lesions to the 4th ventricle, aspiration of the abscesses was not feasible. To look for the source of the brain abscesses, the patient underwent transoesophageal echocardiography (TEE). Of note, the patient had undergone an initial TTE performed on day 1 of admission, due to his jugular vein distention, hypoxia and enlarged pulmonary arteries, and this study, however, had been unrevealing of any abnormalities, because of a suboptimal study due to poor acoustic windows. On TEE, however, it was discovered that he had a sinus venosus type ASD, and possible anomalous venous return, but no vegetations; he also had evidence of a right-to-left shunt (figure 1). To further characterise his cardiac anomaly, cardiac structural CT was performed, which showed the sinus venosus ASD and the right superior pulmonary vein draining into the superior vena cava (SVC) representing partial anomalous venous return, with the SVC largely draining into the left atrium. Given the right to left shunt and the cryptogenic source of the abscesses, odontogenic flora spreading to the brain was suspected,

Figure 1 Bicaval view from TEE. RA right atrium, LA left atrium, IVC inferior vena cava, SVC superior vena cava. The arrow points to the atrial septal defect.

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Figure 2 To the right, brain MRI of the patient at the time of presentation showing right cerebellar ring-enhancing lesion. To the left, a repeat brain MRI after 6 weeks of intravenous antibiotics showing almost complete resolution of the lesion.

for which the patient underwent dental evaluation. Dental evaluation showed multiple tooth caries with poor oral hygiene. Hence, the patient attended oral treatment and dental extraction. During the hospitalisation, the patient's symptoms improved. A repeat MRI of the brain after 6 weeks of intravenous antibiotics showed almost complete resolution of brain abscesses and of the oedema (figure 2). The patient was discharged to a rehabilitation centre, with a future plan to surgically correct his sinus venosus ASD.

DISCUSSION

Brain abscesses are rare entities, with 1500–2500 cases seen annually in the immunocompetent host.¹ Different mechanisms have been described in the literature regarding the mechanisms of bacterial spread to the brain, including contiguous extension from a close inoculation such as in traumatic or postoperative brain abscess formation. However, 25% of brain abscesses are still identified as cryptogenic without obvious source of infection.² In cryptogenic brain abscesses, spread of oropharyngeal flora to the brain is strongly believed to be the source of the abscess. As in our case, poor dental hygiene has been previously identified as a risk factor for brain abscess. One study contends that a transient bacteraemia may occur in healthy persons from flora normally found in the oral cavity due to simple hygienic procedures such as tooth brushing.³ Normally, these potentially pathogenic organisms are contained within the venous circulation, and would be unable to survive the oxygen-rich environment of the pulmonary vascular bed and the actions of the alveolar macrophages.⁴ Although some bacteria might escape this first line of defence, they would be eliminated by the lymphatic system. However, a right-to-left shunt would bypass these defences and allow these organisms to enter the arterial circulation and thus reach the brain.⁵ While surgical treatment and appropriate antibiotic are the mainstay treatment for brain abscesses, in our case, a surgical approach was not feasible given the close proximity of the abscesses to the 4th ventricle.

ASDs are common congenital heart diseases in adults. There are three major types of ASDs: ostium secundum, ostium primum and sinus venosus. Sinus venosus atrial septal defects (SVASDs) represent 4–11% of all ASDs.⁶ The superior form of SVASD develops as a result of the absence of the superior border of the atrial septum, with an overriding SVC. SVASDs are commonly associated with anomalous pulmonary venous return.⁷ Diagnosing SVASDs can be difficult, requiring special imaging such as TEE.⁸ Although in most cases TEE is enough to make the diagnosis to proceed to surgical correction, some cases

might need more sophisticated imaging, such as cardiac CT or MRI, to further delineate the anomaly's anatomy.⁹ Catheter closure of SVASD is not possible, which makes surgical correction of the anomaly the principal treatment.

It is important to keep in mind that, in patients with right-to-left shunt, paradoxical phenomena including abscesses and thrombi are part of their natural history, if these had not been surgically corrected. In our case, it was surprising that the patient never developed criteria of systemic inflammatory response (SIR). Which prompted us to rule out causes of immunodeficiencies including HIV infection and others. Very few case reports have described ASD with right-to-left shunt presenting with paradoxical phenomena such as a brain abscess, and all of them were younger than 50 years of age.¹⁰

Learning points

- Brain abscesses should always trigger further evaluation to look for the source of infection, including regular transthoracic echocardiography (TTE).
- Dilated right atrium or elevated pulmonary artery systolic pressure on TTE should always prompt further work up.
- Surgical drainage and appropriate antibiotics are the mainstay treatment for brain abscesses.
- Congenital heart disease should still be on the list of differentials of paradoxical abscesses even in the elderly.

Competing interests None declared.

Patient consent Obtained.

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