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Novel Partial Anomalous Pulmonary Venous Connection Associated with Brain Abscess

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Background Right-to-left vascular shunts are associated with brain abscess.

Case Report We present a 47-year-old female with a cryptogenic left thalamic abscess on which *Streptococcus mitis* grew upon aspiration. Computed tomography of the chest with contrast agent revealed an anomalous connection between the left superior pulmonary and brachiocephalic veins. A right-to-left shunt was confirmed in a transthoracic echocardiogram study in which bubbles were injected into the left arm; this shunt had not previously been noted upon right-arm injection.

Conclusions We recommend aggressive evaluation for right-to-left shunts in patients who present with cryptogenic brain abscesses. In addition to imaging, this should include a bubble-based study with left-arm saline injection.

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Key Words cryptogenic, brain abscess, pulmonary shunt.

Introduction

Brain abscesses classically result from extension of nearby infection in the head, penetrating head injury, neurosurgery, or hematogenous spread from a remote infectious source through right-to-left shunting.¹ However, up to 30% of brain abscesses have no such associations and thus are deemed cryptogenic.³

Examples of right-to-left shunting are cyanotic heart disease and pulmonary arteriovenous malformations (PAVMs),^{1,4} which form the basis of current recommendations for prophylactic antibiotics in patients with such conditions and for prophylactic closure of PAVMs.⁵ We report an unusual case of a right-to-left shunt found in the setting of a cryptogenic brain abscess.

Case Report

A 47-year-old woman presented with worsening headache and

neck pain over 4 days associated with nausea and vomiting. She denied subjective fevers or chills. She reported traveling to the Dominican Republic 1 month prior to presentation. She had no other medical history and was not on any medications. She denied any recent illnesses or dental procedures.

Her temperature was 38.0°C, heart rate was 70 beats/min, respiratory rate was 18/min, and blood pressures were 140/72 mm Hg. The patient had no evident toxicity and was alert and oriented to person and time but not to place. An oral examination showed good dentition with no notable abscess, pain, or gingival disease. A cardiovascular examination revealed no abnormalities, including murmurs. There was no evidence of skin infections or lesions. She had mild expressive aphasia with clear, unpressured speech. Reception and comprehension were intact. Her cranial nerves and the results of motor and sensory examinations were normal. However, she did have a shuffling gait with loss of balance to the right and clonus at the right ankle.

The laboratory parameters of the patient at admission were remarkable for leukocytosis with a left shift (17.8×10^9 WBCs per liter and 90% neutrophils). The results of electrocardiogram and chest X-ray examinations were normal. Computed tomography (CT) of the brain without contrast agent demonstrated a left thalamic mass with a hypodense center and hy-

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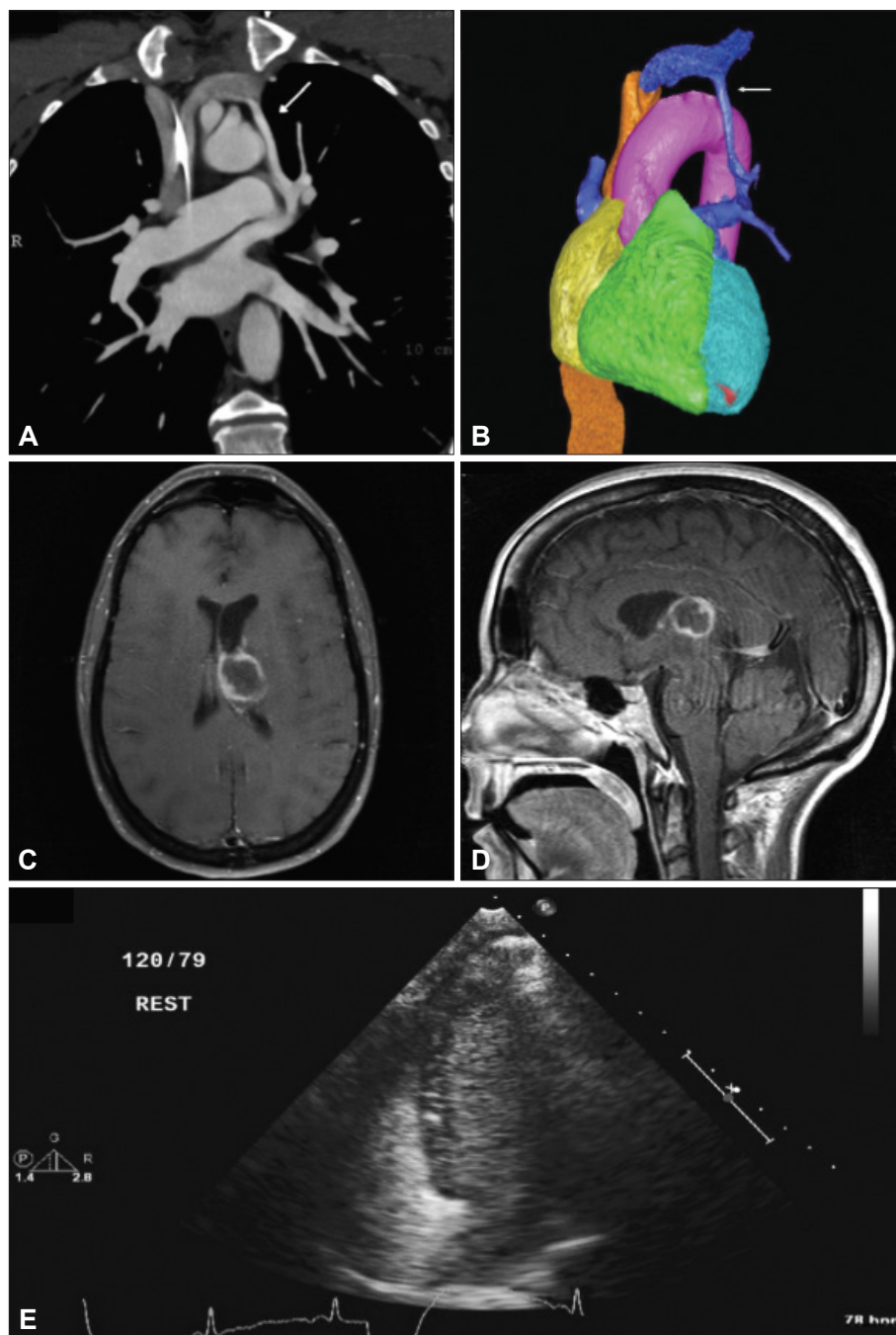


Fig. 1. A: Computed tomography angiogram of the chest. Arrow indicates an anomalous pulmonary venous connection. B: 3-D false-color reconstruction of panel A. Arrow indicates an anomalous pulmonary venous connection. C: Postcontrast T1-weighted transverse MRI scan with rim-enhanced left-side lesion. D: Postcontrast T1-weighted sagittal MRI scan with rim-enhanced left-side lesion. E: 2-D echocardiogram obtained during left-side injection of agitated normal saline.

perdense periphery. A mass effect was noted on the left lateral and third ventricles. Well-aerated paranasal sinuses and mastoid cells were noted. Magnetic resonance imaging of the brain with and without contrast agent revealed a 2.4×2.2×1.9-cm lesion with mild surrounding hyperintense T2-weighted and fluid-attenuated-inversion-recovery (FLAIR) signals compatible with edema (Fig. 1). The enhanced rim demonstrated a hypointense T2-weighted signal. The internal contents of the lesion were hyperintense on T2-weighted and FLAIR signals with corresponding restricted diffusion. These

imaging findings were highly suggestive of a brain abscess.

Stereotactically guided drainage was applied immediately to the patient. Five milliliters of foul-smelling, purulent fluid was aspirated. The patient was initially started on vancomycin, ceftriaxone, and metronidazole after cultures were obtained. *Streptococcus mitis* grew in these cultures, and so the antibiotic coverage was appropriately narrowed.

The patient subsequently underwent transthoracic echocardiography (TTE) and transesophageal echocardiography with agitated-saline contrast agent injected into the right arm; the

Table 1. Previously reported vascular malformations

Malformation	History	Therapeutic intervention	Age (years)	Authors
SVC to left atrium	Previous brain abscess followed by "partial seizure"	Endovascular closure	12	Singh et al. ¹⁴
SVC to left atrium	Previous brain abscess followed by "presumed seizure" and then multiorgan failure secondary to sepsis	None*	33	Hirth et al. ²
SVC to left atrium	Cerebral abscess with a negative workup	Endovascular closure	46	Troost et al. ¹⁰
Superior segmental pulmonary artery to aorta collaterals	Fever, depressed mentation, and cyanosis	Surgical resection	22	Walder et al. ¹⁵

Previously reported cardiopulmonary vascular malformations associated with brain abscess excluding HHT, hepatopulmonary syndrome, structural heart disease, and congenital pulmonary artery-vein complexes. In total, 186 articles dating back to 1893 were reviewed.

*No intervention secondary to death.

HHT: hereditary haemorrhagic teleangiectasia, SVC: superior vena cava.

results were unrevealing. A CT scan of the chest was performed with contrast agent on postoperative day 13 due to a transient episode of hypoxia, which revealed an incidental finding of an anomalous connection between the left brachiocephalic vein and the left superior pulmonary vein. No PAVMs were noted.

Diagnostic cardiac catheterization via the left basilic vein confirmed the anomalous venous connection and the presence of a predominantly left-to-right shunt based on measurement of the oxygen saturation. TTE with agitated saline injected into the left arm this time demonstrated the passage of a large amount of bubbles into the left side of the heart. The patient recovered well after abscess drainage and was discharged to a rehabilitation facility with plans for close follow-up, and eventual percutaneous closure of the anomalous connection.

Discussion

Bacteremia secondary to gingival or other dental disease may lead to brain abscess due to seeding of bacteria via valveless emissary veins that allow direct flow into the venous drainage system of the brain.^{1,6} Similarly, bacteremia from more-distant infections may bypass typical pulmonary filtration via anomalies that allow right-to-left shunting [e.g., cyanotic congenital heart disease, PAVMs, patent foramen ovale (PFO), and persistent venous connections to the arterial system]; thereby gaining access to the arterial structures of the brain.^{1,4}

The idea that a right-to-left shunt may predispose to cerebral abscesses has long been presumed in patients with cyanotic congenital heart disease. Cyanotic heart disease has accounted for 12.8–69.4% of all cases of brain abscesses in several series,⁷ and the incidence of brain abscess in patients with cyanotic heart disease has been reported to range between 5% and 18.7%.⁸

Similarly, PAVMs have been noted to be associated with

both cerebral infarcts and abscesses. In one large series, the prevalence rates of cerebral abscess were 8% and 16% in patients with a single and multiple PAVMs, respectively.⁹ The standard recommended care regimen at high-volume centers is closure of PAVMs prophylactically in order to reduce their cerebral complications.⁵

While the latter two groups of patients appear to implicate persistent right-to-left shunts in the pathogenesis of brain abscesses, it has only recently been appreciated that intermittent right-to-left shunts may also predispose patients to abscesses. Kawamata et al.¹ were the first to report two patients with cryptogenic brain abscesses that had PFOs, and suggested that brain abscesses can arise from paradoxical emboli across a PFO. Several case reports have since noted the presence of PFOs associated with cryptogenic brain abscesses.^{1,4,10} Most compellingly, Mahadevan et al.¹¹ reported on a series of 68 consecutive patients with cerebral abscesses. Of their eight cryptogenic abscesses, seven had evidence of right-to-left shunting: five PFOs, one PAVM, and one persistent shunt from the superior vena cava (SVC) to the left atrium.

Another conduit for intermittent right-to-left shunting involves anomalous systemic venous connections. The most common anomaly of the superior caval circulation is persistence of the left SVC (Table 1).¹² Most (92%) of the left SVCs drain into the right atrium via the coronary sinus, which appears echocardiographically as a dilated coronary sinus. The remaining 8% drain directly into the left atrium, creating a right-to-left shunt that has been reported to be associated with cerebral abscess.¹⁰

We have reported a novel anomalous venous connection associated with a cerebral abscess. Connection of the pulmonary veins to the systemic venous circulation is rare, and represents a type of partial anomalous pulmonary venous connection. The four most-common conduits in order of decreasing frequency are 1) pulmonary veins from the right upper and

or middle lobe to the SVC, usually with a sinus venosus atrial septal defect, 2) all of the right pulmonary veins to the right atrium, 3) all of the right pulmonary veins to the inferior vena cava, and 4) the left upper or both left pulmonary veins draining via an anomalous vertical vein to the left brachiocephalic vein¹³ (as in our case).

The case reported herein is also notable for the negative results obtained in a bubble-based study involving right-arm injection—the right-to-left shunt was only revealed after injecting agitated-saline contrast agent into the left arm. Since right-to-left shunts are strongly associated with cryptogenic brain abscesses, an aggressive evaluation that includes imaging and a bubble-based study with left-arm injection should be considered in such cases. This approach could facilitate both defining the pathogenesis and tailoring treatment for cryptogenic brain abscesses.

Conflicts of Interest

The authors have no financial conflicts of interest.

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