

## CASE REPORT

# *Streptococcus sanguis* brain abscess as an initial manifestation of pulmonary arteriovenous malformation

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**Abstract**

Pulmonary arteriovenous malformation is a vascular anomaly that predisposes to complications of paradoxical embolization including stroke and brain abscess. Here, we present a case of brain abscess associated with pulmonary arteriovenous malformation.

**KEYWORDS**

brain abscess, pulmonary arteriovenous malformation, *Streptococcus sanguis*

## 1 | INTRODUCTION

Pulmonary arteriovenous malformations (PAVMs) are rare pulmonary vascular anomalies when direct communications between the branches of pulmonary artery and pulmonary veins are established without an intervening pulmonary bed. It causes right to left shunt, and if not detected and treated early, it may result in severe neurological complications including brain abscess and stroke.<sup>1,2</sup> Majority of PAVMs occur as part of hereditary hemorrhagic telangiectasia (HHT), whereas the remaining cases are sporadic.<sup>1</sup>

*Streptococcus sanguis* is one of the oral commensal bacteria found in dental plaques and is an unusual cause of intracranial infection.<sup>3</sup> We report a case of brain abscess in which *S sanguis* was isolated from the pus culture of the lesion, and PAVM was subsequently diagnosed by neuroimaging studies as the underlying cause of brain abscess.

## 2 | CASE REPORT

A previously well, 24-year-old man presented with a one-week history of gradual onset of right-sided body weakness, associated with fever, headache, and vomiting. He denied any history of trauma, seizures, bleeding tendency, or recent

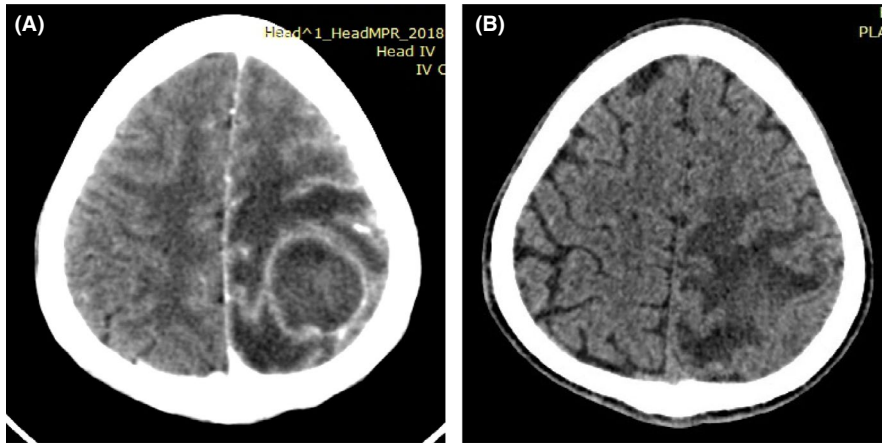
dental procedure. On admission, he was alert and orientated but noted to have bilateral digital clubbing. A pulmonary bruit was heard on auscultation posteriorly over the left lung base, accentuated by inspiration. He was not cyanosed or hypoxic. Neurological examination revealed weakness of the right upper and lower limbs, graded as 4/5 using the Medical Research Council scale. Brisk reflexes and decreased muscular tone were demonstrated in the right upper and lower limbs. A positive Babinski reflex was elicited in the right plantar. Examination of the cranial nerves did not show any neurological deficit. He was noted to have poor oral hygiene and dental caries. There was no mucocutaneous telangiectasia.

The laboratory investigations including differential white cell count and C-reactive protein were all within the normal limits. The differential diagnoses on admission were a cerebrovascular event or a space-occupying lesion including a cerebral abscess. A contrast CT scan of the brain was undertaken urgently and showed an abscess in the left parietal lobe (Figure 1A). A chest radiograph was done because of the pulmonary bruit noted during physical examination showed a well-demarcated, lobulated nodule at the left lower zone (Figure 2).

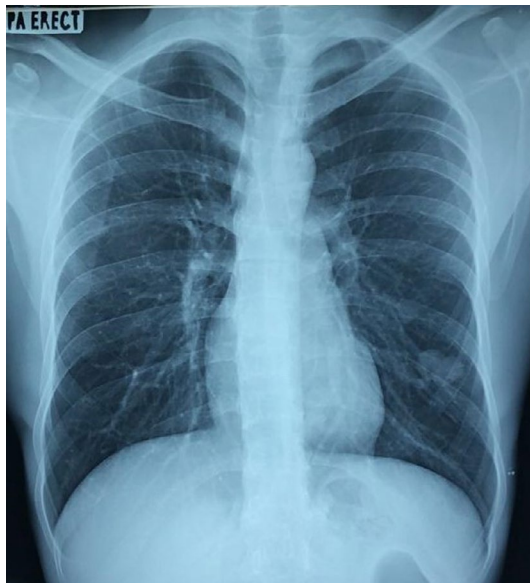
He subsequently underwent left burr hole surgery and ultrasound-guided drainage, in which 20 cc of thick yellowish, foul-smelling pus was evacuated from an encapsulated

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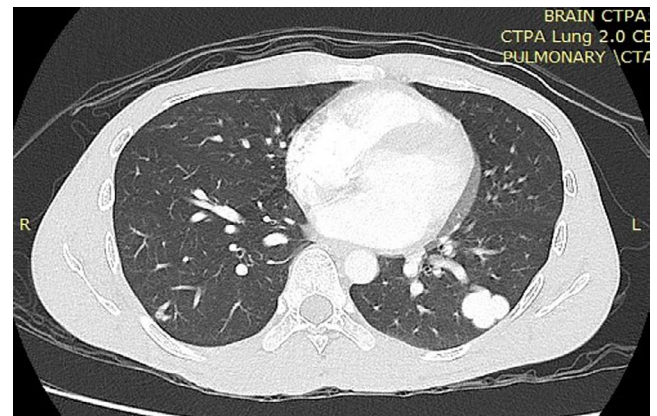
**FIGURE 1** A, CT of the brain showed an abscess with rim enhancement and perilesional edema in the left parietal lobe. B, Resolution of the brain abscess with reducing perilesional edema after treatment



**FIGURE 2** Chest radiograph showed a lobulated radio-opaque mass in the left lower zone

abscess underneath the dura. *Streptococcus sanguis* was isolated from the pus culture and was sensitive to ceftriaxone, erythromycin, cefepime, and linezolid. Histopathological examination of the excised abscess wall was reported as features consistent with an acute inflammation with no evidence of any underlying malignancy.

Due to the high clinical suspicion for PAVM from the physical examination and chest radiograph findings, CT of thorax and pulmonary angiography was done and confirmed the presence of a left lower lobe PAVM (1.7 × 2.3 cm) (Figure 3). The arterial supply derived from the left upper lobe superior segmental branch with draining vein into left the inferior pulmonary vein. Transthoracic echocardiography showed good cardiac contractility with no valvular lesion or vegetation. The patient subsequently underwent tooth extraction while as an inpatient, and culture of the dental pulp also yielded *S sanguis* with similar sensitivities.



**FIGURE 3** CT pulmonary angiography showed a pulmonary arteriovenous malformation in the superior segment of the left lower lobe

He was treated for *S sanguis* brain abscess with intravenous ceftriaxone 2 g twice daily for total 6 weeks' duration. Repeated brain CT after completion of antibiotic showed resolution of brain abscess (Figure 1B). He made a full recovery with complete resolution of the weakness of his right upper and lower limbs with physiotherapy. He was subsequently scheduled for an elective transcatheter embolization of the PAVM.

### 3 | DISCUSSION

Pulmonary arteriovenous malformations are abnormal direct communications between the pulmonary artery and vein, and can present with dyspnoea, epistaxis, haemoptysis, telangiectasias, cyanosis, clubbing, and gastrointestinal bleeding.<sup>1</sup> PAVMs can lead to serious complications as abnormal vascular connections in PAVMs impairing the normal filtering function of lungs and serve as a source of paradoxical embolism and systemic infections.<sup>2</sup> As a consequence, neurological complications can occur, and these include migraine

(43%), transient ischemic attacks (37%), strokes (18%), brain abscess (9%), and seizures (8%).<sup>4</sup>

Up to 60%-90% of congenital PAVMs are associated with hereditary hemorrhagic telangiectasia (HHT).<sup>1</sup> HHT is an autosomal dominant disorder and can be diagnosed with at least two out of four manifestations: telangiectasia, epistaxis, family history, or visceral involvement.<sup>5</sup> The patient described in this case report did not fulfill the criteria for the diagnosis of HHT, neither did he have any acquired causes of PAVM such as chest surgery, trauma, actinomycosis, schistosomiasis, hepatic cirrhosis, and metastatic carcinoma.<sup>6</sup> Therefore, it is likely that the PAVM is idiopathic.

In this present case, we postulated that the cerebral abscess is caused by preceding odontogenic infection as cultures of the brain abscess and dental pulp both yielded the same organism, *S sanguis* with similar sensitivities. This finding was consistent with a study by Boother et al<sup>7</sup> in which 37% of cerebral abscesses had association with untreated dental infections or dental healthcare access. *Streptococcus Sanguis* is a Gram-positive facultative anaerobe of the *Streptococcus viridans* group and a commensal of the human oral cavity. A case report of brain abscess due to *S sanguis* in association with PAVMs was previously reported by Nakamura et al<sup>8</sup> It was hypothesized that the invasion of *S sanguis* from a ruptured nasomucosal vein, subsequently forming septic embolic, which passed through a pulmonary arteriovenous shunt, and eventually contributed to the development of a brain abscess.

Brain abscess can occur as a complication of PAVM. The incidence of brain abscess in patients with PAVM was reported to be around 5%. Momma et al<sup>9</sup> reported that the parietal lobe was the most frequently involved site of brain abscess, which was accountable for 18 of the 53 cases (34%). In the management of brain abscess associated with PAVM, resection or embolization of the PAVM has been advocated in order to prevent recurrent brain abscesses and paradoxical embolization.<sup>2</sup>

Chest radiography is a valuable tool in the diagnosis of PAVMs. The characteristic radiographic features of PAVMs include an oval or round mass of uniform density, mostly lobulated with varying sizes from 1 to 5 cm in diameter, and majority of PAVMs are located in the lower lobes.<sup>2</sup> Chest CT scan is regarded as the gold standard diagnostic tool in PAVMs in view of its ability to provide high anatomical resolution, precise location, and differentiating type of PAVM (simple versus complex).<sup>10</sup> PAVMs can be treated by angiographic embolization or surgical resection.<sup>2</sup>

Our case demonstrated the crucial role of multidisciplinary team working in the management of such a complex patient with multiorgan involvement which included the clinical input of neurosurgery, medicine, dentistry, radiology, and physiotherapy which contributed a significant part in the patient's full neurological recovery.

## 4 | CONCLUSION

Pulmonary arteriovenous malformations are rare pulmonary vascular anomalies that often remain asymptomatic before the development of a first serious complication. Clinicians should be aware that brain abscess can be the first clinical manifestations of PAVMs as a result of right to left shunt. This case highlights that PAVMs should be considered as a possible cause of brain abscess in the absence of an obvious precipitating cause.

## ACKNOWLEDGMENTS

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## CONFLICT OF INTEREST

The authors have no conflicts of interest to declare.

## AUTHOR CONTRIBUTIONS

CYC: prepared the manuscript and involved in patient care. CSYC: contributed to data curation and assisted in manuscript writing. ELCO: provided guidance and edited the final manuscript.

## ETHICAL APPROVAL

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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