Case Report

Brain abscess as the first manifestation of pulmonary arteriovenous malformation: A case report

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Abstract

Pulmonary arteriovenous malformations (PAVM) are rare pulmonary vascular anomalies. Although most patients are asymptomatic, right to left shunt produced by PAVM, could result in easy access of septic or non-septic emboli to systemic circulation, end to serious central nervous system (CNS) complication. Here we report a case of brain abscess in a young man. Its source was initially unknown but multiple arteriovenous malformations were detected incidentally in his thoracic CT, which was performed for ruling out embolism. Although the cases of brain abscesses associated with PAVM are very rare, the brain abscess could be an initial clinical manifestation in asymptomatic PAVM as in the case presented in this report.

Key Words: Arterivenous malformation, brain abscess, pulmonary

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INTRODUCTION

Pulmonary arteriovenous malformations (PAVM) are rare pulmonary anomalies characterized by abnormal connections between pulmonary arteries and veins resulted in intra pulmonary right to left shunt.[1,2]

Most PAVM do not come to attention until early adulthood and then may present with pulmonary and non-pulmonary symptoms.

Among pulmonary symptoms, hemoptysis and dyspnea are more common and non-pulmonary symptoms are most often related to CNS disorders.

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PAVM may be sporadic or associated with hereditary hemorrhagic telangiectasia (HHT) or Osler-Weber-Rendu syndrome.

Here we described a patient with pulmonary AVM who was first manifested with brain abscess with unknown source. To the best of our knowledge, searching Pubmed by keywords of pulmonary arterivenous malformation, brain abscess, Osler-Weber-Rendu and Iran, covering all fields, there has been no reported similar case in the Iranian population.

CASE REPORT

A 25-year-old male came to our hospital, had complains of headache, nausea, vomiting, and fever from five days ago. The general physical examination was unremarkable and neurological exam was also normal.

Primary hematologic and biochemistric laboratory data were unremarkable, except for mild polycythemia. Lumbar puncture (LP) was performed and cerebrospinal

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fluid (CSF) analysis revealed increased protein and white blood cells (mostly neutrophils), but CSF direct smear and also CSF and blood culture were negative.

For more evaluation, brain MRI was performed which showed hyposignal and round lesion (about 3.5 cm) in left occipital lobe in T1w image [Figure 1], associated with high signal intensity capsule and obvious prelesional edema in T2w image [Figure 2] and no signal loss in Fluid Attenuated Inversion Recovery sequence [Figure 3], so brain abscess was the first differential diagnosis for patient.

Then patient underwent craniotomy and occipital brain abscess was the first impression of surgeon during surgery.

Direct smear of lesion's samples showed many gram positive cocci which was culture positive and identified as streptococcus viridians, so gram positive abscess was confirmed.

Patient was not drug abuser and in cardiological consult and echocardiaographic exam, there was not any evidence of endocarditis, so, initial source of brain abscess remained undetermined.

Three days post operation he suffered from right lower limb pain and swelling; he underwent a Doppler ultrasound exam and deep vein thrombosis of right superficial femoral vein was confirmed, then thoracic multislice CT scan was requested to rule out pulmonary thrombo embolism (PTE) because mild dyspnea of patient.

Pulmonary CT angiography was performed with special PTE protocol by 64 multislice CT, and we found partial filling defects in segmental branches of right lower lobe pulmonary artery which were compatible with segmental PTE [Figure 4], but we were also surprised to see multiple obvious pulmonary arteriovenous malformations (AVM) as multiple masses or nodular densities with well defined and lobular margin containing feeding artery and draining vein [Figures 5-7].

After detecting multiple AVM, patient was reassessed and while reviewing his history and physical examination, we found history of recurrent epistaxis (with frequency of three to four times a week) and patient informed that episodes of epistaxis are usually after eating hot food stuff. So far, the patient did not yet have the need for nasal packing or blood transformation, conversely he had high hemoglobin (about 15.5) in the day of admission, indicating mild polycythemia. This polycythemia could be due to degrees of hypoxemia secondary to multiple arterivenous shunts, which is similar to other types of right to left shunts.

Also, his father and one of his sisters had recurrent epistaxis without any other complications.

In repeated physical exam, we noted telangiectatic lesions in his leg and ankle, so, four of four curacao criteria[3] for HHT were fulfilled and Osler-Weber- Rendu syndrome was confirmed for the patient. There was no obvious clubbing or cyanosis. Also, abdominopelvic sonographic study was unremarkable and there was no any hepatic, splenic, or pancreatic lesion. Consultation with radiologist for interventional procedures and also with surgeon was requested for patient, but unfortunately patient left hospital on his own request and responsibility and refused further therapeutic options.

DISCUSSION

Abnormal vascular connections in PAVM results in bypassing filtering effect of pulmonary capillaries with

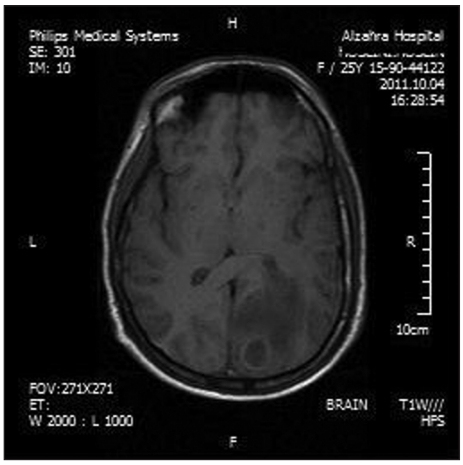

Figure 1: T1W image of brain

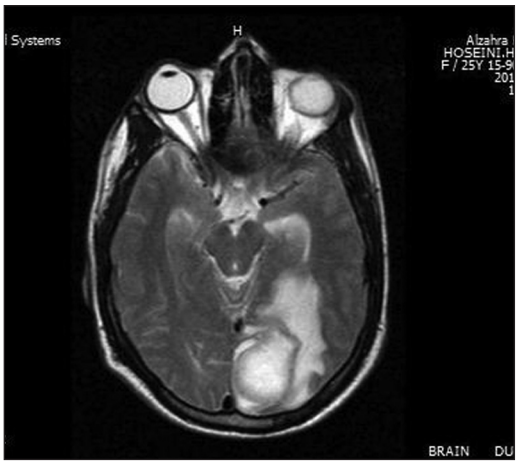

Figure 2: T2W image of brain

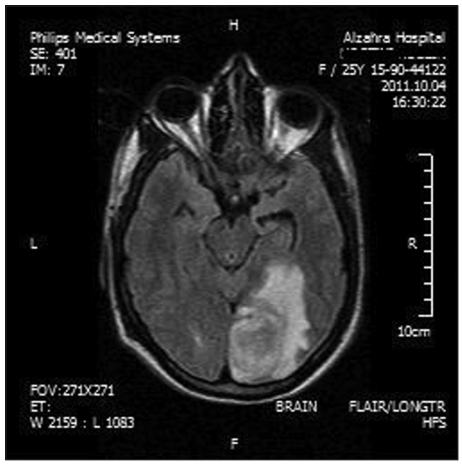

Figure 3: FLAIR image of brain

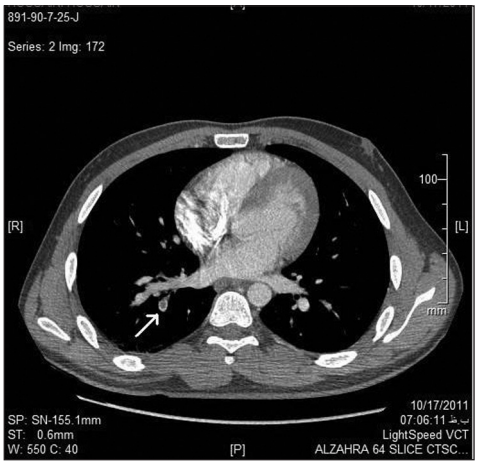

Figure 4: Thoracic CT shows filling defect in Segmental branch of RT lower lobe(arrow)

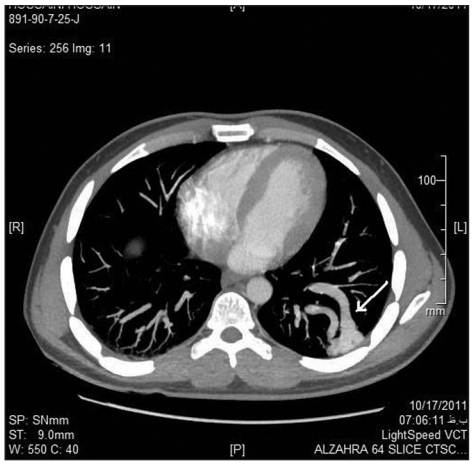

Figure 5: Thoracic CT reveals AVM of left lower lobe (arrow)

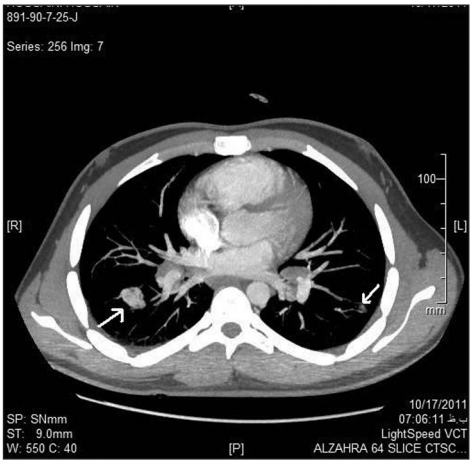

Figure 6: Thoracic CT shows AVM in Superior segment of both lower lobes (arrows)

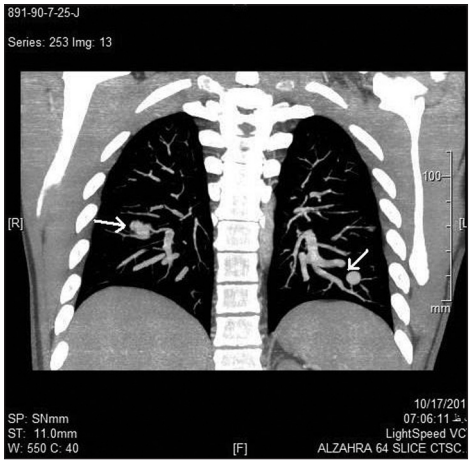

Figure 7: Thoracic CT shows AVM in each lower lobe (arrows)

potential increased risk of paradoxical septic or non septic emboli. In consequence, the brain is usually the first and most frequently target and overally right to left shunts predispose patients to cerebral abscess or stroke.[3,4] Apart from this, right to left shunt in PAVM cause hypoxemia, dyspnea, and polycythemia.

PAVM is commonly associated with hereditary hemorrhagic telangiectasia (HHT) or Osler-Weber-Rendu syndrome. In this way, about 70% of all patients with PAVM have HHT and conversely, from known cases of HHT, about 15% have PAVM.[5]

Each patient who is suspected to suffer from HHT should be evaluated to rule out the presence of visceral vascular malformations. Lungs, liver, and brain are the most common locations with vascular malformation in this syndrome.[6]

Although PVAM occurs rarely, it is common in rare autosonal dominant HHT, which occurs about one in 5000.[3] This syndrome is characterized by four criteria, known as curacao criteria, which are: 1) spontaneous recurrent epistaxis 2) telangiectasia 3) proven visceral AVM 4) first degree relation with HHT.

If three or four criteria are fulfilled, definite diagnosis of HHT will be considered, while two present criteria indicate possible HHT and in the case of only one criteria, this syndrome is unlikely.[3]

In our patient, telangiectasia, visceral AVM, and recurrent epistaxis were present and regarding to history of epistaxis in his father and sister, first degree relative with HHT was also highly suggested, considering all these factors HHT was confirmed for him.

Mathis *et al.,* described brain abscess in his twenty six cases with HHT and noted that in all cases pathogens as anaerobic germs and particularly streptococcus was detected and also noted that against non-HHT cases with brain abscess, in HHT subjects no staphylococcus infection was reported.[7]

It is known that brain abscess is the most serious neurological complication of PAVM and occurs in about 5 to 10% of patients with PAVM.[8]

Pulmonary PAVM in HHT patients is mostly located in the lower lobe of lungs,[9] as in our case. Idiopathic PAVM are similar in shape and imaging findings, except for greater number of solitary versus multiple AVM and also lack of lower lobe predominance, in idiopathic cases.[10]

Brain abscess of our case was in occipital lobe, however, as Momma *et al.,* reported[11] parietal lobe is the most common site of abscess.

The diagnosis is usually straight forward on CT scan,[12-14] and pulmonary CT angiography is the gold standard for diagnosis.[15]

Therapeutic options include angiographic embolization and also surgical resection. All cases of PAVM need treatment for preventing possible complications. Although surgery is the treatment of choice in patients with isolated lesions; however, in patients with multiple lesions, embolization is the selective method.[16]

Finally, it is recommended to search for PAVM in patients with cryptogenic brain abscess.[17]

REFERENCES

1. Klein JS. Pleura, chest wall, diaphragm and miscellaneous chest disorders. In: Brant WE, Helms CA, editors. Fundamentals of diagnostic radiology. 3rd ed. USA: Lippincott Williams and Wilkins; 2007. p. 556.
2. Kinoshita T, Gohara R, Koga T, Sueyasu Y, Terasaki H, Rikimaru T, *et al*. Visualization of pulmonary arteriovenous malformation by three dimensional computed tomography: A case report. Kurume Med J 2003;50:161-3.
3. Lee YS, Kim S, Kang EK, Park JD. A case of hereditary hemorrhagic telangiectasia. Korean J Pediatrics 2007;50:1018-23.
4. Hirth A, Disney P, Thorne S. Brain abscess associated with an unusual cause of right to left shunt. Heart 2007;93:34.
5. Dines DE, Arms RA, Bernatz PE, Gomes MR. Pulmonary arteriovenous fistulas. Mayo Clin Proc 1974;49:460-5.
6. Tabakow P, Jarmundowicz W, Czapiga B, Czapiga E. Brain abscess as the first clinical manifestation of multiple pulmonary arteriovenous malformations in a patient with hereditary hemorrhagic telangiectasia (Rendu-Osler-Weber disease). Folia Neuropathol 2005;43:41-4.
7. Mathis S, Dupuis-Girod S, Plauchu H, Giroud M, Barroso B, Ly KH, *et al*. Cerebral abscesses in hereditary haemorrhagic telangiectasia: A clinical and microbiological evaluation. Clin Neurol Neurosurg 2011;114:235-40.
8. Hall WA. Hereditary hemorrhagic telangiectasia (Rendu-Osler-Weber disease) presenting with polymicrobial brain abscess. J Neurosurg 1994;81:294-6.
9. Wang HC, Yang PC, Kuo SH, Luh KT. Pulmonary arteriovenous malformation: Analysis of 10 cases. J Formos Med Assoc 1998;97:97-100.
10. Wong HH, Chan RP, Klatt R, Faughnan ME. Idiopathic pulmonary arteriovenous malformations: Clinical and imaging characteristics. Eur Respir J 2011;38:368-75.
11. Momma F, Ohara S, Ohyama T, Moto A, Okada H, Harada H. Brain abscess associated with congenital pulmonary arteriovenous fistula. Surg Neuroln 1990;34:439-41.
12. Lawler LP, Fishman EK. Multi-detector row CT of thoracic disease with emphasis on 3D volume rendering and CT angiography. Radiographics 2001;21:1257-73.
13. Lawler LP, Fishman EK. Arteriovenous malformations and systemic lung supply: Evaluation by multidetector CT and three-dimensional volume rendering. AJR Am J Roentgenol 2002;178:493-5.
14. White RI Jr, Pollak JS. Pulmonary arteriovenous malformations: Diagnosis with three-dimensional helical CT-a breakthrough without contrast media. Radiology 1994;191:613-4.
15. Khurshid I, Downie GH. Pulmonary arteriovenous malformation. Postgrad Med J 2002;78:191-7.
16. Pejhan S, Rahmanjoo N, Farzanegan R, Rahimi M. Surgically treatable pulmonary arterivenous fistula. Ann Thorac Cardiovasc Surg 2012;18:36-8.
17. Drouet A, Le Moigne F, Donat A, Guilloton L, Felten D. Brain abscess as the first clinical manifestation of isolated pulmonary arteriovenous malformation without Rendu-Osler disease. Rev Neurol 2011;167:29-34.

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