Brain Abscess in Young Adult with Double Outlet Right Ventricle (DORV): A Case Report

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Abstract

Intracranial lesions frequently occur in association with congenital malformation of the heart. Intracranial abscess and cerebral thrombosis
are the two most serious complications of the brain due to congenital cyanotic heart disease. We reported a case of brain abscess occurring in patients with double outlet right ventricle (DORV). The identification of focal infection and appropriate treatment with parenteral antibiotics,steroid, antiplatelet and anticonvulsant improved patients clinically.

Keywords: brain abscess; congenital; double outlet right ventricle

Introduction

Intracranial lesions frequently occur in association with congenital malformation of the heart.Intracranial abscess and cerebral thrombosis are the two most serious complications of the brain due to congenital cyanotic heart disease.[1]

Case

A 20-year-old girl presented with acute onset partial secondary generalized seizures few hours before admission. Patient lost her conciousnes during seizures that occurs three times and come off about five minutes. She had no fever, meningismus, signs of raising intracranial pressure, head injury, or previous history of seizures. She has already diagnosed for cyanotic congenital heart defect since four month old. Her vital sign were within the normal limit. The remarkable physical examinations were abnormal heart sounds which are pansistolic murmur at left lower sternal border, and also clubbing finger in the extremities.

The electrocardiogram (ECG) showed right atrial and ventricle hypertrophy with right axis deviation (Figure 1).

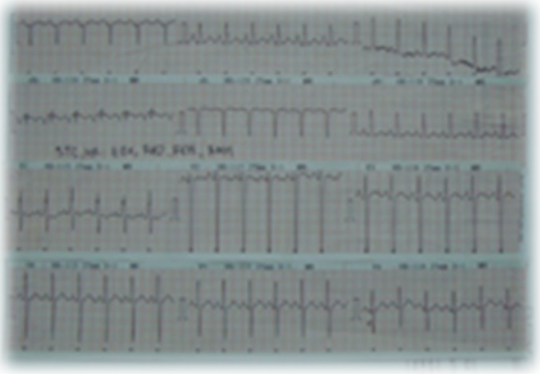
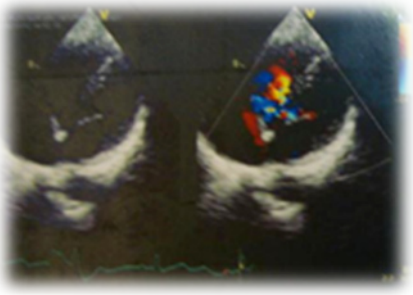


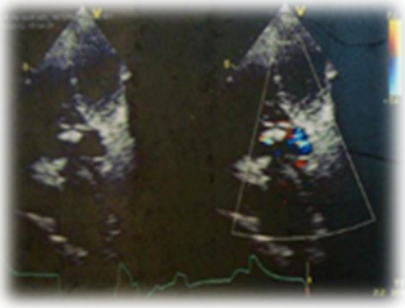
Figure 1. The electrocardiogram showed sinus tachycardia with right atrial enlargement and right ventricle hypertrophy. The axis is right axis deviation.

The remarkable laboratory findings were increasing hemoglobin (Hb 18,6 g/dL) and red blood count (RBC) 7,730/µL, however there was no increasing of white blood count (WBC). Blood cultures and bacterial sensitivity were also negative. Oxygen saturation in this patient was between 72-80%. Transthoracal echocardiogram (TTE) showed double outlet right ventricle (DORV) with transposition of the great arteries (TGA), ventricular septal defect (VSD) with right to left shunt and severe pulmonary stenosis (Figure 2).

Computed Tomography (CT) imaging of the brain showed hypodense lesions with the presence of ring enhancement in the left fronto-



A.



B.

Figure 2.(A) The echocardiogram showed ventricular septal defect (VSD) with right to left shunt; **(B)**The echocardiogram showed double outlet right ventricle (DORV) with transposition of the great arteries (TGA) and severe pulmonary stenosis.

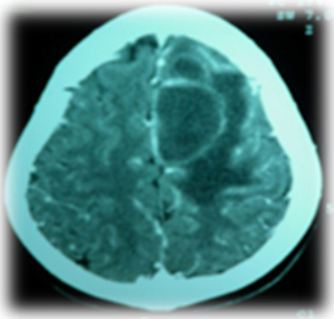


Figure 3. Computed Tomography (CT) showed hypodense lesions with the presence of ring enhancement in the left fronto-parietal lobes surrounded by perifocal oedema that shifted the midline to the right side.

parietal lobes surrounded by perifocal oedema that shifted the midline to the right side (Figure 3).

The Panoramix X-ray (OPG) showed that this patient have a focal infection from her teeth. This patient was treated with parenteral antibiotics, steroid, antiplatelet and anti-convulsant. After being treated for 11 days, her condition was improved clinically.

Discussion

Double outlet right ventricle (DORV), a clinically significant congenital heart defect, occurs in 1-3% of individuals with congenital heart defects.[2] The term "DORV" refers to any cardiac anomaly in which both the aorta and pulmonary trunk originate, predominantly or entirely, from the right ventricle. In this situation, the left ventricle has no direct outlet to either great artery and ejects through an interventricular communication, usually referred to as a VSD (which is almost invariably present) into the right ventricle. Rarely there may be no "VSD" and the LV is then extremely hypoplastic.[3]

Brain abscess is a focal, intracerebral infection that begins as a localized area of infection and develops into a collection of pus surrounded by a well-vascularized capsule.[4] In our case we presented brain abscess in a 20-year-old girl who came to the emergency room with seizures few hours before admission. The most common presentation of brain abscess is headache and vomiting due to raised intracranial pressure. Seizures have been reported in up to 50% of cases.[5] Brain abscess can develop in 5-18% population with cyanotic heart disease (CHD).[6] Tetralogy of Fallot is the most common cardiac anomaly associated with brain abscess. Transposition of great vessels, tricuspid atresia, pulmonary stenosis, and double-outlet right ventricle have also been reported as predisposing factor.[5] In one report among 82 cases of brain abscess, 62 patient (75,2%) had congenital heart disease, resulting in a right to left shunt.

The most common form of congenital heart disease was Fallot's Tetralogy, which occured in 38 cases (61,2%), followed by complete transposition of the great vessels in 6 cases (9,6%), and DORV occured in only 2 cases (3,2%), whilst another reports found that brain

abscess occured only 2,8% in patients with DORV.[7,8]

In cyanotic congenital heart disease, systemic venous return to the right-side of the heart is shunted across the defect into the systemic circulation, resulting in persistent arterial desaturation and cyanosis. Decreased arterial oxygenation can result in compensatory polycythemia.[4]

Patients with cyanotic heart disease could have low-perfusion areas in the brain due to chronic severe hypoxemia and metabolic acidosis as well as increased viscosity of blood due to secondary polycythemia. These low-perfusion areas commonly occur in the junction of gray and white matter, and they are prone to seeding by microorganism that may be present in the bloodstream.[5] Intracardiac right to left shunt by-pass allows direct entry of blood containing bacteria to the cerebral circulation without pulmonary filtration.[6] It has been suggested that two conditions are necessary for brain abscess formation: (1) intermittent bacteremia, and (2) focal encephalomalacia.

Patient with cyanotic congenital heart disease may therefore be susceptible to focal encephalomalacia because of their severe hypoxemia and the increased viscosity associated with polycythemia.[7] In our patient, the oxygen saturation was between 72-80% and have increasing of the haemoglobin and red blood count. Poor dental structure with defective enamel and dental caries contributes to the risk of brain abscess.[9] This can be found in our patient from the Panoramic X-Ray (OPG).

The blood culture and bacterial senstivity were negative in our patient. It was reported in a study that blood cultures in 39 patients with cyanotic brain abscess were mostly sterile (89.8%).[6] In one report among 10 patients with brain abscess in cyanotic congenital heart disease, the blood cultures were negative in all patients.[10] Treatment of a brain abscess involves aspiration of the pus or excision of the abscess, followed by parenteral antibiotic therapy.[5] Legg advocated anticonvulsant therapy for 5 years to all patients with cerebral abscess. Discontinuation of antiepileptic drugs can be considered when patient is seizure free for at least 2 years after surgery and EEG shows no epileptic activity.[11] Steroid administration should be generally avoided unless the patient demonstrates signs of meningitis or disproportionate cytotoxic edema posing a life threatening problem.[6]

Patients with cyanotic heart disease may have increase of blood viscosity and thereby reduce cerebral blood flow. This condition increase the risk of cerebrovascular events, the majority of which are cerebral infarcts related to vessel occlusion rather than intracranial hemorrhage.[12] Our patient was planned to be done aspiration, but she refused. Then she was treated with parenteral antibiotics, steroid, antiplatelet and anti-convulsant. After being treated for 11 days, her condition was improved clinically.

Conclusion

People with cyanotic congenital heart disease are at risk of developing brain abscess. Intracardiac right-to-left shunt bypass, by which blood is not filtered through pulmonary circulation where bacteria are intercepted by phagocytosis, may allow direct entry to cerebral circulation.

References

1. Kagawa M., Takeshita M., Yato S., Kitamura K. 1983. Brain abscess in congenital cyanotic heart disease. J Neurosurg. 58:913-917.
2. Obler D., Juraszek AL., Smoot LB., Natowicz MR. 2008. Double outlet right ventricle: aetiologies and associations. J Med Genet 45:481-497.
3. Wilkinson J. 2003. Double outlet right ventricle. Orphanet Encyclopedia. http://

www.orpha.net/data/patho/GB/uk-DORV.pdf.

4. Lumbiganon P., Chaikitpinyo A. 2007. Antibiotics for brain abscesses in people with cyanotic congenital heart disease (Review). Cochrane Database of Systematic Reviews Issue 3. DOI: 10.1002/14651858.CD004469.pub2.
5. Moorthy RK., Rajshekhar V. 2008. Management of brain abscess: an overview. Neurosurg Focus. 24(6):E3.
6. Muzumdar D., Jhawar S., Goel A. 2011. Brain abscess: An overview. Int J Surg. 9:136-144.
7. Fischbein CA., Rosenthal A., Fischer EG., Nadas AS., Welch K. 1974. Risk factors for brain abscess in patients with congenital heart disease. Am J Cardiol. 34:97-102.
8. Auvichayapat N., Auvichayapat P., Aungwarawong S. 2007. Brain abscess in infants and children: a retrospective study of 107 patients in Northeast Thailand. J Med Assoc Thai. 90:1601-1607.
9. Engle MA. 1976. Cyanotic congenital heart disease. Am J Cardiol. 37:283-308.
10. Minton PR., Vogel JHK., Pryor R., Blount SG. 1967. Brain abscess in cyanotic congenital heart disease. Am J Cardiol. 19:143.
11. Legg NJ., Gupta PC., Scott DF. 1973. Epilepsy following cerebral abscess. A clinical and EEG study of 70 patients. Brain. 96:259-268.
12. Ammash N., Warnes CA. 1996. Cerebrovascular events in adult patients with cyanotic congenital heart disease. J Am Coll Cardiol. 28:768-772.