

## Case Report

# Atypical multiple brain abscesses with congenital single ventricle: a case report

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Received June 7, 2017; Accepted February 16, 2018; Epub March 15, 2018; Published March 30, 2018

**Abstract:** Multiple brain abscesses due to heart disease with congenital single ventricle is relatively rare in clinical practice. Herein, we reported a 28-year-old male patient with multiple brain abscesses who suffered from congenital heart disease (single ventricle). This patient was admitted due to 2-day headache and half-day body twitching without other symptoms. CT suggested intracranial hemorrhage. This patient developed fever after treatment. Then, enhanced magnetic resonance imaging (MRI) indicated spontaneous right frontal lobe hemorrhage, which suggested multiple brain abscess. After antibiotic treatment and brain abscess drainage, left hemiplegia was observed in this patient. The treatment for this patient was extremely risky and difficult due to the congenital heart disease. This patient was finally diagnosed with atypical multiple brain abscesses. Two operations have been conducted on this patient. Left limb muscle strength did not recover to normal level after treatment. This patient was followed up till Dec 2015.

**Keywords:** Congenital heart disease, atypical multiple brain abscess, cerebral hemorrhage

### Introduction

Multiple brain abscesses are a rare and potentially life-threatening condition requiring early diagnosis and early treatment [1]. Congenital heart disease (CHD) is an important predisposing factor for hematogenous brain abscess, especially in children and adolescents with serve congenital heart disease [2-4]. It is reported that single brain abscess in children accounts for 69.49%, and multiple abscesses account for 30.5-42%. Multiple abscess is more common in children with cyanotic congenital heart disease [5, 6]. Herein, we reported a male adult with CHD suffering from multiple brain abscesses who was successfully treated in our hospital.

### Case Report

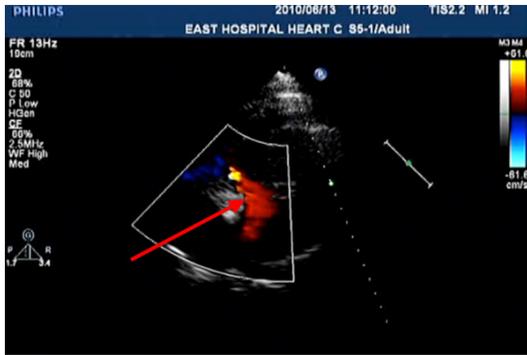
A 28-year-old male patient visited our hospital due to sudden headache for 2 days and half-day body twitching. He was diagnosed with congenital single ventricle XXX years ago and thereafter received long-term oral metoprolol treatment. Cardiac ultrasonography indicated

left ventricular hypertrophy, suggesting CHD (**Figure 1**).

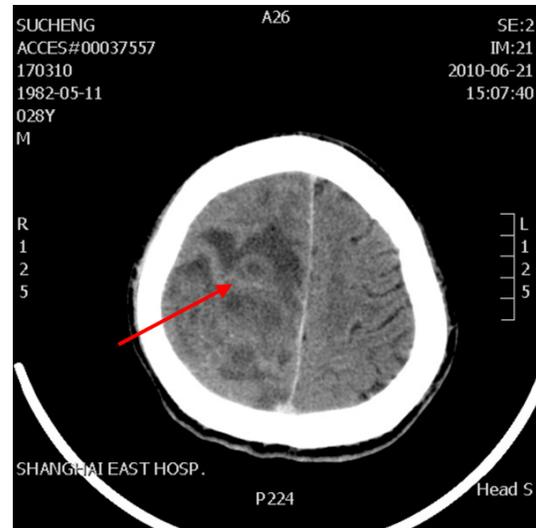
Brain computed tomography (CT) and enhanced magnetic resonance imaging (MRI) revealed that the patient had spontaneous right frontal lobe hemorrhage secondary epilepsy. On the basis of findings from examinations after admission and the medical history, this patient was initially diagnosed with spontaneous right frontal lobe hemorrhage, CHD, and secondary epilepsy. Then, hemostasis, dehydration and other conservative treatments were administered. After 3-day treatment, the patient C-reactive protein (CRP) was 53 mg/L, white blood cell count was  $10.16 \times 10^9/L$ , neutrophils accounted for 75.5%. Cranial CT found the tumor with subarachnoid hemorrhage (**Figure 2**). Then, anti-infective treatment was initiated with ceftazidime and the body temperature reduced to normal level. However, one week later, the headache deteriorated in this patient.

The subsequent cranial CT showed right space-occupying edema with subarachnoid hemorrhage in the parietal lobe (**Figure 3**). The man-

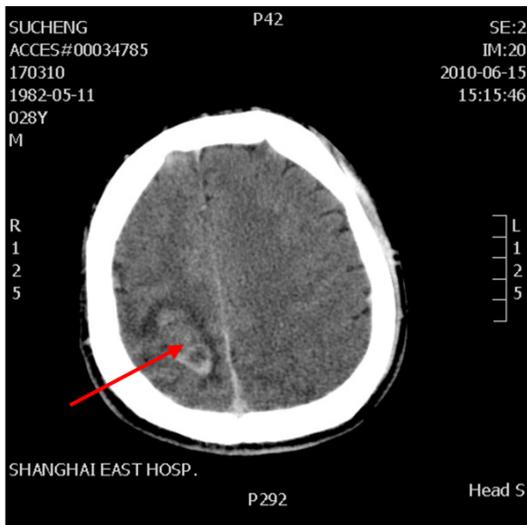
## Multiple brain abscesses with congenital heart disease



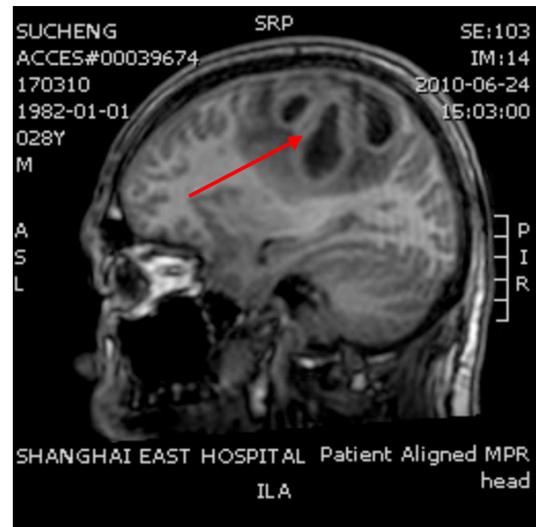
**Figure 1.** Cardiac ultrasonography suggests a single ventricle (arrow).



**Figure 3.** CT scan suggest that the absorbance of hemorrhage, and a cystic lesion at the site of hemorrhage (arrow).



**Figure 2.** CT scan suggests intracranial hemorrhage, edema around the lesion (The high density in the image is the hemorrhage, the surrounding low density shadow is the edema zone) (arrow).



**Figure 4.** Sagittal MRI suggests multiple intracranial cystic lesions (arrow).

nitro treatment was performed to reduce intracranial pressure, but the headache was not significantly improved.

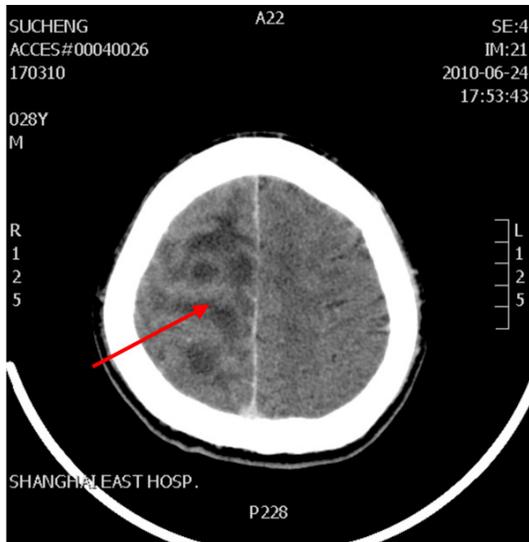
After 12-days serial treatment, the symptom was not significantly improved, but the left limb paralysis was present, and muscle strength was graded 0. This patient was subsequently transferred into intensive care unit (ICU) for further treatment. Enhanced MRI revealed the right frontal lobe lesions and evident brain edema, and then brain abscess or cerebral glioma was considered (**Figure 4**). A second enhanced cranial CT also showed multiple brain abscesses in the right frontal lobe, which confirmed findings from MRI (**Figure 5**).

We speculated that CHD in this patient caused multiple brain abscesses. Meropenem and

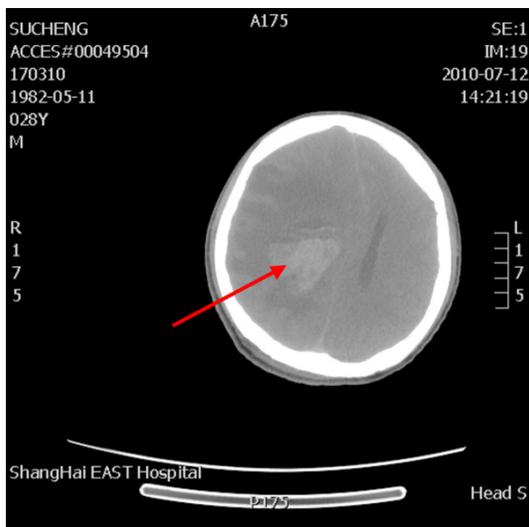
vancomycin hydrochloride was intravenously administered for anti-infective treatment. During the treatment, cranial CT and enhanced MRI revealed there was no change in the brain images.

Considering the risk for irreversible neurological impairment due to brain abscesses, immediate puncture and drainage of brain abscesses were done in this patient. 4 days later, the drainage tubes were removed. Unfortunately, the patient developed vomiting, his consciousness deteriorated significantly, the right pupil was 0.5 cm in diameter, and left pupil was 0.2

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**Figure 5.** CT scan shows similar findings to MRI and suggests multiple intracranial cystic lesions (arrow).



**Figure 6.** After puncture, intracranial hemorrhage becomes recurrent. CT scan suggests increase in intracranial pressure and shift of central line (arrow).

cm in diameter. The emergent cranial CT indicated cerebral hernia and hematoma at the right frontal lobe (Figure 6). The patient received emergent craniotomy. After surgery, cefoperazone sulbactam, vancomycin, and diflucan were used for post-operative anti-infective treatment.

The symptoms were gradually improved after surgery, and the disease condition became stable after 79-day therapy. Examination on discharge showed right limb muscle strength of grade 5, left upper limb muscle strength of

grade 3, left lower extremity muscle strength of grade 4, and negative bilateral babinski sign. This patient was followed up to Dec 2015. CT scan (Figure 7) was performed during follow-ups, and the latest CT showed that although there will be occasional seizures, and disease condition remained stable (Figure 7H).

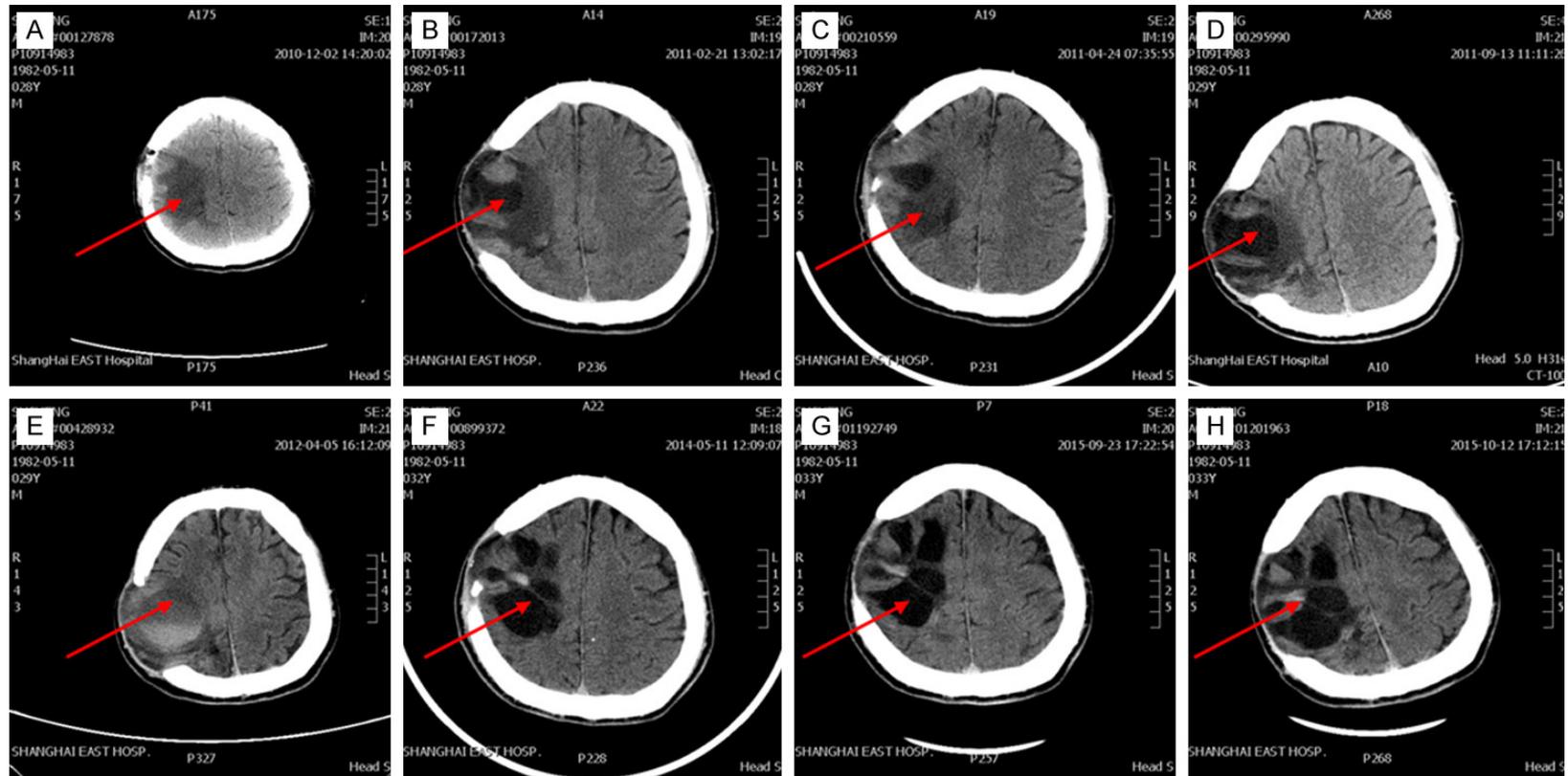
### Discussion

This report is a very rare case. Patients with single ventricle have a very short life cycle. Adult patients with single ventricle are rare. Through this case, we should pay more attention to such patients, avoiding missed diagnosis. Thus, doctors can make correct diagnosis and treatment plan as soon as possible. Intracranial hematoma and brain abscess can be treated by surgery. But early surgery may cause infection diffusion when abscess wall does not form completely. Therefore, it is important to diagnose correctly at the early stage when clinical symptoms and signs are not typical.

Brain abscess is a focal pyogenic infection of the brain parenchyma, and frontal-temporal lobe is the most common site of brain abscess, followed by frontal-parietal, partial, cerebellar, and occipital lobes. The major predisposing factors of brain abscess include contiguous infection, trauma, and hematogenous spread from a distant lesion [7, 8]. Brain abscess is a common complication of CHD [9]. The causes of CHD patients complicated with cerebral abscess may include: cerebral hypoxia, increased blood viscosity due to the compensatory erythrocyte increase. The diagnosis of brain abscess is usually based on clinical and imaging findings. Routine laboratory examination is not specific in the diagnosis of brain abscess [10, 11].

The treatments of brain abscess include pharmacotherapy and surgery. The medical treatment is mainly anti-infective treatment. Surgical treatment employs puncture and drainage of brain abscess. In recent years, some new methods are developed for the treatment of brain abscess, such as MRI assisted stereotactic puncture of multiple brain abscesses, minimally invasive puncture and drainage, cranial microsurgery [12-14]. Precise localization is a prerequisite for the surgical treatment of brain abscess [15, 16]. Invasive intracranial aspergil-

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**Figure 7.** CT scans during follow-ups. A. a CT scan at Dec 2010; B. a CT scan at Feb 2011; C. a CT scan at Apr 2011; D. a CT scan at Sep 2011; E. a CT scan at Apr 2012; F. a CT scan at May 2014; G. a CT scan at Sep 2015; H. 5 years later (Oct 2015), CT scan showed soft foci in the right hemisphere. The general condition is stable and intracranial pressure is normal. Arrows indicate the formation of soft foci.

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losis is associated with high morbidity and mortality [17-19]. This is a significant improvement since 1960s in the treatment of brain abscess due to the development of imaging technique, better neurosurgery and use of antibiotics of higher generation.

### Acknowledgements

We thank Dr. Guangxue Wang for the advice on discussion, Li Xu for nursing assistance. This work was supported in part by the Shanghai Municipal Commission of Health and Family Planning [201440319], and Medical Science Development Fund of Jiangsu University (JLY20160146).

### Disclosure of conflict of interest

None.

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### References

- [1] Brouwer MC, Coutinho JM and van de Beek D. Clinical characteristics and outcome of brain abscess: systematic review and meta-analysis. *Neurology* 2014; 82: 806-813.
- [2] Canpolat M, Ceylan O, Per H, Koc G, Tumturk A, Kumandas S, Patiroglu T, Doganay S, Gumus H, Unal E, Kose M, Gorkem SB, Kurtsoy A and Ozturk MK. Brain abscesses in children: results of 24 children from a reference center in central Anatolia, Turkey. *J Child Neurol* 2015; 30: 458-467.
- [3] Goodkin HP, Harper MB and Pomeroy SL. Intracerebral abscess in children: historical trends at Children's hospital Boston. *Pediatrics* 2004; 113: 1765-1770.
- [4] Kao KL, Wu KG, Chen CJ, Wu JJ, Tang RB, Chang KP and Wong TT. Brain abscesses in children: analysis of 20 cases presenting at a medical center. *J Microbiol Immunol Infect* 2008; 41: 403-407.
- [5] Chinbo M, Addebbous A, Moutachakir M, Rada N, Bouskraoui M, Chabaa L and Soraa N. [Gemella haemolysans brain abscess in a child with complex congenital heart disease]. *Ann Biol Clin (Paris)* 2014; 72: 487-490.
- [6] Ciurea AV, Stoica F, Vasilescu G and Nuteanu L. Neurosurgical management of brain abscesses in children. *Childs Nerv Syst* 1999; 15: 309-317.
- [7] Brook I. Microbiology and treatment of brain abscess. *J Clin Neurosci* 2017; 38: 8-12.
- [8] Roman GC. Defining dementia: clinical criteria for the diagnosis of vascular dementia. *Acta Neurol Scand Suppl* 2002; 178: 6-9.
- [9] Atiq M, Ahmed US, Allana SS and Chishti KN. Brain abscess in children. *Indian J Pediatr* 2006; 73: 401-404.
- [10] Bhand AA. Brain abscess—diagnosis and management. *J Coll Physicians Surg Pak* 2004; 14: 407-410.
- [11] Brouwer MC and van de Beek D. Management of bacterial central nervous system infections. *Handb Clin Neurol* 2017; 140: 349-364.
- [12] Champey J, Pavese P, Bouvaist H, Kastler A, Krainik A and Francois P. Value of brain MRI in infective endocarditis: a narrative literature review. *Eur J Clin Microbiol Infect Dis* 2016; 35: 159-168.
- [13] Fuentes S, Bouillot P, Regis J, Lena G and Choux M. Management of brain stem abscess. *Br J Neurosurg* 2001; 15: 57-62.
- [14] Su TM, Lan CM, Tsai YD, Lee TC, Lu CH and Chang WN. Multiloculated pyogenic brain abscess: experience in 25 patients. *Neurosurgery* 2003; 52: 1075-1079; discussion 1079-1080.
- [15] Nakajima H, Iwai Y, Yamanaka K and Kishi H. Successful treatment of brainstem abscess with stereotactic aspiration. *Surg Neurol* 1999; 52: 445-448.
- [16] Sarma S and Sekhar LN. Brain-stem abscess successfully treated by microsurgical drainage: a case report. *Neurol Res* 2001; 23: 855-861.
- [17] Chen S, Pu JL, Yu J and Zhang JM. Multiple aspergillus cerebellar abscesses in a middle-aged female: case report and literature review. *Int J Med Sci* 2011; 8: 635-639.
- [18] Rosa C, Araujo R, Rodrigues AG, Pinto-de-Sousa MI and Pina-Vaz C. Detection of Aspergillus species in BACTEC blood cultures. *J Med Microbiol* 2011; 60: 1467-1471.
- [19] Shamim MS, Enam SA, Ali R and Anwar S. Craniocerebral aspergillosis: a review of advances in diagnosis and management. *J Pak Med Assoc* 2010; 60: 573-579.