



Case report

Brain abscess: A rare complication in a child with tetralogy of Fallot

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ABSTRACT

Brain abscess is a rarely occurring fatal complication accounting for 5%–18.7% of the population with cyanotic congenital heart diseases. Cyanotic heart diseases are associated with a right-to-left shunt that bypasses pulmonary circulation and results in tissue hypoxia and cyanosis. The most common cyanotic congenital anomaly seen is the tetralogy of Fallot. Herein, we discuss a case of uncorrected tetralogy of Fallot presenting as a parieto-occipital abscess in a 13-year-old boy. The patient was managed conservatively on intravenous antibiotics due to the small size of the abscess and then referred to cardiothoracic surgeons for the repair of the defect. The purpose of reporting this case is to highlight the importance of early detection and correction of cardiac defects to prevent serious complications resulting in childhood morbidity and mortality.

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Introduction

Tetralogy of Fallot (TOF) is a common cyanotic congenital heart disease (CCHD) that comprises of four defects: ventricular septal defect, hypertrophy of right ventricle, overriding of aorta and obstruction of the right ventricular outflow tract [1]. The highest prevalence of congenital heart diseases (CHD) has been found in Asian children, with 9.3 per 1,000 live births [2]. Every year about 40,000 children are born with CHD in Pakistan [3].

Brain Abscess is a relatively unusual but potentially life-threatening infection of brain parenchyma, which can occur in around 5%–18.7% of the population with CHD [4,5]. The main predisposing factors are chronic hypoxia resulting in polycythemia, hyperviscosity and poor host immunity [6]. It frequently presents with headache, fever, seizures, changes in mental status, focal neurological deficits, nausea, and vomiting. In developing countries like Pakistan, delay in surgical repair of TOF puts children at greater risk of developing adverse neurological complications, particularly brain abscess. We report the case of a young boy with untreated TOF to focus on the importance of early diagnosis and prompt management of such patients to decrease the childhood morbidity and mortality.

Case report

A 13-year-old boy was admitted to the Cardiology Department of Civil Hospital, Karachi, Pakistan with complaints of shortness of breath, high-grade fever, headache and two episodes of generalized tonic-clonic fits with altered level of consciousness for the last three days. He was diagnosed with TOF in the late infancy and had not undergone any surgical correction because of severe financial constraints. The patient had a significant history of shortness of breath since childhood, palpitation and generalized weakness. However, there was no history of vomiting, chest pain, any previous medication or surgery.

On general physical examination, clubbing and cyanosis were present. His temperature was 101 °F, pulse rate 106 beats/minute; blood pressure 110/75 mmHg and respiratory rate of 20 breaths/minute and his SpO₂ was varying in between 85%–90% in room air. The patient did not show any peripheral signs of infective endocarditis. On Cardiac Examination, the apex beat was palpable in the left 5th intercostal space within the midclavicular line. Left parasternal heave was also present. Heart sounds were normal and there was a harsh ejection systolic murmur, not radiating grade 3/6 audible at the left sternal border, more on inspiration. He was alert, well-oriented and showed no focal deficit on neurological examination. The remainder of the systemic examinations was within normal limits.

His laboratory investigations revealed a hemoglobin of 13.8 g/dl and hematocrit of 44.7 % suggesting no polycythemia. Total leukocyte count was 13.9 × 10⁹/L and neutrophils were 71 %. Initial blood culture showed growth of Gram-negative *Escherichia coli* b

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acteria. Other serum tests were unremarkable. No apparent cardiomegaly was found on chest x-ray. Echocardiography illustrated large ventricular septum defect with overriding of aorta, hypertrophied right ventricle and thickened pulmonic valve with restricted movement suggesting pulmonary valve stenosis with high transvalvular gradients. There was no evidence of endocarditis on echocardiogram. Contrast-enhanced computed tomography (CT) scan of the brain confirmed the diagnosis of brain abscess by demonstrating a well-defined ring-enhancing hypo-dense lesion in the right parieto-occipital region, measuring $1.4 \times 1.5 \times 1.5$ cm (CCxTVxAP dimensions) with surrounding cerebral edema (Fig. 1A and B).

The patient was managed conservatively on intravenous antibiotics (Vancomycin and Meropenem 500 mg 12 hourly) for two weeks since the size of the abscess was small. He was also given acetaminophen (15 mg/kg/dose per oral six hourly) and anticonvulsant (Sodium Valproate 500 mg per oral 12 hourly). The patient showed marked improvement in his symptoms within three days of therapy. A repeated contrast CT brain was done after



Fig. 2. Contrast CT brain two weeks after the antibiotics therapy demonstrates marked reduction in the size of the abscess and surrounding edema (red arrow) (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.).

two weeks and it depicted regression in the size of brain abscess and perilesional edema (Fig. 2). Blood culture sample was also obtained after two weeks and revealed no growth. He was discharged on oral antibiotics (Cefotaxime and Metronidazole) for a further two weeks. On follow up after one month, the patient was fully recovered and no neurological deficits were observed. He was then referred to cardiac surgery for the correction of TOF.

Discussion

Tetralogy of Fallot (TOF) is a congenital heart disease that combines four major developmental defects of the heart. Such defects with an underlying right-to-left sided shunt present with cyanosis early in childhood and thus, alternatively named as cyanotic heart diseases. Complications of untreated TOF such as improper growth and development of the child, delayed milestones, secondary polycythemia, and a more severe infective endocarditis are characteristic, however, a rare albeit serious complication, brain abscess has also been reported as a presenting complaint [7].

Cerebral abscess, generally thought of as an infectious disease, is expected to be more common in developing countries where infections are endemic, sanitation is poor, and basic medical facilities are unavailable that result in late detection of disease. Our patient belonged to the rural side of Pakistan. There are many factors responsible for the occurrence of brain abscess in CCHD patients. Bypassing the pulmonary circulation, the blood in these patients is not filtered by normal alveolar phagocytes. This increases the probability of direct entrance of pathologic microorganisms into the circulation of brain. This together with the fact that the brain might be hypo-perfused due to severe hypoxemia and metabolic acidosis resulting from secondary polycythemia, allows the pathogens to seed such under-perfused regions [4,7]. Characteristically, hematogenous spread of this kind from a distant source is attributed to multiple cerebral abscesses.

Brain abscess is two to three times more common in men than women. Even though rare in occurrence, it is potentially fatal and is associated with high morbidity especially affecting people in their forties [8]. Generally, cerebral abscesses in CCHD patients are

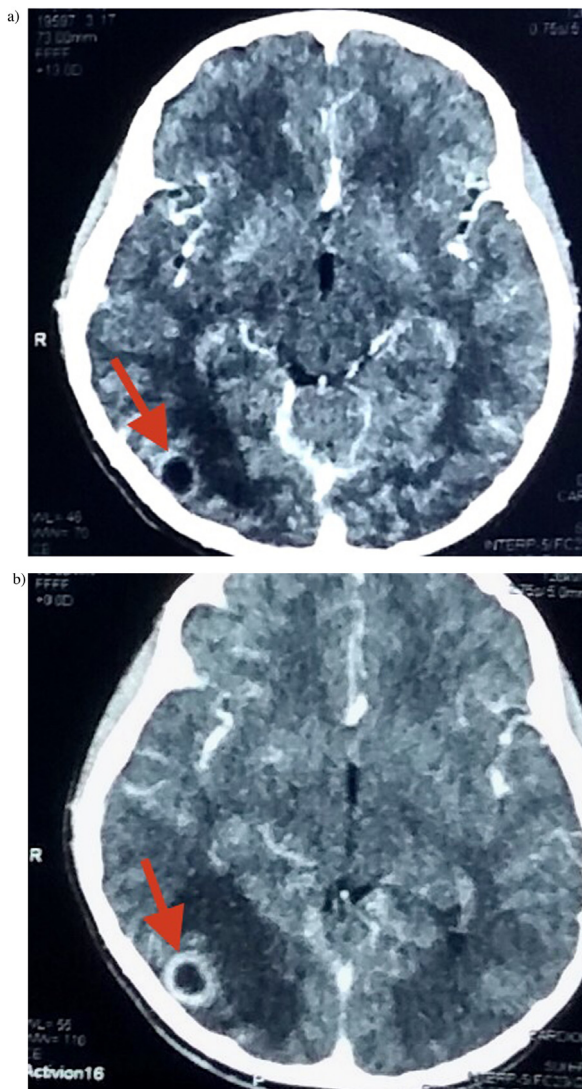


Fig. 1. (a) Initial contrast-enhanced CT brain showing a $1.4 \times 1.5 \times 1.5$ cm ring enhancing lesion in the right parieto-occipital region with perilesional edema (red). (b) Initial contrast-enhanced CT brain showing a $1.4 \times 1.5 \times 1.5$ cm ring enhancing lesion in the right parieto-occipital region with perilesional edema (red) (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.).

attributed to infection by non-hemolytic streptococci [7], *Streptococcus milleri* being the most common cause in particular. Blood culture report in our patient, however, suggested E.coli as the causative agent. E.coli, gram-negative bacteria, is a rare cause of brain abscess since only two patients have been affected by an E. coli brain abscess in the past 20 years [9].

The delay in the diagnosis and consecutive management of brain abscess in CCHD patients leaves the underlying condition unaddressed for a while sufficient to complicate the disease process. Mortality in untreated patients ranges from 27.5%–71% [4]. Larger or deep-seated abscesses should be aspirated immediately and repeatedly. Following aspiration, the patient should be started on appropriate antibiotic therapy specifically targeting the organism discovered in culture. Empirical medical therapy is an exclusive treatment for cases where the size of abscess is less than 2 cm in diameter, the patient is neurologically stable and being monitored by repeated CT scans [4,10]. Our patient was treated with antibiotics only because his abscess size was 1.4 × 1.5 × 1.5 cm with no neurological deficit. Other forms of intervention such as craniotomy and excision have poor outcomes in terms of mortality rate which can be as high as 71 % and are thus reserved for progressively worsening abscesses where conventional approach remains ineffective [10].

Conclusion

Considering the fact that brain abscess complicates uncorrected cyanotic congenital heart diseases, physicians must hold a high index of suspicion for potentially dealing with a case of CCHD-related brain abscess to avoid unnecessary delay in diagnosis and management. This is especially important since brain abscess itself continues to be a challenge to the medical world despite the introduction of updated diagnostic radiological procedures, newer and more effective antibiotic regimen, and better neurosurgical techniques. Most importantly, parents of children diagnosed with CCHD should be counseled on how to recognize serious signs and symptoms of brain abscess and when to seek urgent medical attention.

Authorship conformation form

All authors have participated in (a) conception and design, or analysis and interpretation of the data; (b) drafting the article or revising it critically for important intellectual content; and (c) approval of the final version.

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References

- [1] Bailliard F, Anderson RH. Tetralogy of fallot. *Orphanet J Rare Dis* 2009;4:2, doi: <http://dx.doi.org/10.1186/1750-1172-4-2>.
- [2] van der Linde D, Konings EE, Slager MA, Witsenburg M, Helbing WA, Takkenberg JJ, et al. Birth prevalence of congenital heart disease worldwide: a systematic review and meta-analysis. *J Am Coll Cardiol* 2011;58:2241–7, doi: <http://dx.doi.org/10.1016/j.jacc.2011.08.025>.
- [3] Hussain S, Sabir MU, Afzal M, Asghar I. Incidence of congenital heart disease among neonates in a neonatal unit of a tertiary care hospital. *J Pak Med Assoc* 2014;64:175–8.
- [4] Pandian JD, Moosa NV, Cherian PJ, Radhakrishnan K. Brainstem abscess complicating tetralogy of Fallot successfully treated with antibiotics alone. *Neurol India* 2000;48:272–5.
- [5] Sethi S, Kapil S. Scalp block for brain abscess drainage in a patient with uncorrected tetralogy of Fallot. *World J Clin Cases* 2014;2:934–7, doi: <http://dx.doi.org/10.12998/wjcc.v2.i12.934>.
- [6] Atiq M, Ahmed US, Allana SS, Chishti KN. Brain abscess in children. *Indian J Pediatr* 2006;73:401–4, doi: <http://dx.doi.org/10.1007/BF02758560>.
- [7] Begum NNF, Sarker FR, Begum M, Yesmin T, Hossain B, Ferdous J, et al. Management of a critical case of double outlet right ventricle (DORV) and cerebral abscess by multiple interventions. *JAFMC Bangladesh* 2016;11:81–4, doi: <http://dx.doi.org/10.3329/jafmc.v11i1.30679>.
- [8] Alvis Miranda H, Castellar-Leones SM, Elzain MA, Moscote-Salazar LR. Brain abscess: current management. *J Neurosci Rural Pract* 2013;4:S67–81, doi: <http://dx.doi.org/10.4103/0976-3147.116472>.
- [9] Akuzawa N, Osawa T, Totsuka M, Hatori T, Imai K, Kitahara Y, et al. Secondary brain abscess following simple renal cyst infection: a case report. *BMC Neurol* 2014;14:130, doi: <http://dx.doi.org/10.1186/1471-2377-14-130>.
- [10] Ashraf M, Ahmed S, Ahmad S, Hussain M. Burr hole aspiration of brain abscess in children with cyanotic heart disease. *J Coll Physicians Surg Pak* 2017;27:483–5.