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# Cerebral Abscess in a Septic Toddler with Left to Right Shunt

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**Keywords:** Brain abscess; Sinus venosus defect; Urinary tract infection; Citrobacter koseri

**Abbreviations:** Qp: Pulmonary blood flow; Qs: Systemic blood flow; MRI: Magnetic Resonance Imaging; CSF: Cerebrospinal Fluid; TTE: Transthoracic Echocardiography; TEE: Transesophageal Echocardiography.

### Abstract

We describe a case of a 17-month old diagnosed with multiple small left frontal cerebral abscesses with moderate left lateral ventriculomegaly. His initial presentation was high grade fever, vomiting and focal neurological deficits in the setting of a urinary tract infection. A Transthoracic Echocardiography (TTE) with agitated saline contrast study revealed superior sinus venosus defect with Partial Anomalous Pulmonary Venous Return (PAPVR) of the right upper pulmonary vein segment draining to the superior vena cava and a Patent Foramen Ovale (PFO) with right to left shunting with Valsalva maneuver. His initial CSF examination had shown profound leukocytosis which responded excellently to intravenous antibiotics. Follow up brain MRI showed complete resolution of the abscess and cerebral ventriculitis. Following complete recovery, he underwent an uneventful sinus venosus defect and PAPVR repair via two patch technique along with closure of the PFO to prevent further complications.



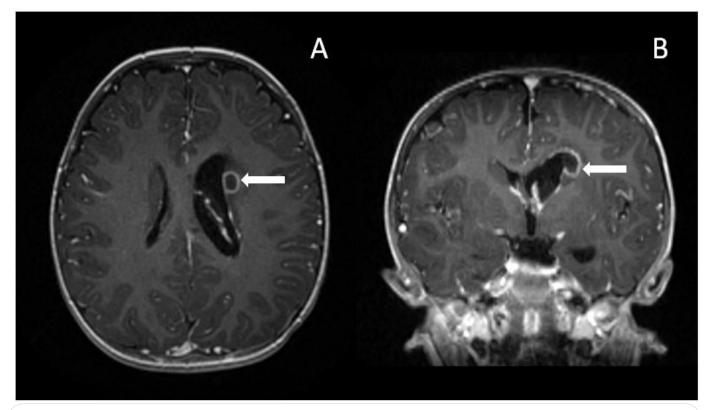
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### Introduction

Cerebral abscess is a rare and potentially life threatening condition in pediatric population. Children with congenital heart disease especially the cyanotic, prosthetic heart valves and devices or immunocompromised state are particularly at increased risk [1]. Intermittent right to left shunting in acyanotic heart diseases like patent foramen ovale or atrial septal defects as a cause of brain abscess have been reported [2,3,4]. We describe a rare case of a toddler who presented with neurological deficit secondary to brain abscess and found to have superior sinus venosus defect with partial anomalous pulmonary venous return.

### **Case report**

A 17 month old fully vaccinated boy with unremarkable past medical history who initially presented with high grade fever and vomiting. Review of systems was otherwise negative. Preliminary work up was positive for urinary tract infection secondary to Citrobacter Koseri for which he was started on oral antibiotics. The fever persisted for 3 days, he started to develop worsening lethargy and he began to demonstrate dysconjugate gaze. Brain MRI showed multiple cerebral abscesses in the frontal lobe with intraventricular extension and left lateral ventriculitis, there was secondary moderate left lateral ventriculomegaly with associated interstitial edema (Figure 1 & 2). His complete blood cell count showed a WBC count of 11700/µL (Neutrophils 53%, lymphocyte 37%, monocytes 9%), hemoglobin of 11.2 gm/ dl and platelet count of 523000/µL. C-reactive protein was normal (0.7 mg/dL). His initial CSF exam showed profound leukocytosis (Appearance: Bloody, WBC count – 13,405/µL, neutrophils 76%, Lymphocytes 18%, RBC count 38,000, total protein 298 mg/dL), his CSF culture was negative for any aerobic or anaerobic bacterial growth. Suspicion for a septic embolism prompted a focused transthoracic echocardiography (TTE) with agitated saline contrast study performed in the surgical suite which right to left shunting at the atrial level with ventilator Valsalva manouvre. Detailed TTE, the following day revealed a superior sinus venosus defect with partial anomalous pulmonary venous return (PAPVR) of the right upper pulmonary vein segment draining to the right sided superior vena cava and a patent foramen ovale (PFO) with left to right shunting (Figure 3). Cardiac magnetic resonance confirmed the diagnosis and showed increased pulmonary blood flow (Qp - 4.6 L/min vs Qs of 2.18 L/min) with a pulmonary to systemic flow ratio (Qp:Qs) of 2:1 and severe right ventricular dilation. He underwent an emergent neuroendoscopic exploration of the left cerebral ventricular system with washout and placement of an external ventricular drain. After completing 6 weeks of IV antibiotics, follow up brain MRI showed complete resolution of the abscess and ventriculitis (Figure 4). Following complete recovery, he underwent an uneventful sinus venosus defect and PAPVR repair via two patch technique along with closure of the PFO to prevent further complications. During his follow up visit, he showed complete recovery of his neurological deficits.



**Figure 1:** Post-enhanced T1 axial **(A)** and coronal **(B)** images at the time of presentation. There is sub-ependymal ring-enhancing lesion with hypointense center corresponding to a small abscess (arrow), with surrounding vasogenic edema. The left lateral ventricle is moderately enlarged. Notice the presence of ependymal enhancement (arrow).

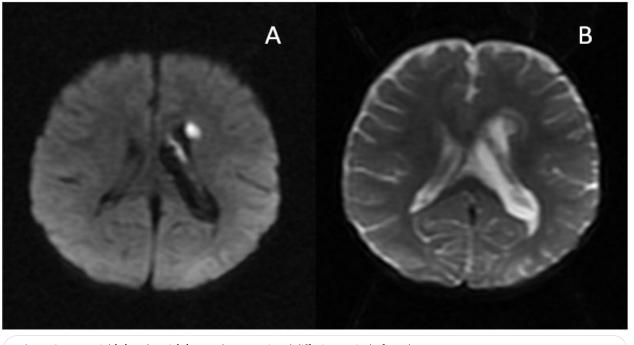
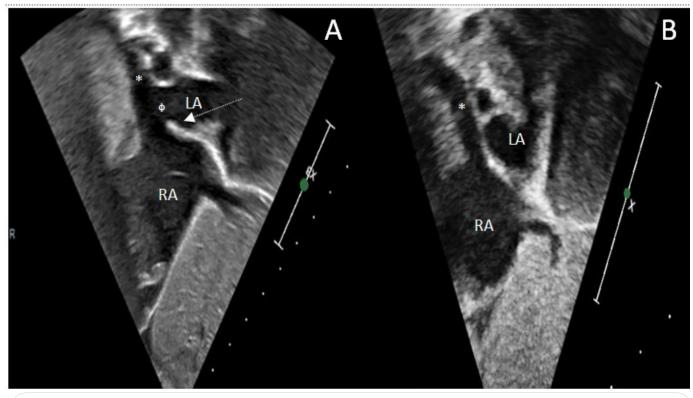
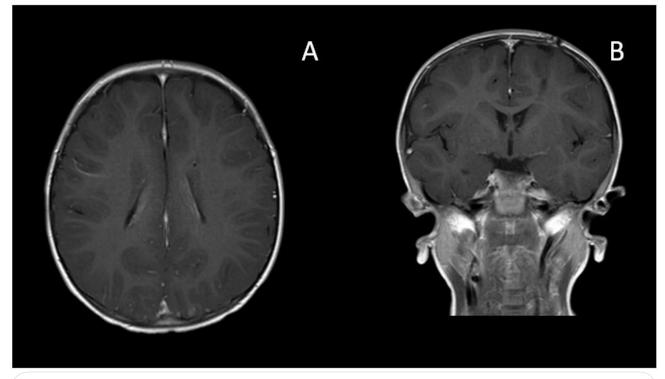


Figure 2: DWI axial (A) and ADC (B) map show restricted diffusion typical of an abscess.



**Figure 3:** (A) Transthoracic echocardiogram with subcostal short axis view showing superior sinus venosus defect ( $_{\phi}$ ), appreciate superior vena cava (\*) that appears to be overriding the interatrial septum (arrow) in our patient. (B) Normal heart with no sinus venosus defect and SVC connected to the right atrium (RA).



**Figure 4:** Follow up MRI 4 months after treatment. Post enhanced axial **(A)** and coronal **(B)** images show complete resolution of the abscess and edema. The ventricles are normal in size.

### Discussion

We encountered an unusual presentation of a left to right cardiac shunt leading to brain abscess possibly as a complication of urinary tract infection.

Brain abscess is a focal pyogenic infection of the brain's parenchyma. Pediatric brain abscess is a rare but serious infection, often involving patients with specific risk factors and burdened by a high risk of morbidity and mortality. The annual incidence of bacterial brain abscesses in the general population is as high as 0.3 to 1.3 per 100,000 populations; recent case series of pediatric brain abscesses have estimated the annual incidence to be approximately 0.5 per 100,000 children [5]. Due to its low incidence, it is challenging to accurately determine if the annual incidence is increasing as a result of highly developed diagnostic cranial imaging such as magnetic resonance imaging or decreasing as a result of better vaccination availability, antibiotics and timely interventions for congenital heart diseases.

The most frequent intracranial locations are frontal-temporal, frontal-parietal, partial, cerebellar, and occipital lobes. The major predisposing factors include a hematogenous spread from a distant focus or contiguous spread of local infection. The microbial etiology depends on the site of the primary infection; the patient's age, underlying condition, and immune status; and the geographic location. The organisms most commonly isolated are anaerobic bacteria, aerobic and microaerophilic streptococci, Enterobacteriaceae, and Staphylococcus aureus. Specimens obtained during surgery or stereotactic Computerized Axial Tomography (CT) guided aspiration should be sent for aerobic, anaerobic, mycobacterial and fungal culture and, when indicated, for protozoa. In neonates, most infections associated with brain abscesses are caused by Gram-negative bacteria, especially by Citrobacter Koseri and Proteus Mirabilis. However, other pathogens should be taken into consideration, such as Gram-positive bacteria, fungi, or mixed infections.

Citrobacter Koseri is a physiological bacteria of human digestive tract, they may also be found in contaminated food, water and sludge. Although Citrobacter Koseri does not usually cause infections, it has a very strong affinity for the brain tissue [6]. In our patient, urine cultures were positive for Citrobacter Koseri. Interestingly, the CSF culture remained negative, which may be explained by the bacterial clot hypothesis. This hypothesis states that bacterial clots localize in the small brain vessels and consequently cause brain abscesses. This can be found especially in infections caused by Citrobacter Koseri due to its strong affinity for brain tissue. Consequently, inflammation within the CNS develops without seeding bacteria into the CSF, which explains sterile cultures.

Before abscess encapsulation and localization, antimicrobial therapy, accompanied by measures to control increasing intracranial pressure, are essential. Once an abscess has formed, surgical excision or drainage combined with prolonged antibiotics (usually 4-8 weeks) remains the treatment of choice.

Pathophysiology of intra-cardiac right to left shunts as a cause of cerebral abscess has been well described in the literature. Gautier et al [2] Have described a case of patent foramen ovale with paradoxical embolism presenting as cryptogenic brain abscess. The literature shows that asymptomatic left to right intracardiac shunts are an unusual cause of cerebral abscess in pediatric population [7]. However, Abdullah et al reported a case of sinus venosus defect causing brain abscess in a 12-year-old child. As opposed to paradoxical embolism in sinus venosus defect, transient streaming of infected systemic venous blood into the left atrium secondary to superior vena cava overriding the atrial septum is a plausible hypothesis. It is important to be aware that crying may elicit the Valsalva reflex and there could be more than expected right to left shunting in a patient with atrial level communication. Our patient with similar cardiac lesion presented at a much younger age with urosepsis, focal neurological findings and evidence of right heart volume overload. Degree of compromise in neurologic conditions on initial evaluation, in particular alteration in mental status, is consistently found to be predictive of ultimate prognosis [8-14]. He required an emergent ventricular drain placement and repair of his cardiac lesion following completion of his antibiotic treatment to prevent progressive right heart dilation and dysfunction. Transesophageal echocardiography with an agitated saline contrast study is recommended in patient where transthoracic imaging does not provide the diagnosis, however in our patient TTE was the diagnostic tool used followed by a cardiac MRI to quantify the pulmonary blood flow.

There are no pediatric specific cardiac imaging guidelines in a patient with idiopathic brain abscess, clinicians currently rely on expert opinion. We recommend initial imaging in the form of transthoracic echocardiography with agitated saline contrast study and transesophageal approach if TTE is suboptimal.

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