ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Pediatric Neurosurgery

Editor
Frank Van Calenbergh, MD
University Hospitals; Leuven, Belgium

SNI
Open Access

Case Report

Multiple Abscesses in the Frontal, Temporal and Brainstem regions in a 4.5-year-Old Girl- An Illustrative Case Report

Retaj Mohammad, Darach Crimmins

Department of Neurosurgery, Children's Health Ireland at Temple St, Dublin, Ireland.

E-mail: *Retaj Mohammad - r.mohammad0114@gmail.com; Darach Crimmins - darach.crimmins@cuh.ie

***Corresponding author:**
Retaj Mohammad,
Department of Neurosurgery,
Children's Health Ireland at
Temple St, Dublin, Ireland.

r.mohammad0114@gmail.com

Received : 21 March 2023
Accepted : 02 June 2023
Published : 16 June 2023

DOI
10.25259/SNI_249_2023

Quick Response Code:

ABSTRACT

Background: Brainstem located abscesses are rare in the pediatric population. Diagnosis of brain abscess can be challenging as patients may present with nonspecific symptoms and the classical triad of headache, fever, and focal neurological deficit is not always present. Treatment can be conservative or a combination of surgical intervention with antimicrobial therapy.

Case Description: We present the first case of a 4.5-year-old girl with acute lymphoblastic leukemia that developed infective endocarditis (IE) and subsequently developed 3 suppurative collections (frontal, temporal, and brainstem). The patient had negative cerebrospinal, blood, and pus culture growth and subsequently underwent burr-hole drainage of the frontal and temporal abscesses with a 6-week course of intravenous antibiotic therapy with an uneventful postoperative course. At 1 year, the patient is left with minor right lower limb hemiplegia and no cognitive sequelae.

Conclusion: The decision to surgically intervene for brainstem abscesses is dependent on surgeon and patient factors including the presence of multiple collections, midline shift, the aim of source identification in sterile cultures, and the patient's neurological condition. Patients with hematological malignancies should be monitored closely for IE which is a risk factor for hematogenous spread of brainstem located abscesses.

Keywords: Acute lymphoblastic leukemia, Brainstem abscess, Burr hole, Pediatric

INTRODUCTION

Brain abscess (BA) is a focal suppurative intracranial infection characterized by the formation of a well-define capsule involving the brain parenchyma.[18] BA is a relatively rare pathology with an estimated incidence of 0.76/100,000 person-year/annum.[4] Moreover, only 25% of cases are observed in the pediatric population.[32] Despite the rare occurrence, BA is associated with high morbidity and mortality. Lesions located in the brainstem region represent 1% of pediatric cases.[1]

Diagnosis of BA can be challenging due to the wide range of symptoms children which may present with. The classical triad of headache, fever, and a focal neurological deficit (FND) is only observed in up to 28% of cases.[25] In addition, children and specifically the infant population may find it difficult to describe a headache. As clinical presentation is vague, diagnosis is confirmed on neuroimaging through a computed tomography (CT) or magnetic resonance imaging (MRI) scan with contrast demonstrating the characteristic ring-enhancing lesion.[6]

The spread of infection may be contiguous or hematogenous. Common predisposing factors in childhood include congenital heart disease and immunosuppression.[34] Sinus infections and dental sources are also very common in children.[1]

The optimal management remains a controversial topic widely debated by physicians. Treatment can be conservative with antibiotic therapy or may involve surgical intervention.[18] There are no official treatment guidelines for the management of BA. A combination approach is often recommended, and medical-only treatment is reserved for specific cases. Location is a possible indicator for conservative treatment in which the frontal lobe has been the most frequent nonsurgically treated region.[25] Brainstem lesions are more difficult to treat to the anatomic proximity to significant regions in the brain. Successful reports of conservatively treated pediatric brainstem abscesses have been published.[1,10,23,35] However, with new surgical advancements, more surgeons are opting for a surgical excision with a craniotomy or surgical aspiration with a burr-hole technique. The optimal surgical intervention for brainstem located abscesses is not clear.

Prognosis and outcomes of BA have improved with new advancements in neuroimaging modalities, broadening of the vaccination programs in children, novel antibiotics with improved bacterial cover and blood brain barrier (BBB) penetration, and new surgical techniques.[18] However, some patients are still left with neurological and cognitive impairment. The outcome of brainstem abscesses specifically remains unclear when compared to infections in other locations.

To the best of our knowledge, there have been only 27 published cases of pediatric brainstem abscesses.[1,2,8,10-17,19,22-24,26-29,33,35-39,41] We report the 28th case of a pediatric patient with acute lymphoblastic leukemia (ALL) that developed multiple suppurative intracranial collections including a pontine abscess that was treated with a surgical intervention and a course of antibiotics. To the best of our knowledge, this is the 1st reported case of pediatric brainstem abscess associated with ALL.

CASE PRESENTATION

A 4.5-year-old girl presented to the emergency department with a 2-day history of progressive fatigue, fever, and tachycardia. The patient was diagnosed with ALL 4 months prior and was receiving chemotherapy and following up with the oncology team. The patient's Glasgow coma scale (GCS) on admission was 15/15 and systems examination demonstrated tachycardia but otherwise unremarkable.

Investigations

Initial blood work showed leukopenia with a white cell count (WCC) of 1.4 × 10^9 L (4–13.5) and an elevated C-reactive protein (CRP) of 25 mg/L (<5). All other parameters were unremarkable.

Due to the patient's cardiac symptoms, an echocardiogram was requested. The echocardiogram revealed vegetations and a diagnosis of infective endocarditis (IE) was made. The patient underwent CT [Figure 1] scanning which demonstrated three ring-enhanced lesions in the frontal lobe, temporal lobe, and brainstem (pons) measuring up to 0.7 cm and surrounding vasogenic edema with a right to left midline shift.

Laboratory procedures

A lumbar puncture was performed but both the cerebrospinal fluid (CSF) cultures and blood cultures were negative. After 2 days of admission to the neurosurgical department, the decision for surgical intervention was made due to sterile cultures and difficulty to obtain source control as well as worsening neurological symptoms where the patient started to develop right lower limb weakness. The patient subsequently underwent a burr-hole drainage of the frontal and temporal collections using frameless navigation. This was done using an open right frontal technique with two burr holes and there was no intraoperative imaging during the procedure. Pus culture was aspirated but was also sterile with no growth. The patient was commenced on Piperacillin-Tazobactam and Vancomycin IV with Amphotericin B and Voriconazole for fungal cover. The patient underwent a total of 6-week duration IV-only antibiotic therapy. The patient also received prednisolone steroid therapy. Midazolam was prescribed as required for seizure prophylaxis. Following the burr-hole aspiration, the patient had an uneventful postoperative course with no surgical complications or re-operation. Postoperative CT scan showed no evidence of

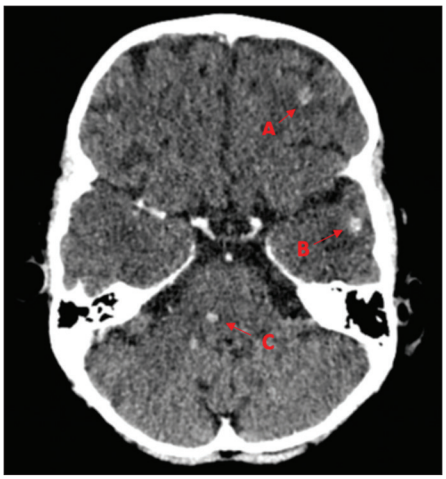

Figure 1: Axial preoperative computed tomography scan with contrast demonstrating three ring-enhanced lesions located in the frontal lobe (A), temporal lobe (B), and the pons (C).

hemorrhage and resolution of the midline shift and vasogenic edema. The Glasgow outcome scale (GOS) at discharge was 3/5 due to moderate right lower limb hemiplegia.

At the 1-year follow-up, the patient still suffered from mobility impairment requiring a wheelchair for transportation for long distances. However, GOS at the 1-year follow-up was 4/5 due to minor right lower limb hemiplegia which was an improvement from the GOS at discharge (3/5) as the patient had some improved motor function and required minor assistance with daily activities. The patient was not academically delayed and did not have intellectual disability. For the European Quality 5D Youth quality of life questionnaire, the patient reported "a lot of problems" with mobility and "some problems" with pain and discomfort occasionally requiring painkillers. The European Quality Visual Analog Scale for this patient was 70%. Otherwise, the patient reported no problems with self-care, usual activities, and personality and mood disturbance.

DISCUSSION

Clinical presentation of BRs in the pediatric population is often nonspecific and vague. The classical triad of fever, headache, and FND has been observed in up to 28% of cases and, therefore, is an unreliable indicator.[25] Our patient presented with a 2-day history of fatigue, fever, and tachycardia but did not display the classical triad which is reported in the literature. In a recent review of pediatric brainstem abscesses including 24 patients, the classical triad is observed in 20.8% of cases.[1] The most common presenting signs in the brainstem cohort were a neurological deficit with cranial nerve palsies being the most prevalent deficit observed. However, our patient did not display any neurological signs on presentation and was fully conscious on admission with a GCS of 15/15. The patient presented with tachycardia but an otherwise normal cardiovascular exam. A diagnosis of IE was made after the visualization of new vegetations on the aortic valve. This may be due to the patient's central lines and catheters as she was receiving chemotherapy for ALL. This demonstrates that cardiac manifestation of ALL is not uncommon and should be always considered and evaluated in patients with nonspecific symptoms such as fever and fatigue. Tabaei *et al* report a similar case of a 6-year-old boy with ALL that developed IE that eventually required surgical resection.[30] The most common location of brainstem infections occur in the pons region followed by the midbrain and medulla oblongata, respectively.

Multiple BAs vary in the literature and have been reported in up to 50% of cases in children.[9] Our patient had three collections (two frontal and one brainstem) diagnosed on MRI. This may be due to the history of ALL and immunocompromised status. A compromised immune status and a diagnosis of IE have been associated with the development of multiple abscesses in children.[3] In addition, these patients are more likely to develop opportunistic infections and have a higher complication rate.[21]

Laboratory parameters such as an elevated WCC and inflammatory markers including CRP or erythrocyte sedimentation rate may indicate an infectious etiology. However, they are unreliable in the diagnosis of BA as they may be normal in up to 40% of cases.[7] Our patient presented with a high CRP of 25 mg/L but low WCC of 1.4×10^9/L. The leukopenia is explained by the patient's cytotoxic medications for the treatment of ALL.

Initial microbiological cultures for CSF and blood cultures were negative for any growth. Yogev and Bar-Meir estimate a positive growth rate of 10% of blood cultures in the setting of BA.[40] Blood culture and CSF culture growth rate for brainstem located abscesses in children was lower than general pediatric BA series.[1] Intraoperative pus was also sent for culture which was negative leading to a therapeutic dilemma. Including our case, 11/20 (55%) reported cases had positive pus culture growth.[1,39] There are no official treatment guidelines for suppurative intracranial infections and the empiric antimicrobial regimen is often guided by the likely predisposing source of the infection.[6]

The patient was commenced on Piperacillin-Tazobactam and Vancomycin IV and Amphotericin B and Voriconazole for fungal cover. The patient underwent a total of 6-week duration IV antibiotic therapy. Broad-spectrum antimicrobial therapy should be further narrowed after organism identification and sensitivity reporting. It is challenging to determine the appropriate empiric regimen in patients with ALL when cultures are sterile. Due to the likelihood of fungal infections in this cohort, antifungal agents should always be included. The patient received steroid therapy for cerebral edema and a midline shift observed on MRI scans. There are no clear indications for steroid therapy in BA and it should be used cautiously due to the potential harmful side effects, interference with pathophysiology of abscess formation and impact on antimicrobial BBB penetrance.[18,21]

Surgical indication for brainstem abscesses is debatable with various opinions expressed by different surgeons and institutions. Successful nonsurgical treatment of pediatric brainstem abscesses has been reported.[1,10,23,35] These cases were reported in older children but not in the infant population. The decision is highly dependent on surgeon and patient factors including whether the lesions are solitary or multiple as highlighted by our case. In this case, the patient had two other larger collections in the frontal and temporal regions and the decision was made to aspirate the larger collections to minimize cerebral edema and midline shift. This was done 2 days after the culture results were sterile as the patient also started developing new neurological deficits such as limb weakness. Therefore, a combination of source control to identify and guide antibiotic treatment with symptom control to prevent further neurological deterioration

were the main driving factors for surgical intervention. The patient continues for a total of 6 weeks of intravenous antibiotics and followed up postoperatively clinically and radiologically for persistence of infection and recurrence of the collections.

The aim of source identification was not achieved as pus culture was sterile as well. The patient continued on the same empiric regimen for a total of 6 weeks. However, the GOS outcome was improved at the 1 year follow-up compared to discharge, 4/5 and 3/5, respectively. At the discharge mark, the patient had significant mobility impairment and reduced power in the right lower limb whereas after 1 year, the patient was semi-independent and had minor limb weakness.

There is no difference in outcome in the published literature when comparing different techniques such as burr-hole drainage, endoscopic drainage, or craniotomy. Minimally invasive techniques are preferred as they are safer than an open surgical approach.[10] On the other hand, craniotomy open surgery has been associated with less recurrence rates, length of intrahospital stay, and an overall reduction of antibiotic therapy duration.[20,31]

The prognosis of BA has improved over the past five decades with the advancements of neuroimaging and microbiological culture techniques. Before the CT era, the mortality of children with BA was remarkably high at 40–60% and since has dropped to below 10%.[6,18] Specifically, a lower mortality rate of 0–3% has been reported with the stereotactic aspiration approach.[5,21] In this review, 10.7% (3/28) of children with brainstem abscesses have died. Neurological and cognitive outcome have also been explored in this cohort and seem to be improving. One-third of children with brainstem abscesses reach full neurological recovery and up to 70% are left with mild neurological deficits.[1] In our case, mobility impairment was the most impacted determinant of quality of life at 1 year follow-up. However, research on long-term outcomes in this population remains scarce.

CONCLUSION

We present the first case to our knowledge, of a 4.5-year-old girl with ALL and multiple collections including a brainstem abscess. The decision to surgically intervene for brainstem abscesses is dependent on surgeon and patient factors including the presence of multiple collections, midline shift, the aim of source identification in sterile cultures, and the patient's neurological condition. In our case, the frontal and temporal collections were aspirated instead of the smaller brainstem lesion in an effort to obtain source control and improve the neurological deficits. Despite sterile microbiological cultures, this patient recovered well with minor right hemiplegia at 1 year follow-up. There is no superior surgical technique for brainstem located abscesses. Further studies should look at long-term outcomes and empiric regimens in the immunocompromised patient demographic.

Ethical approval

Informed consent was obtained from the patients' guardian. Ethical approval and data protection agreement were obtained from the Research Ethics Committee of Temple St Children's University Hospital, Dublin, Ireland (REC-055–21).

Authors' contributions

Retaj Mohammad was involved in study design, ethical approval and consent seeking, data collection, and analysis and the writing of the manuscript. Darach Crimmins was involved in study design and overall supervision of manuscript.

Acknowledgments

The authors are grateful to the patient and their family for providing follow-up information and allowing this case to be shared.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Antkowiak Ł, Putz M, Mandera M. Clinical features, microbiology, and management of pediatric brainstem abscess. Childs Nerv Syst 2020;36:2919-26.
2. Arzoglou V, D'Angelo L, Koutzoglou M, Di Rocco C. Abscess of the medulla oblongata in a toddler: Case report and technical considerations based on magnetic resonance imaging tractography. Neurosurgery 2011;69:E483-6.
3. Atiq M, Ahmed US, Allana SS, Chishti KN. Brain abscess in children. Indian J Pediatr 2006;73:401-4.
4. Bodilsen J, Dalager-Pedersen M, van de Beek D, Brouwer MC, Nielsen H. Incidence and mortality of brain abscess in Denmark: A nationwide population-based study. Clin Microbiol Infect 2020;26:95-100.
5. Brouwer MC, Coutinho JM, van de Beek D. Clinical characteristics and outcome of brain abscess: Systematic review and meta-analysis. Neurology 2014;82:806-13.
6. Brouwer MC, Tunkel AR, McKhann GM 2nd, van de Beek D. Brain abscess. N Engl J Med 2014;371:447-56.
7. Brouwer MC, van de Beek D. Epidemiology, diagnosis, and treatment of brain abscesses. Curr Opin Infect Dis 2017;30:129-34.
8. Danziger J, Allen KL, Bloch S. Brain-stem abscess in childhood.

Case report. J Neurosurg 1974;40:391-3.
9. Dou ZZ, Guo LY, Liu LL, Li MH, Hu HL, Hu B, *et al.* Clinical characteristics and outcome analysis of 94 children with brain abscess in beijing: A single-center retrospective study. Pediatr Infect Dis J 2021;40:109-15.
10. Fuentes S, Bouillot P, Regis J, Lena G, Choux M. Management of brain stem abscess. Br J Neurosurg 2001;15:57-62.
11. Ghannane H, Laghmari M, Aniba K, Lmejjati M, Benali SA. Diagnostic and management of pediatric brain stem abscess, a case-based update. Childs Nerv Syst 2011;27:1053-62.
12. Hadi Almuhammed H. Pediatric brainstem abscess: A case-based review. Med J Basrah Univ 2016;34:50-5.
13. Imai H, Ono N, Zama A, Tamura M. Diagnosis and treatment of brainstem abscess using magnetic resonance imaging and microsurgical aspiration-case report. Neurol Med Chir (Tokyo) 1995;35:160-4.
14. Jamjoom ZA. Solitary brainstem abscess successfully treated by microsurgical aspiration. Br J Neurosurg 1992;6:249-53.
15. Kashiwagi S, Abiko S, Aoki H. Brainstem abscess. Surg Neurol 1987;28:63-6.
16. Kim JH, Jung TY, Jung SH, Lee KH, Kim SK. Pediatric brainstem abscess with hemorrhage mimicking diffuse intrinsic pontine glioma: A case report. Childs Nerv Syst 2015;31:2359-62.
17. Kozik M, Ozarzewska E. A solitary abscess of the medulla oblongata. Eur Neurol 1976;14:302-9.
18. Mameli C, Genoni T, Madia C, Doneda C, Penagini F, Zuccotti G. Brain abscess in pediatric age: A review. Childs Nerv Syst 2019;35:1117-28.
19. Messina AV, Guido LJ, Liebeskind AL. Preoperative diagnosis of brain-stem abscess by computerized tomography with survival. Case report. J Neurosurg 1977;47:106-8.
20. Mut M, Hazer B, Narin F, Akalan N, Ozgen T. Aspiration or capsule excision? Analysis of treatment results for brain abscesses at single institute. Turk Neurosurg 2009;19:36-41.
21. Muzumdar D, Jhawar S, Goel A. Brain abscess: An overview. Int J Surg 2011;9:136-44.
22. Nauta HJ, Contreras FL, Weiner RL, Crofford MJ. Brain stem abscess managed with computed tomography-guided stereotactic aspiration. Neurosurgery 1987;20:476-80.
23. 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.
24. Prasanna Kumar M, Krishnamurthy S, Venkateswaran VS, Mahadevan S, Lalitha M, Sistla S, *et al.* Brainstem micro-abscesses caused by Burkholderia pseudomallei in a 10-month-old infant: A case report. Paediatr Int Child Health 2017;37:230-2.
25. Raffaldi I, Garazzino S, Gattinara GC, Lipreri R, Lancella L, Esposito S, *et al.* Brain abscesses in children: An Italian multicentre study. Epidemiol Infect 2017;145:2848-55.
26. Rajshekhar V, Chandy MJ. Successful stereotactic management of a large cardiogenic brain stem abscess. Neurosurgery 1994;34:368-71.
27. Ramdasi RV, Patil MK, Muzumdar DP. Solitary pyogenic brainstem abscess. Neurol India 2016;64:572-4.
28. Robert CM Jr., Stern WE, Brown WJ, Greenfield MA, Bentson JR. Brain stem abscess treated surgically. Wtih special note upon the employment of thorium dioxide. Surg Neurol 1975;3:153-60.
29. Russell JA, Shaw MD. Chronic abscess of the brain stem. J Neurol Neurosurg Psychiatry 1977;40:625-9.
30. Sadeghpour Tabaei A, Koochakzadeh L, Khorgami M, Tabaei SS. Acute lymphoblastic leukemia with infective endocarditis presented with unusual intracardiac mass. Case Rep Cardiol 2017;2017:1528416.
31. Sarmast AH, Showkat HI, Kirmani AR, Bhat AR, Patloo AM, Ahmad SR, *et al.* Aspiration versus excision: A single center experience of forty-seven patients with brain abscess over 10 years. Neurol Med Chir (Tokyo) 2012;52:724-30.
32. Shachor-Meyouhas Y, Bar-Joseph G, Guilburd JN, Lorber A, Hadash A, Kassis I. Brain abscess in children - epidemiology, predisposing factors and management in the modern medicine era. Acta Paediatr 2010;99:1163-7.
33. Suzer T, Coskun E, Cirak B, Yagci B, Tahta K. Brain stem abscesses in childhood. Childs Nerv Syst 2005;21:27-31.
34. van der Velden FJ, Battersby A, Pareja-Cebrian L, Ross N, Ball SL, Emonts M. Paediatric focal intracranial suppurative infection: A UK single-centre retrospective cohort study. BMC Pediatr 2019;19:130.
35. Vaquero J, Cabezudo JM, Leunda G. Nonsurgical resolution of a brain-stem abscess. Case report. J Neurosurg 1980;53:726-7.
36. Wabi G. Brain stem abscess. Indian Pediatr 1993;30:547-8.
37. Wang HS, Kuo MF, Huang SC. Medical cure of a brainstem abscess and serial brainstem auditory evoked potentials. Dev Med Child Neurol 1992;34:911-5.
38. Wiswell W, Jerzewski K, McGuire M. A rare case of a medulla oblongata brain abscess in a 23-month-old child. Pediatr Emerg Care Med Open Access 2019;4:1.
39. Xu B, Zhang Y, Yu J. Brainstem tuberculous abscesses successfully treated by microsurgical excision: A case report and review of the literature. Oncol Lett 2017;13:2708-12.
40. Yogev R, Bar-Meir M. Management of brain abscesses in children. Pediatr Infect Dis J 2004;23:157-9.
41. Yoshikawa T, Goodman S. Brain-abscess. West J Med 1974;121:207-19.

How to cite this article: Mohammad R, Crimmins D. Multiple Abscesses in the Frontal, Temporal and Brainstem regions in a 4.5-year-Old Girl- An Illustrative Case Report. Surg Neurol Int 2023;14:209.