J Exp Neurol. 2025;6(2):105-110.

Journal of Experimental Neurology Case Report

TICI 3 Revascularization after Atypical Presentation of Basilar Occlusion in the Setting of a Pediatric Patient

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Received date: April 01, 2025, **Accepted date:** May 14, 2025

Citation: Furr T, Weintraub A, Blanden A, Brescia N. TICI 3 Revascularization after Atypical Presentation of Basilar Occlusion in the Setting of a Pediatric Patient. J Exp Neurol. 2025;6(2):105-110.

Abstract

Stroke at a young age is a rarely reported entity whether it is uncommon or overlooked. When it occurs, it can result in lifetime neurological deficits and disabilities for the child and an overwhelming emotional and mental challenge to caregivers. One of the reasons that can lead childhood stroke to be overlooked is an atypical presentation onset with a headache or acute symptomatic seizure. As time is brain, these differences in childhood stroke can lead to a delay in diagnosis and symptoms considered as a stroke mimic. Furthermore, posterior circulation stroke can present nonspecific symptoms that can challenge the early diagnosis, leading to delay in treatment and missing the window of safety. Although guidelines recommend considering IV thrombolytic (last known well within 4.5 hours) or endovascular thrombectomy (EVT) (last known well within 6 hours) for children who may benefit from as long as images are confirming, there is still minimal data on childhood ischemic stroke. Here we report a case of a pediatric patient with basilar artery occlusion who presented with an uncommon presentation of thunderclap headache and received successful therapy with mechanical thrombectomy.

Keywords: Basilar artery, Brain imaging, Cerebrovascular regulation, Clinical Neurology

Introduction

Posterior circulation strokes account for around 20% of all acute ischemic strokes (AIS) [1]. It is evident that basilar artery occlusion (BAO) strokes have a poor outcome and can have up to 80% dependency or mortality despite thrombolysis or EVT [2]. BAO affects not only the brainstem including the pons and midbrain but also causes infarct of the thalamus and some parts of temporal and occipital lobes. The clinical picture can exhibit symptoms like headache, altered mental status, motor, sensory, visual, and oculomotor dysfunction, as well as vertigo and speech or swallowing issues [3]. Although challenging considering the nonspecific range of symptoms, early recognition is vital for BAO as nine hours after the symptom onset, the possibility of a favorable outcome is very low. Our case report represents an atypical presentation of BAO as a thunderclap headache as the main symptom, that could have been missed without extensive physical examination and diagnostic reasoning.

Case Presentation

We present a 16-year-old female with a past medical history of asthma, oral contraceptive (OCP) use, and acne taking spironolactone who presented with a thunderclap headache. Full case timeline can be seen in **Table 1**. History begins with the patient being in her usual state of health until the day before the hospital presentation when she woke up feeling unwell, nauseated, dizzy and had a severe thunderclap headache at midnight. When attempting to stand and walk she was off balance. Due to these symptoms, she was brought to the pediatric emergency department by her parents. On presentation, she was agitated and encephalopathic requiring intubation for airway protection and respiratory assistance. CT head without contrast was obtained which showed no acute pathologies and she was admitted to the pediatric intensive care unit (PICU). Neurology was consulted for evaluation of thunderclap headache later that day at 1100.

Table 1. Showing case timeline while patient was in hospital and outpatient setting on discharge.	
Case Timeline:	
1.	Presented with thunderclap headache, nauseated, dizziness and off balance
2.	CT head wo contrast showing no acute pathologies, needing intubation for encephalopathy and respiratory assistance.
3.	Later that day, neurology consulted showing concerning physical examination and emergently seen for CT angiogram arterial and venous phase; concluded with showing BAO. Hyperacute MRI showing DWI/FLAIR Mismatch and acute infarct in the pons.
4.	Received TICI 3 revascularization with mechanical thrombectomy.
5.	Ultrasound transcranial doppler showing grade 3 right to left interatrial or intrapulmonary shunt. Bilateral venous doppler negative.
6.	Hyper-coagulable work up negative.
7.	Discharge NIHSS 3 for limb ataxia and partial gaze palsy.
8.	Genetic testing done in the outpatient setting negative, remained on aspirin 81 mg daily and refrain from estrogen containing pills and spironolactone.

The Bedside examination was concerning for demonstrated tonic extensor posturing to noxious, equal round and reactive pupils, sluggish vestibula-ocular reflexes, and corneal stimulation yielded extension. Additionally, she demonstrated bilateral ankle clonus, significant hyperreflexia, positive Babinski reflex, and pathologic split of Achilles reflex to the iliopsoas. NIHSS score was at 30 (LOC 3, LOC questions 1, LOC commands 2, visual 3, upper extremity drift 4 and 4, lower extremity drift 4 and 4, language 3, extinction and inattention 2).

Due to these findings, she was emergently sent for a CT angiogram arterial and venous phase to evaluate for an aneurysm, developing hydrocephalus, dural venous sinus thrombosis, swelling, and herniation due to meningitis or other inflammatory conditions, and other differentials for causes of a thunderclap headache **(Table 2)**. The scan demonstrated a BAO **(Figure 1)**. Hyperacute MRI was obtained which demonstrated significant DWI flair mismatch of an acute infarct in the pons and she was transferred to the angiography suite for mechanical thrombectomy in which she received a TICI 3 revascularization **(Figures 2** and **3)**.

After the mechanical thrombectomy, post-operative imaging was significant for DWI hyperintense lesions smaller than those shown on the MRI before the procedure **(Figure 3)**. There was significant FLAIR hyperintensity in the pons indicative of edema with anterior pons leptomeningeal contrast enhancement.

She remained in the pediatric ICU for two days and had a thirteen-day hospital course followed by a rehab course for 10 days. On the day of discharge, her NIHSS score was 3 for limb ataxia and partial gaze palsy (Intranuclear ophthalmoplegia (INO) on the right and 6th nerve palsy on the left). An extensive stroke in young workup was done to understand the etiology

Table 2. Differential diagnosis for thunderclap headache categorized in common, less common, and uncommon/rare causes.
Differential Diagnosis Thunderclap Headache
Common Causes:
Subarachnoid hemorrhage
Reversible cerebral vasoconstriction syndromes
Less Common Causes:
Cerebral venous thrombosis
Cervical artery dissection
Infections
Spontaneous intracranial hypotension
Ischemic stroke
Posterior reversible encephalopathy syndrome
Uncommon/Rare Causes:
Pituitary apoplexy
Colloid cyst of 3rd ventricle

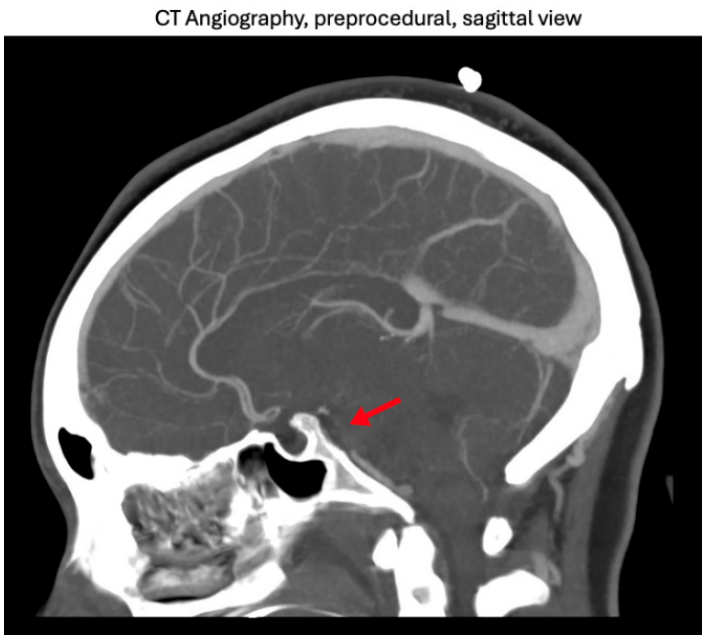

Figure 1. Preprocedural CT angiography representing occluson of basilar artery (red arrow).

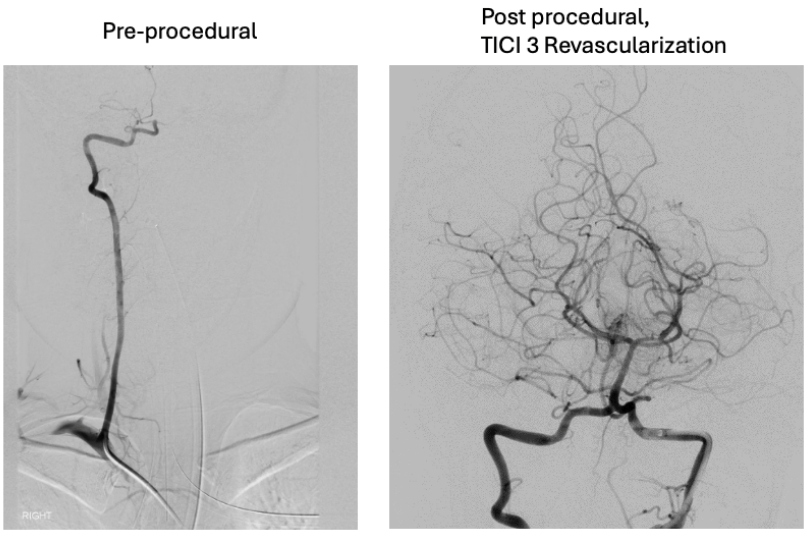

Figure 2. Pre-procedural angiography showing occlusion at basilar artery (left). Post procedural angiography with TICI 3 revascularization post mechanical thrombectomy (right).

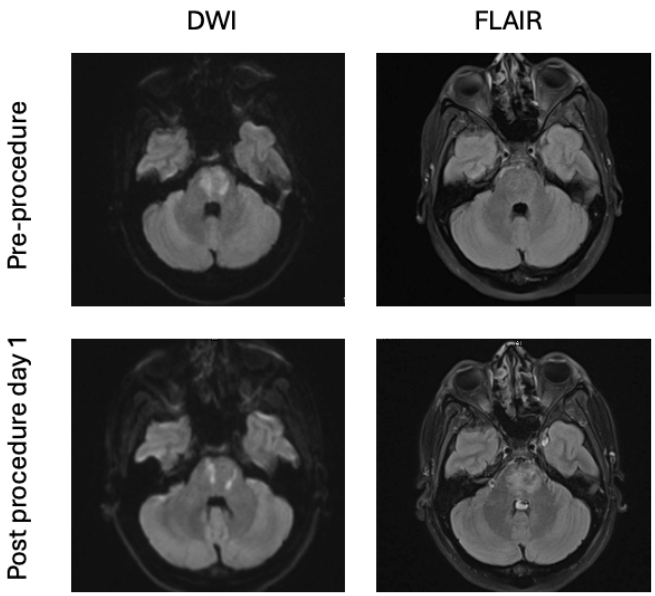

Figure 3. MRI without contrast. DWI and FLAIR pre-procedure and post-procedure day 1.

and was found to have a grade 3 right-to-left interatrial or intrapulmonary shunt that was demonstrated on transcranial Doppler ultrasonography. Hyper-coagulable workup was negative for protein S levels, low fibrinogen, and no elevation in autoimmune levels. Lumbar puncture was negative for bacterial, viral, and fungal causes of infection. Bilateral lower extremity venous doppler study was done which ruled out DVT. Outpatient TEE with bubble was planned and in the meantime she was loaded with aspirin and was started on aspirin 81 mg daily. The presumed etiology was determined as paradoxical embolism in the setting of atrial versus pulmonary shunts with a hypercoagulable state due to OCP use. Outpatient genetic testing was planned to rule out hereditary hypercoagulability syndromes, as it has been noted that ischemic strokes are rare in pediatric populations with even more rarity of ischemic strokes being BAO. Patient's 3-month follow-up had an NIHSS of 0 with a remarkable recovery from her basilar stroke. Her genetic testing came back as a carrier for the *GJB2* and *STXBP2* genes deeming her to be negative for a genetic hyper-coagulable cause of her stroke. A TEE was done in the outpatient study which showed a small atrial shunt through a patent foramen ovale. She had it occluded with a 25 mm gore cardioform septal occluder. Her continued therapy will be to remain on aspirin 81 mg daily and to refrain from oral combined estrogen containing pills and spironolactone. Anticoagulation was considered, but we deemed that aspirin and anticoagulation at the same equipoise as there was absence of evident venous thrombosis or hypercoagulable state with everything being negative. The OCP use can be held for remainder of life.

Discussion

The basilar artery is likely the most important artery to the posterior circulation of the brain. What makes a stroke so devastating in this area is that the artery supplies several life sustaining areas of the brain. The diagnosis of basilar artery occlusion in the pediatric population can be difficult to make as the presentation can present like many other neurological pathologies including seizures at onset of the stroke and also a locked in state or unresponsiveness. Our patient initially presented with a finding that could be mimicked as a subarachnoid hemorrhage or one of the headache pathologies which can be seen in **Table 2**. Ultimately, being the uncommon nature of a BAO in a pediatric population, there should be an extensive work-up even if half of the time there will be a conclusion of no clear etiology.

Multiple studies are showing the safety and efficacy of EVT on large and medium vessel occlusions of anterior circulation. At the same time, a sparse number of trials have been published for BAO. BEST (Basilar Artery Occlusion Endovascular Intervention versus Standard Medical Treatment**)** and BASICS (The Basilar Artery International Cooperation Study) trials revealed a possible benefit of EVT in patients who presented with moderate to severe symptoms with lack of significant

benefit on outcomes at 3 months after stroke [2]. However, with this trial, many issues may have hindered the validity of the results.

Henceforth, ATTENTION (Endovascular Treatment for Acute Basilar Artery Occlusion) and BAOCHE (Basilar Artery Occlusion Chinese Endovascular) trials were designed to compare EMT versus medical therapy within 24 hours after the onset of symptoms (within 12 hours, and 6-24 hours respectively). They showed significant benefit in lowering disability and mortality, specifically for the patients presented with moderate-to-severe symptoms [4,5]. ATTENTION and BAOCHE trials showed EMT was beneficial independent of baseline characteristics, intracranial atherosclerotic disease (ICAD), and age and time passed since the onset of symptoms [4,5]. The intracranial bleeding risk with BAO EVT was similar to the anterior circulation EVT patients. It is important to take note that with these studies, the patient population criteria was over the age of 18 years old.

However, when we discuss intravascular treatment for the pediatric population, the studies are sparse. Intravascular treatment is controversial as there is a difficult nature of performing clinical trials on pediatric patients due to the ethical dilemma of control groups not getting potential beneficial treatment and also low incidence of stroke in the pediatric population. Case control studies have shown there to be superior clinical outcomes with mechanical thrombectomy over medical management for large vessel occlusions [6]. There also shows benefits in long-term and short-term neurological outcomes after basilar artery thrombectomy for patients aged 1-18 [7]. We now know that mechanical thrombectomy has its advantages but still we must understand the risks in the pediatric population. These include diminished artery size in children, weight-dependent restrictions for contrast, and potential radiation exposure [8].

In the 2015 AHA guideline, it was stated that mechanical thrombectomy may be reasonable with patients under 18 years of age while acknowledging the risks are not established yet [9]. This prompts the necessity to have a discussion with the patient and their family members about the potential risks that may become evident. With the right patient based on no contraindications to mechanical thrombectomy and a clear understanding from the family about the potential risks and lack of clinical trial evidence, we are seeing more institutions offering pediatric mechanical thrombectomy to the basilar artery. As seen with our patient, we thought it was deemed appropriate to advance with mechanical thrombectomy given the natural history of the patient's stroke having preserved brainstem function on exam, and out of the thrombolytic window. It was important to discuss the risk and benefits with the family. Discussion about the limitation of the studies for mechanical thrombectomy in this age group was done. Recovery for these patients can have no recovery, permanent neurological disability or death with or without the procedure. Fortunately, this ended with significant improvement for the patient as her NIHSS was a 3 on discharge and an NIHSS of 0 on follow-up.

Conclusion

Our patient presented with an atypical presentation that led to the initial thinking of subarachnoid hemorrhage for her headache and infection for her altered mental status. With key physical examination and proper study ordering, the patient was properly diagnosed with BAO and treated properly to optimize full recovery. There was clear evidence of a successful mechanical thrombectomy for this pediatric patient having full recovery. This case report brings light on the efficacy of mechanical thrombectomy of BAO in the pediatric population and also a clear understanding of BAO as a differential diagnosis in an atypical presentation.

Acknowledgement

We want to recognize our mentors, advisors, and colleagues at SUNY Upstate University Hospital for support with making this case report. We also want to thank the patient and family to allow us to present this patient's case.

Author Contribution Statement

All authors that are listed in this manuscript played a large part and provided a large contribution with the making of the manuscript and editing. All contributions were made in an equal fashion.

Funding/Conflicts of Interest/Disclosures

There are no conflicts of interest, funding received, or disclosures to report for this manuscript.

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