

Case Report

Acute posttraumatic pediatric cerebral venous thrombosis: Case report and review of literature

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Abstract

Background: Pediatric cerebral venous thrombosis (CVT) is a common sequelae of infection, coagulopathies, and dehydration in the pediatric population. Acute posttraumatic CVT is an uncommon etiology of pediatric CVT that presents a unique management challenge. There are no established guidelines outlining the treatment of this small subset of patients.

Case Description: We present a case of a 12-year-old boy with posttraumatic CVT who was safely treated with anticoagulation therapy, and had resolution of his symptoms and radiographic improvement within 3 days of therapy. The relevant literature is reviewed.

Conclusion: Anticoagulation therapy may be safely used in the treatment of acute posttraumatic CVT in pediatric patients, and may reduce the incidence of clot propagation, hospitalization time, and cost of treatment.

Key Words: Anticoagulation, cerebral venous thrombosis, head trauma, pediatric, sigmoid sinus thrombosis, trauma

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INTRODUCTION

Pediatric cerebral venous thrombosis (CVT) is a potential life-threatening condition that requires a high index of suspicion to diagnose. The etiologies of pediatric CVT are multifocal and include underlying infection, coagulopathies, and trauma.^[16] Among these, trauma-induced CVT is relatively uncommon but poses a unique management challenge. In a literature review of pediatric CVT, 846 cases were reported [Table 1];^[1-10,12-14,16-22] only 28 cases (3%) were reported in the context of a head trauma. Three of the cases reported did not specify the treatment the patient eventually received, 23 cases received no intervention,

all of which achieved complete resolution of symptoms over the course of several weeks (2-24 weeks), and the remaining 2 cases received anticoagulation (AC) therapy and made a complete recovery. We seek to report a well-documented case of acute posttraumatic CVT in a 12-year-old boy who was successfully treated with AC therapy, which resulted in improved cerebral venous flow within 3 days of therapy.

Review method

We used the following search terms to find reported cases of pediatric CVT in Google Scholar Search: "Cerebral venous thrombosis in childhood", "post trauma cerebral venous thrombosis", "anticoagulation pediatric cerebral

Table 1: Literature review of reported cases of pediatric cerebral venous thrombosis and management

Reported cases of pediatric CVT							
Author	Year	Time interval (years)	Total cases of pediatric CVT	Trauma induced cases	Treatment of all Pts	Treatment of post-traumatic CVT	Recovery of post-traumatic CVT
Belman (2)	1990	1	1	0	Treated underlying cause	-	-
Barron (1)	1992	10	25	0	Treated underlying cause	-	-
Medlock (10)	1992	6	13	0	Treated underlying cause	-	-
Taha (20)	1993	4	5	5	-	Conservative	Complete after 2 to 10 weeks in 4 pts, 1 pt complete after 6 months
Rich (14)	1993	1	1	0	Treated underlying cause	-	-
deVeber (5)	1998	4	32	0	22 pt treated with AC; treated underlying cause	-	-
Vielhaber (21)	1998	3	32	0	-	-	-
Philips (13)	1999	Not reported	6	0	Thrombolytic	-	-
Stiefel (19)	2000	2	8	8	-	Conservative	Complete after 3 to 24 weeks
Carvalho (3)	2001	13	31	0	Treated underlying cause	-	-
Huisman (7)	2001	4	19	9	2 pts with AC; treated underlying cause	Conservative	Complete
deVeber (4)	2001	6	160	0	53 pts with AC; treated underlying cause	-	-
Keane (8)	2002	1	1	0	AC	-	-
Sousa (17)	2004	1	1	1	-	Conservative	Complete
Sebire (16)	2005	Meta analysis	42	2	18 pts with AC; treated underlying cause	Not reported	-
Muthukumar (12)	2005	1	1	1	-	Anticoagulation	Complete after 3 weeks
Kenet (9)	2007	9	396	1	250 pts with AC; treated underlying cause	Not reported	-
Soysal (18)	2008	1	1	0	Thrombolytic + AC	-	-
Wasay (22)	2008	9	70	0	3 pts Thrombolytic; 15 pts AC; treated underlying cause	-	-
Georgoulis (6)	2011	1	1	1	NA	Anticoagulation	Complete after 6 months of therapy
Total			846	28 (3%)			

CVT: Cerebral venous thrombosis, AC: Anticoagulation

venous thrombosis”, and “sigmoid sinus thrombosis after closed head injury in children”. Reported articles were reviewed for cases of posttraumatic CVT in pediatric patients, treatment methods and outcomes were evaluated and reported in Table 1. Reviews that contained mixed age groups (adults and pediatric patients) were used, however, only the pediatric cases were considered.

CASE REPORT

History of present illness

We report a case of a 12-year-old boy with no significant past medical history who presented at Phoenix Children’s Hospital Emergency Room following a motor vehicle accident (MVA). The patient was sharing a seatbelt with one other sibling in the back seat of a 4-door sedan. The vehicle was involved in a “T-bone” collision, which resulted in a temporal head injury against the side door. At the time of presentation, the patient complained of neck tenderness, pain at the right skull base, and nausea.

The patient did have poor recollection of the accident; however, there was no indication of loss of consciousness. The patient denied any neck or head pain prior to the accident, and denied any fever, chills, emesis, or seizures. Prior to his presentation, the patient was otherwise healthy, performed well in school, has no family history of coagulopathies, was not taking any medications, and had a negative review of systems except what was discussed earlier. On physical examination, the patient was alert and oriented and appeared well nourished and well hydrated. His pupils were equal and reactive to light, extraocular movements were intact, his smile was symmetric and his hearing present in both ears. He had no otorrhea or rhinorrhea, but some ecchymosis was noted around the right ear. There was some neck tenderness at the level of C3, a neck brace was applied as a precautionary measure pending imaging results. He had full power and normal tone in his upper and lower extremities. His pulses were equal bilaterally to palpation. Overall, the patient was clinically stable but remained somewhat confused with

a Glasgow coma scale (GCS) of 14. The patient was admitted to the pediatric intensive care unit pending imaging studies.

Initial imaging studies

A computed tomography (CT) of the head [Figure 1a] on presentation revealed a small petrous bone fracture on the right side of the skull. Several small pneumocephalic collections were evident surrounding the fractured mastoid air cells. Bony fragments were visible within the vicinity of the sigmoid sinus. A magnetic resonance venography (MRV) [Figure 1b] was ordered to evaluate the cranial vascular integrity, which demonstrated a flow void of the right sigmoid sinus and the right external jugular vein.

Hospital course

The patient's coagulation profile was within normal limits, he was admitted and started on IV heparin and monitored with serial exam for changes in neurologic function. The patient remained stable and on the third day of admission, a repeat MRV [Figure 2] demonstrated persistent flow void of the sigmoid sinus, but improved flow through the external jugular sinus. At this point, the patient's nausea and headache had resolved and he was switched to low molecular weight heparin (enoxaparin) and transferred to the pediatric floor. The remainder of the hospitalization was uneventful, the patient was discharged on day 7 with 6 weeks of low-molecular weight heparin (LMWH) therapy with weekly clinical follow up.

DISCUSSION

The incidence of CVT is 0.67 per 100,000 children per year, neonates make up to 43% of reported cases.^[4] CVT in neonates presents with seizures and nonspecific neurologic signs (change in eating habits, irritability, poor sleep, etc.).^[4] In contrast, older children most commonly present with headache, papilledema, lethargy, and focal neurologic signs.^[3,4,16] In children, CVT most commonly affects a

single dural sinus, the superficial sagittal and transverse sinuses represent 47.5% and 12% of cases, respectively; in the same study, 30% of cases involved more than two sinuses.^[9] There are numerous risk factors associated with pediatric CVT, underlying infection is the most common risk factor, however, dehydration, coagulopathies, and malignancies are common predisposing conditions.^[3,4,9] Among these, head trauma represents a smaller subset and we estimate it to be an important risk factor in 3% [Table 1] of pediatric CVT. Older children are more likely to present with posttraumatic CVT^[7] and the thrombosis occurs acutely (1-5 days of presentation).^[6,17,20] Although posttraumatic CVT can occur in the absence of a skull fracture, most cases involve a bone fracture along the adjacent dural sinus.^[7,20]

Diagnosis

It is important to consider the age of the patient when using diagnostic imaging in suspected cases of pediatric CVT. Although in the setting of head trauma, CT is commonly used to evaluate for possible intracranial hemorrhage and fractures, suspected thrombosis requires further studies to confirm the diagnosis if time permits. CT is only 84% accurate in diagnosing CVT in children,^[4] for this reason, MRV is an important tool to confirm or rule out CVT when it is suspected. Surprisingly, a small series of trauma-induced CVT showed the traditional CT finding of "empty delta" sign in all of their reported cases ($n = 9$).^[7] It is important to note, however, that such findings are limited by the retrospective nature of the study design. In neonates, Doppler ultrasound can be 83% sensitive in diagnosing CVT^[4] and has the added benefit of limiting radiation exposure, which is easily accessible, and more cost effective than conventional CT.

Management

Per published CTV guidelines,^[15] our patient was evaluated for underlying thrombocytopenia,

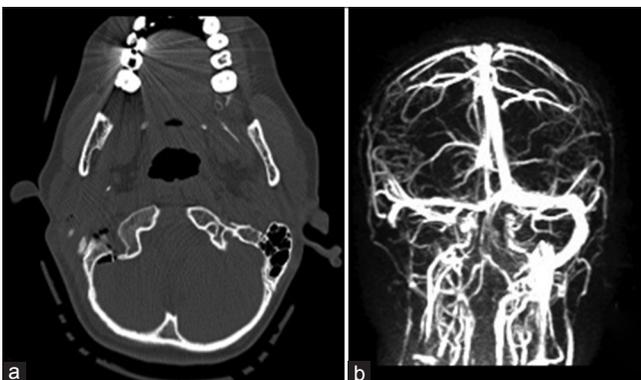


Figure 1: A 12-year-old boy with evidence of (a) petrous bone fracture and pneumocephalus on axial CT at the time of presentation. (b) MRV at the time revealed a flow void of the sigmoid sinus and external jugular vein, confirming the diagnosis of sigmoid sinus thrombosis



Figure 2: Repeat MRV demonstrating persistent flow void through the sigmoid sinus, but improved flow through external jugular vein after 3 days of anticoagulation therapy

coagulopathies, and infection all of which were negative. Unlike many of the published case reports of posttraumatic CVT,^[17,19,20] which managed patients without medical or surgical intervention, our patient was initially managed with IV heparin and subsequently switched to subcutaneous LMWH as an outpatient. Although the decision to use AC therapy in the setting of a head trauma with evidence of a petrous bone fracture can be daunting, a recent update in the management of nonneonatal pediatric CVT recommend the use of such therapy even in setting of nonsignificant intracranial hemorrhage.^[15] In the cases reviewed, only 2 out of the 28 (7%) posttraumatic CVT cases used AC therapy. In the first case,^[12] a patient with right transverse sinus thrombosis was found to have an elevated antiphospholipid antibodies (16.1 IgM phospholipid [MPL] U/ml; normal <10 MPL U/ml) a known risk factor for CVT. The patient was treated with acenocoumarol with a goal international normalized ratio (INR) of 2.5-3 for 3 weeks as inpatient; there were no complications of therapy reported. The second case^[6] reported a presentation of posttraumatic pediatric CVT with evidence of intraparenchymal hemorrhage adjacent to a thrombosed sigmoid sinus. That patient was bridged with 5 days of subcutaneous heparin to warfarin therapy for 6 months with a target INR <2.0. There was no reported complication of this treatment regimen, and the patient made a complete recovery after the 6-month period. Similarly, in our reported case of posttraumatic pediatric CVT, the patient was initially treated with IV heparin for the first 24 h period, and later switched to LMWH. The reversibility and minimal daily management of heparin therapy makes it an excellent choice for AC therapy, especially if there is evidence of worsening intracranial hemorrhage. This treatment option was also easy to administer by the family, and allowed the patient to be treated as an outpatient with weekly coagulation studies. Following reevaluation after the initial 6 weeks of Lovenox (enoxaparin sodium) injections, the patient was continued on this therapy for a total of 3 months before being transitioned to 3 months of daily Coumadin, which was more easily tolerated by the patient who completed a total of 6 months of AC therapy per pediatric CVT guidelines.^[15] During this time the patient made a complete recovery with no complications.

Although others have decided to opt for the use of conservative therapy in the treatment of posttraumatic pediatric CVT,^[7,17,19,20] it is important to stress that conservative treatment is not benign, and the lack of reported complications in the literature is not necessarily an indication of its safety, but rather a consequence of a low incidence rate (3%). Although there are no published randomized control trials evaluating the use AC therapy in the treatment of pediatric CVT, a

pilot cohort study of 22 pediatric patients with CVT reported the use of various AC (heparin, LMWH, and warfarin) regimens and reported no deaths or clinically significant intracranial hemorrhage.^[5] Importantly, a larger prospective study demonstrated that conservative therapy was associated with propagation of venous thrombi when compared with AC therapy.^[11] In a multicenter European registry of pediatric CVT, AC therapy was used in 250 (65%) patients; of these 1 patient died after therapy, importantly however, 7 (5%) patients who were treated conservatively died secondary to edema-induced herniation,^[9] a complication of the underlying CVT. Similarly, a smaller multicenter study treated 15 (20%) patients with AC therapy, of which 1 died; unfortunately, 7 children who were managed conservatively died from complications related to the underlying CVT.^[22] These findings making a compelling argument for the early use of AC intervention, which may help reduce the reported morbidity/mortality associated with the conservative management of pediatric CVT.

Prognosis

After 3 days of AC therapy, our patient's symptoms of nausea and headache had completely resolved. On the third day of treatment, a repeat MRV [Figure 2] demonstrated a persistent flow void in the sigmoid sinus; however, flow through external jugular vein had improved. On last follow-up (6 months postinjury) the patient continues to be asymptomatic and experienced no negative sequelae from AC therapy. This rapid recovery is most likely attributed to the use of AC therapy, and the time to symptomatic and radiographic improvement is an important factor when considering the cost and efficacy of treatment. In the literature, the prognosis of pediatric CVT was mostly dependent on presenting symptoms and underlying condition; seizure, venous infarcts, and coma were predictors of poor outcome.^[4,22] Importantly, it has been reported that nonadministration of AC therapy is an independent risk factor for recurrent CVT.^[9]

CONCLUSION

Acute posttraumatic CVT is an uncommon etiology of pediatric CVT, as such its management is not clearly established. We report a case of posttraumatic CVT in a 12-year-old boy who was safely treated with AC therapy, and had symptomatic and radiographic improvement within 3 days of therapy. This readily reversible medical therapy may help reduce mortality and morbidity associated with conservative management. In addition, the simplicity of drug management may hasten recovery and thereby reduce hospitalization time. Further randomized controlled trials are necessary to fully assess the efficiency of such therapy in this specialized patient population.

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