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Post-Traumatic Vein of Labbe Thrombosis Presenting as Hemorrhagic Venous Infarct in a Pregnant Female: An Interesting Rare Entity with Diagnostic Challenges

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Abstract

Vein of Labbe thrombosis is an uncommon subtype of cerebral venous thrombosis (CVT). It clinically presents as headache, focal seizures, mass effect or altered sensorium. Severity of illness is multifactorial: degree of thrombosis, patency, blockage or hypoplasia of opposite vein, presence or absence of collateral anastomosing circulation, proximity with ventricles, size of infarct etc. Though it's a rare site to be involved by thrombosis but most frequent among young pregnant women as a subtype of CVT. There are five variants of Vein of Labbe reported in literature among which left sided type I Vein of Labbe variant is more common among young females. Therapeutic dose of Low molecular weight heparin (LMWH) is the treatment of choice in acute event of CVT during pregnancy followed by prophylactic dose till postpartum period. We present a rare and interesting case of Vein of Labbe thrombosis in which history of trauma misled the early diagnosis. A young pregnant female presented to ED with complaints of loss of consciousness, vomiting, generalized headache and right ear bleeding within 24hours of encountering a road traffic accident. The patient was evaluated as cerebral injury and Vein of Labbe (left) thrombosis was an incidental finding noticed in MRV imaging with normal platelet counts and coagulation profiles. She was managed conservatively and show drastic improvement without administration of LMWH. Antithrombotic agents were not used in this case because of her progressive clinical improvement, reduction in size of infarct with time, no evidence of development of new infarct, normal coagulation profile and post-traumatic thrombotic event. This case is presented to emphasize that CVT may develop secondary to trauma and not always as a consequence of hypercoagulable state in pregnancy, vein of Labbe thrombosis must be looked into consideration in traumatic case of CVT if patient is a young pregnant women and antithrombotic agents are not mandatory in treatment if coagulation profile is normal. This case is interesting in its clinical presentation, development of CVT in spite of normal coagulation profile, rare site of thrombosis (Vein of Labbe), clinical improvement without administration of anti-thrombotic agents, good clinical outcome and prognosis with no residual effects.

Keywords: Cerebral Venous Thrombosis (CVT); Vein of Labbe; Pregnancy; Antithrombotic Agents

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Abbreviations

CVT: Cerebral Venous Thrombosis; CTA: Computed Tomography Angiography; MRV: Magnetic Resonance Venogram; ED: Emergency Department; GCS: Glasgow Coma Scale; LMWH: Low Molecular Weight Heparin; ICP: Intracranial Pressure; CSF: Cerebrospinal Fluid; FLAIR: Fluid-Attenuated Inversion Recovery; DWI: Diffusion Weighted Imaging; ADC: Apparent Diffusion Coefficient; SDH: Subdural Hemorrhage

Introduction

Cerebral venous thrombosis (CVT) is a common complication during pregnancy, secondary to the underlying physiological hypercoagulable state. The risk of CVT increases with trimesters. Pubmed searching tool shows probably the first reported case of thrombosis and embolism of the cerebral blood vessels in pregnancy, labor and puerperium in 1963 [1]. Incidence of CVT is higher among young adults (31 - 50 years) with female predominance (F:M ratio is 1.86:0.75) [2]. Coutinho JM., *et al.* reported CVT among 18% pregnant female in their study [3]. CVT is responsible for 2% of strokes during pregnancy and peripartum period [4].

The usual clinical presentation of CVT may vary from nonspecific symptoms like headache, nausea/vomiting and seizures to focal neurological deficits and altered sensorium. These are reversible with early diagnosis and optimal management.

Post-traumatic venous infarct is a rare entity presenting during pregnancy and difficult to diagnose in absence of symptoms related with localized mass effect or seizures. The clinical outcome will depends upon multiple factors like degree and extent of trauma, type of trauma, time interval between traumatic event and initiation of hospital care, associated comorbidities (obesity, hyperlipidemia, coagulation abnormality, infection, malignancy etc), previous medications, feasibility and availability of diagnostic modality, patient compliance towards therapy etc.

Thrombosis of Vein of Labbe is an uncommon event in pregnancy with variable clinical presentation. The risk factors for Vein of Labbe thrombosis among female population include young age, pregnancy, puerperium, use of oral contraceptive pills, coagulation factor abnormality, Anti-phospholipid syndrome, autoimmune disorders etc. According to Ramsawak L., *et al*, left sided Vein of Labbe thrombosis is more common than right sided [5]. The extent of cerebral injury and disease severity is dependent upon presence of collateral circulation, unilateral and or bilateral thrombotic event and stage at which interventions initiated after insult.

The diagnostic modalities available for CVT include Magnetic resonance imaging with venogram (MRV) and computed tomography angiography (CTA). In pregnancy MRV is the gold standard and CT will be preferred when MRV is inconclusive. The clinical outcome is usually favorable with good prognosis [5].

Several case reports and systematic reviews reported that there is increase in risk of CVT with gestational age however obstetric outcome is unrelated with CVT. Young age and pregnancy is among the most important risk factor in development of CVT secondary to hormonal imbalance.

The treatment of choice for single acute event of CVT is low molecular weight heparin (LMWH) in therapeutic dose because it neither cross the placenta nor affect the fetal wellbeing. LMWH is also preferred in post-acute phase in pregnancy and during lactation. Prophylactic dose of LMWH will be continued throughout the pregnancy and peurperium [6].

We present this case because of several rare things associated with it - post-traumatic rare complication associated with pregnancy, rare clinical presentation in presence of hemorrhagic infarct, rare venous infarct affecting unilateral Labbe vein presenting as right ear bleeding, challenges in selection of diagnostic modality and therapeutic management in view of pregnancy, no role of antithrombotic agents.

Case Report

A young female with 28 weeks of pregnancy was transferred into ED from a local hospital after primary stabilization within 24 hours of encountering a road traffic accident with loss of consciousness for five minutes, two episodes of vomiting, generalized headache, and right ear bleeding. On examination she had GCS score of 12 ($E_4V_3M_5$), tachycardia and hypotension. Her airways were patent and stable vitals. She had no cervical spine tenderness and other neurological examinations were within normal limit. On obstetric history she was found to have gravida three and two live births (P_2L_2). The hematological examination shows normal hemoglobin and platelet count with mild leucocytosis and neutrophilia.

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Her coagulation profile and biochemical parameters were unremarkable although her D-Dimer was mildly elevated at 24 hours of onset of event.

MRI Brain: A large area of hyperintensity in the left temporal

lobe causing mass effect over adjacent neuroparenchyma in the

form of effacement of sulcogyral spaces, ipsilateral lateral ventricle with midline shift of 7.5 mm to right was observed suggestive of intraparenchymal bleed [Figure 1]. Undisplaced fracture of right squamous temporal bone extending longitudinally to the middle ear and right hemomastoid with collection in the right middle ear cavity was observed on CT Brain.

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Figure 1: MRI Brain: Area of hemorrhagic infarct (Yellow arrow), Midline shift (arrow head).

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MRI venogram of Brain reported a large area of blooming in the left temporal lobe causing mass effect over adjacent neuroparenchyma in the form of effacement of sulcogyral spaces, ipsilateral lateral ventricle with midline shift of 7.5 mm to right suggestive of intraparenchymal bleed. Areas of FLAIR hyperintensities showing diffusion restriction on DWI with corresponding low signals on ADC involving the periphery of the above lesion and left frontal lobe suggestive of post – traumatic acute infarcts. In addition to this thin rim of subdural hematoma (SDH) involving left frontal and temporal region and SDH involving left parietal lobe was also observed (Figure 2).



Figure 2: MR veinogram.

After the diagnosis, patient was managed conservatively and obstetric consultation obtained. Drugs safer in pregnancy were administered. (Table 1). Anticonvulsants started following which her symptoms subside gradually thereafter. The use of antithrombotic agents is the issue of concern in CVT secondary to traumatic head injury, patent major vessels of CNS, normal platelet count and coagulation profile and due to anastomosing type of vein of Labbe. She was responding well hence shifted to respective parent department where she was followed up as per obstetric care guidelines.

Discussion

CVT is a rare disease with low incidence (5 per million) and female predominance but serious neurologic disorder that is potentially reversible with prompt diagnosis and appropriate medical treatment. The incidence is more often in pregnancy and pu-

Name of drug	Dose	Duration	Category
Inj Levetiracetam	1gm stat f/b 500 mg BD	3 days	С
Inj Mannitol 20%	100 ml in 30 min TID	3 days	В
Inj Dexamethasone	4 mg BD	3 days	
Inj Amoxicillin + clavulanic acid	1.2 gm BD	5 days	В

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Table 1: Medications used in treatment.

erperium (11.6 per million). The disease is associated with a wide spectrum of etiologic factors and clinical presentations. Main risk factors include hypercoagulable states, medications (OC pills, steroids), post-trauma, post lumbar, pregnancy, puerperium, Inflammatory bowel disease, hyperhomocysteinemia and collagen vascular diseases [7]. Etiologically CVT may be secondary to abnormalities in pattern of blood flow, abnormalities in coagulation profiles and infiltrative or inflammatory conditions. A study by Enevoldson., *et al.* reported specific etiology for CVT are protein S deficiency, protein C deficiency, dural arteriovenous malformation, intracerebral arteriovenous malformation, bilateral glomus tumors, systemic lupus erythematosus, Wegener's granulomatosis, Non-Hodgkin's lymphoma [8].Majority of the events are secondary to focal or systemic coagulative derangements.

Patho-physiological explanation of absence of focal neurological deficit in present case is unilaterality of the lesion resulting in maintained collateral circulation. CVT delays venous emptying and thus reduces in CSF reabsorption resulting in an increase in venous and capillary pressure. This may lead to focal edema secondary to increased retrograde pressure characterized by venous dilatation, hematoma and focal non-systematized ischemic injury. These may cross arterial boundaries and show features of vasogenic and cytotoxic edema.

The most common symptom is headache with most common venous sinus involved are superior sagittal sinus and transverse sinus.

Post-traumatic development of CVT are very rare cases that are usually unsuspected and easily missed cause of delayed intracerebral hemorrhage following head trauma [9].

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Cerebral venous thrombosis in closed head trauma is a call to look beyond fractures and hematomas. Cerebral imaging is a fundamental diagnostic modality of choice for CVT diagnosis. MRV is the gold standard for diagnosis of Vein of Labbe thrombosis because it is more sensitive for deeper venous structures. Contralateral hypoplastic venous draining sinuses are associated with elevated intracranial pressure (ICP) in unilateral cerebral venous thrombosis resulting in bleeding manifestations. ICT is usually normal in Vein of Labbe thrombosis as was in present case.

Vein of Labbe, also known as inferior anastomotic vein, is a part of superficial central venous system. It drains the temporal lobe and tissue adjacent to the Sylvian fissure. It is the most important draining vein of the temporal lobe. It crosses the temporal lobe between the Sylvian fissure and the transverse sinus. It is the largest venous channel on the lateral surface of the brain that connects the superficial middle cerebral vein and inferior cerebral vein draining into the transverse sinus. The superior anastomotic vein of Trolard is usually smaller, connecting the superior sagital sinus and the superficial middle cerebral vein. The neuroanatomic topographical typing by Minca DI., *et al.* reported Type 1 (Squamosal-petrosalmastoid) Vein of Labbe variant in <50% cases they studied. They reported that left sided type I Vein of Labbe variant was more common among females than right sided [10].

Sasaki., *et al.* suggested that the proximity of cerebral cortical veins to the overlying dura predisposes them to surgical injury therefore Type 1 variant of Vein of Labbe interruption during otologic surgery may produce devastating neurologic consequences. One should take care not to damage Type 1 Vein of Labbe variant while approaching tegmen tympani [11].

Naidoo J., *et al.* concluded in their study that Vein of Labbe occlusion or injury in a poorly anastomosed superficial cortical venous system may lead to temporal lobe swelling, brainstem compression and a posterior hemorrhagic infarct [12]. In present case, the altered sensorium, low GCS and right ear bleeding was well explained by longitudinal fracture of right squamous temporal bone and temporal lobe edema as a consequence of traumatic injury of brain.

Boukobza M., *et al.* stated that Labbe vein thrombosis is a rare phenomena that usually occurs along with extensive dural venous

sinus thrombosis and evidenced a strong association between the extent of thrombosis in the transverse sinus and the presence of Labbe vein thrombosis [13].

Acute development of Vein of Labbe thrombosis and hemorrhagic infarct will be treated with therapeutic dose of LMWH followed by prophylactic dose of LMWH till postpartum period. In our case, since patient had normal coagulation profile and clinical improvement with conservative management, LMWH was not administered. We followed up the patient after discharge from hospital till her delivery and postpartum period to know if she developed a second event but failed to find any. This may explain that her hemorrhagic infarct and vein of Labbe thrombosis were secondary to trauma induced activation of coagulation system and not due to hypercoagulable state of pregnancy in view of normal coagulation profile.

Conclusion

CVT is a common complication in pregnancy due to hyper- coagulable state but as site specific incidence is concerned, vein of Labbe thrombosis is a rare complication. The underlying thrombotic event may be missed out when patient presents with history of trauma and bleeding. This may mislead diagnostic and therapeutic intervention within time. Judicious administration of antithrombotic agents is another area of challenging decision and we experienced that CVT can be managed with appropriate selection of therapeutic agents and antithrombotic agents are not always required. The learning points from this case are

- Always explore thrombotic events among pregnant patients presenting with headache, vomiting and altered sensorium with history of trauma in spite of normal coagulation and routine parameters
- Sometimes traumatic bleeding may mislead the diagnosis but CT and MRV will be of definite help.
- Vein of Labbe thrombosis is a rare but important cause of hemorrhagic infarct that require immediate attention and treatment to improve prognosis.
- All cases of CVT may not require antithrombotic agents as first line treatment of choice.

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