

Complication case

Bleeding Complication of Pial Arteriovenous Fistula with HHT after Embolization and Post Procedure Anticoagulation

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Purpose

Pial arteriovenous fistulas (AVF) are rare disease but more common occur in patient with hereditary hemorrhagic telangiectasia (HHT). Endovascular coiling has been reported as an effective treatment. However this method harbors complication of venous thrombosis and intracranial hemorrhage developing after endovascular occlusion of pial AVF.. This is a case report of bleeding complication after endovascular coiling and anticoagulant administration of patient with pial AVF related to HHT. Administering anticoagulant after the procedure to prevent venous thrombosis due to stasis is still need to be studied further

Case Report

A 24 year old male presented with right middle cerebral artery AVF with giant venous pouch. Patient had history of recurrent seizures and progressive declining of cognitive function. He also had recurrent epistaxis since he was 10 year old until in his 20s with episodes of fatigues and dispneu. His father also has recurrent epistaxis until in his 60s without any known co-morbidities. The pial AVF was embolized by coils and patient was given low molecular weight heparin (subcutaneous injection, 0.4 mL/12 hours) after procedure. At day 3, patient got seizure and severe headache and hemorrhage was detected oozing from the site of the fistula by CTA. Patient was immediately referred to neurosurgeon to evacuate the hematoma as well as the giant venous pouch. Patient regained his consciousness with left hemiparesis after the surgery. But 2 weeks later he developed difficulty breathing due to hemothorax and patient passed away due to acute respiratory distress.

Discussion/ Result

Bleeding complication post embolization of this pial AVF fistula in our opinion could be related to 1) the using of anticoagulation since It had shown on CT with contrast injection that source of the bleeding was oozing from the site of coiled fistula, 2) HHT triggered bleeding, we diagnosis this patient has HHT based on Curacao 4 criterias (1) spontaneous epistaxis, (2) mucocutaneous telangiectasia on skin or mucosa, (3) visceral AVMs (brain, lung, liver etc.), and (4) diagnosis of HHT in a first degree relative using the same criteria in this case is his father. Patient who meet ≥ 3 of the 4 criteria are labeled as definite HHT. Approximately 10-20% of patients with HHT mostly HHT1 are affected by cerebral AVM. Pial AVFs are thought to be an exceedingly rare in a sporadic AVM population, but are seen in $\sim 10\%$ of HHT AVM patients, thus the presence of pial AV fistula should triggered an investigation for HHT. Small studies on natural history of superficial single-hole pial AVFs in patients with HHT demonstrated high rates of neurological deficits and hemorrhage. On

study of 8 patients receiving conservative management of pial AVFs demonstrated mortality in 63% of patients because of bleeding of the AVFs. This patient despite bleeding at the site of brain AV fistula also experienced hemorrhagic complication hemothorax which was possibly related to bleeding of unknown existence of pulmonary AVM.

Conclusion

Embolization of high-flow AVF fistula with giant venous pouch could result in venous stasis and thrombosis, but administration of anticoagulant especially in pial AV fistula related to HHT need some further study. This case report has some remaining questions (1)Is to embolize this patient with HHT was a correct decision? (2)when is the right time to administer anticoagulant for pial AV fistula embolization with enlargement of venous system? (2) Is it safe to administer anticoagulant to patient with HHT?

Keywords: anticoagulant, bleeding complication, endovascular coiling, HHT, pial AV fistula