Case Report

A rare case of sulcal hemorrhage after ischemic stroke – A case report

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Abstract

Atraumatic Non-aneurysmal sulcal subarachnoid hemorrhage is very rare. Sulcal subarachnoid hemorrhage (sSAH) is characterized by isolated bleeding in one or a few adjacent sulci. Central sulcus hemorrhage is a rare imaging finding. There are many causes for sSAH. In older patients, sSAH is due to Cerebral Amyloid Angiopathy (CAA), while in younger patients, reversible cerebral vasoconstriction syndrome (RCVS) is the most frequent etiology. Imaging studies help in the evaluation of sSAH. We report a rare case of an isolated central sulcus hemorrhage on computed tomography. sSAH usually occur on the side with acute ischemic stroke, and it is unusual for sSAH to occur on the opposite side of the infarct territory, but in our case sSAH occurred on opposite side, but after a gap of 3 years.

Key words

Sulcal Subarachnoid Hemorrhage (sSAH), Cerebral Amyloid Angiopathy (CAA), Reversible Cerebral Vasoconstriction Syndrome (RCVS), Ischemic Stroke.

Introduction

Intracranial hemorrhage could be located within the deep white matter, cortical/ subcortical, subarachnoid, subdural, epidural, or intraventricular locations. The most common etiologies for intracerebral bleeding include trauma and hypertension. Cerebral amyloid angiopathy (CAA) is a common cause of non-traumatic peripheral intracerebral hemorrhage in a normotensive patient, and may present as an isolated cortical or subcortical hemorrhage. Imaging studies help in the evaluation of nonspecific clinical features and helps to know the etiology of the hemorrhage and associated complications. Prompt diagnosis of acute as well

as chronic presentations of cerebral amyloidosis is important and need an appropriate management and care [1]. Atraumatic and non-aneurysmal sulcal subarachnoid hemorrhage (sSAH) is a rare type of intracerebral hemorrhage with various etiologies [2]. The main clinical symptoms at presentation usually are focal and transient neurological deficit and thunderclap headache. A few patients have progressive headache and partial or generalized epileptic seizures.

MRI abnormalities associated with sSAH reported are prior hemorrhages, microbleeds, severe leukoencephalopathy and hemosiderosis suggesting CAA, vasogenic edema in parieto-occipital areas compatible with a Posterior Reversible Encephalopathy Syndrome (PRES), cortical venous thrombosis and concomitant acute cortical stroke, reversible cerebral vasoconstriction syndrome (RCVS) vascular malformation, and over anticoagulation. sSAH is a rare condition related to a wide spectrum of etiologies, including covid-19 infection [3]. Brain MRI magnetic resonance angiography and eventually digital subtraction angiography helped in the identification of an underlying etiology. CAA, reversible cerebral vasoconstriction syndrome (RCVS) and Posterior Reversible Encephalopathy Syndrome (PRES) represented more than 50% of the etiological mechanisms. Among older patients, sSAH was mainly related to CAA while in younger patients, RCVS represented the most frequent etiology [4].

Case report

A 45-year-old male was admitted with complaints of diffuse headache of 1 day duration and developed 2 episodes of seizures after 6 hours. He had 2 vomiting. Headache was sudden in onset and gradually progressive associated with vomiting; there were no aggravating or relieving factors. Seizures were sudden in onset and were generalized type of seizures, frothing of mouth and, up-rolling of eyes was present, deviation of mouth to the right side was seen during seizures and was associated with bedwetting, regained consciousness after 45 minutes of seizure attack. No history of muscle weakness.

Past history: Patient had CVA-cerebral thrombosis, left MCA territory with right hemiparesis 3 years ago. Patient was a known case of Hypertension since 4 years on Atenolol 25 mg. No history of any thyroid disorders, DM, TB, epilepsy, COPD.

Personal history: Patient was a chronic alcoholic, chronic smoker since 10 years, no significant family history.

General examination: Patient was conscious, coherent, co-operative, normal built and well nourished. No Pallor, Icterus, Cyanosis, Clubbing, Koilonychias, Lymphadenopathy. No pedal edema.

Vitals: Temp- afebrile, BP=140/90 mm of Hg in sitting position in right arm. Pulse=80/min regular in rhythm with no radio-radial or radio-femoral delay.

CNS examination:

Intellectual functions: Patient was conscious/coherent/co-operative, oriented to time, place, person, right-handed. Memory - Immediate, recent, remote all were normal. Normal concentration and intelligence. Spontaneous speech, good comprehension. No aphasia. No hallucinations/delusions.

Cranial nerves: Normal.

Motor system

Bulk, Tone, power – were normal in all the 4 limbs

Deep tendon reflexes were normal in all the 4 limbs

Plantars were extensor in left foot, flexor in the right foot.

Pes cavus was present. No abnormal movements seen.

Cerebellar functions: No titubation, no nystagmus, no pendular knee jerk, no dysdiadokinesia, no past pointing, finger nose test was normal on both sides. Gait - normal.

Sensory system – all modalities of sensations were normal

Peripheral nerves were not thickened
Skull and spine were Normal
CVS examination: S1, S2 heard no, murmurs
Abdomen examination: Abdomen was soft and not distended, non-tender, no organomegaly
Respiratory system examination: Bilateral normal vesicular breath sounds were present with no adventitious sounds.

Investigations: Hb - 16.4 g%, RBC count – 4.1mill/cu mm, WBC count - 6700 cells/cu mm, N-68%, L- 24%, M - 4%, E - 4%, B - 0. Platelet count –2.0 lakh/cu mm. LFT-total bilirubin - 0.9 mg/dl, SGOT-39 U/L, SGPT – 41 U/L, ALP – 106 U/L, Total protein - 6.1 g/dl, Albumin - 3.1 g/dl, Globulin – 3.0 g/dl. Serum Electrolytes: Na – 140 mmol/L, K - 4.0 mmol/L, Cl – 109 mmol/L. RBS – 83 mg/dl. Blood urea – 30 mg/dl. Serum creatinine - 1.0 mg/dl. BT – 3 mins 20 secs, CT – 8 minutes, PT – 12 secs. RTPCR – Negative; NECT Brain (**Figure - 1**) shows Acute sulcal bleed measuring 23 x 15 mm noted involving right parietal lobe white matter, no mass effect, no midline shift noted. Chronic infarcts were noted involving right external capsule and anterior limb of left internal capsule. Chronic lacunar infarcts were noted involving bilateral thalami, posterior limb of left internal capsule. Chronic small vessel ischemic changes were noted involving bilateral periventricular white matter. Carotid Doppler - Thin linear hypoechoic plaque in left CCA, normal bilateral carotid doppler indices with no significant luminal narrowing. IMT – right CCA – 0.5 mm, left CCA – 0.6 mm. Chest X-ray PA view- normal. ECG - Normal.

Diagnosis: CVA - Right sulcal subarachnoid hemorrhage in parietal lobe, left hemiparesis, old CVA- left MCA territory, right hemiparesis (improved), hypertension.

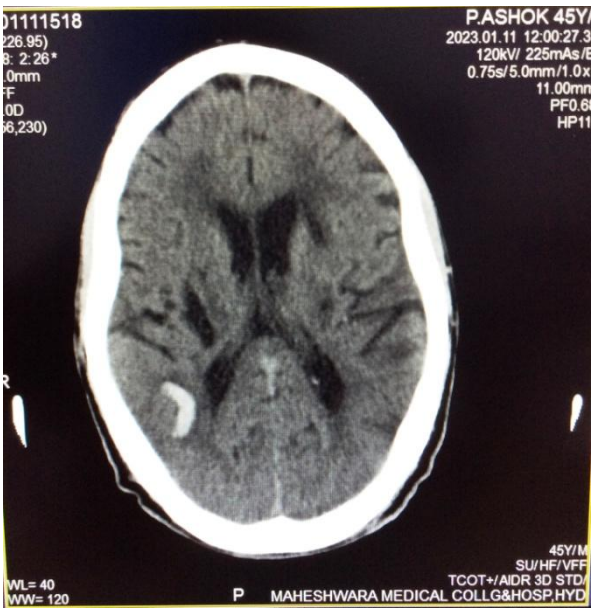
Treatment given

1. Tab. Nimodipine – 30 mg – 2 Tablets - TID
2. Inj. Optineuron - 1AMP - OD
3. Tab. Nicardia retard – 10 mg - TID
4. Physiotherapy

Discussion

sSAH is a rare condition related to a wide spectrum of etiologies, including COVID – 19 infection [3]. Various etiologies can present with intracranial hemorrhage, especially within the cortical or subcortical areas on imaging [5]. Brain MRI magnetic resonance angiography and digital subtraction angiography helped in the diagnosis. CAA, reversible cerebral vasoconstriction syndrome (RCVS) and Posterior Reversible Encephalopathy Syndrome (PRES) represented more than 50% of the etiological causes. Among older patients, sSAH was mainly related to CAA while in younger patients, RCVS represented the most frequent etiology [4].

Figure – 1: NECT Brain shows Acute sulcal bleed measuring 23 x 15mm noted involving right parietal lobe white matter, no mass effect, no midline shift noted. Chronic infarcts were noted involving right external capsule and anterior limb of left internal capsule. Chronic lacunar infarcts were noted involving bilateral thalami, posterior limb of left internal capsule. Chronic small vessel ischemic changes were noted involving bilateral periventricular white matter.



Kumar S, et al. and Singhal AB, et al. reported a clinical association and causes of cSAH in the largest mixed pathogenesis case series [6, 7]. cSAH represents significant SAH subcategory

(6% versus 7% reported previously) [7]. CAA is the commonest cause in patients >60 years old, and RCVS in those <60 years old. Presentations are distinct, the former with slowly evolving transient focal neurological symptoms (TFNS), the latter usually with thunderclap headache (but sometimes rapid onset focal neurological deficits or seizures). In contrast with the series of Kumar, et al. [6]; Ashan Khurram, et al. [2] in their series the commonest presenting complaint was TFNS (66%) not headache (62%), perhaps reflecting a lower RCVS proportion and higher median age. In addition to CAA and RCVS, their study also documented multiple cases with posterior reversible encephalopathy syndrome, cerebral venous sinus thrombosis, and parent vessel stenosis. Most patients presenting with TFNS were >60 years old. Sixteen patients had probable CAA and 6 patients were possible CAA. The central sulcus seems to be an area particularly prone to cSAH-induced TFNS. TFNS are commonly misdiagnosed as transient ischemic attacks [8].

Ashan Khurram, et al. [2] in their study demonstrated a minimum population incidence of 5.1 per million/year. This is an underestimated because of many limitations. An ideal unbiased cSAH incidence study would require comprehensive, prospective, expert, blinded neuroimaging review of a large heavily investigated population during several years. They concluded that, non-traumatic cSAH occurs relatively commonly. CAA and RCVS are most frequent causes. Increased awareness will prevent misdiagnosis as transient ischemic attack (or seizures) and prevent potentially harmful treatment [9].

The pattern of bleeding in CAA is peripheral, cortical or subcortical, micro-and macro-hemorrhages with sparing of the deep white matter, basal ganglia, and thalami. Cerebellar and intraventricular involvement is rare. The common etiologies for hemorrhage in a non-traumatic setting are usually related to hypertension and aneurysms. Hypertensive hemorrhages occur centrally within the thalamus and basal ganglia.

Cerebral aneurysms are more common with a family history and present with rupture and bleeding, usually within the vicinity of the aneurysm. Trauma related hemorrhages are usually seen within the inferior frontal and temporal regions that are prone to contusion and are associated with epidural or subdural hematomas in the region of coup and contre coup injuries. Vascular malformations present as isolated cortical or extra-axial hemorrhages. CT or MR angiograms help in characterization of possible causes of peripheral intracranial hemorrhages such as aneurysms and vascular malformations.

Cerebral amyloidosis is one of the etiologies for spontaneous non-traumatic cortical or subcortical bleed in a normotensive patient. Amyloid deposition in the brain occurs within various pathologies such as Alzheimer's dementia, Creutzfeldt Jacobs's disease, spongiform encephalopathies, and postradiation necrosis or can be rarely hereditary [9]. Deposition of amyloid within the cortical, subcortical, and leptomeningeal cerebral vessels results in increased fragility, hemorrhages, microaneurysms, and vascular irregularity or stenosis [10, 11]. Cerebral amyloidosis can be asymptomatic or clinically present with symptoms related to acute or chronic hemorrhage and ischemia. Cerebral amyloidosis commonly presents as peripheral cortical or subcortical hemorrhage but other rare and nonspecific patterns have also been described [12, 13, 14]. The finding of a central sulcus hemorrhage is rare, but it is mostly associated with amyloid angiopathy [15, 16].

An acute hemorrhage can be identified easily on non-contrast CT imaging. MR imaging, especially FLAIR (fluid attenuation inversion recovery) and gradient echo (GRE) or susceptibility weighted imaging (SWI) are helpful for confirmation and characterization of smaller cortical or subcortical bleeds. It also

helps to identify other areas of chronic involvement. Boston criteria provide integration and standardization of clinical and imaging findings with diagnostic categorization (definite, probable with pathological evidence, and probable and possible diagnoses) based on clinical, histopathological, and MRI findings [17].

A biopsy is not usually warranted, and if obtained, staining of the amyloid with Congo red under polarized light demonstrates characteristic yellow green birefringence. Radiological work up in an elderly non-traumatic normotensive patient with cortical or subcortical hemorrhage excludes other causes of hemorrhage. Imaging in conjunction with clinical presentation establishes a putative diagnosis of cerebral amyloidosis. Management is currently limited and relies on optimal control of blood pressure, use of corticosteroids, and avoidance of antiplatelet agents and Warfarin [18]. Further understanding of the molecular pathogenesis would help in understanding the role of immunosuppressants and lipid lowering drugs and in turn help in the development of disease modifying drugs [11].

Acute ischemic stroke co-occurring with sSAH has been infrequently reported. Nearly all cases of sSAH have been described to occur on the side with acute ischemic stroke, and it is unusual for sSAH to occur on the opposite side of the infarct territory [19]. But in our case ischemic stroke on opposite side, preceded sSAH by 3 years.

Hemorrhagic cerebrovascular events, either due to aneurysmal rupture or spontaneous subarachnoid hemorrhage (SAH), are not rare in COVID-19. Several mechanisms such as coagulopathy, cytokine storm, viral endotheliopathy, hypertension, and immune modulation might play a role in the pathogenesis of SAH in COVID-19 [3]. Several reports of SAH in COVID-19 patients are present in the literature [20, 21, 22 , 23]. But in our case the RTPCR test was negative

SARS-CoV-2 may gain access to CNS via two principal routes, namely the hematogenous and the retrograde neuronal (axonal) dissemination. In the hematogenous entry, SARS-CoV-2 may enter the CNS through the blood-brain barrier (BBB) by any of the three mechanisms-

- by direct entry into CNS via infection of the endothelial cells,
- via angiotensin-converting enzyme-2 (ACE-2) receptors expressed on the endothelium, or
- via migration by infecting the monocytes and macrophages.In the neuronal entry, SARS-CoV-2 enters the CNS via the olfactory neurons through the nose [20, 24, 25, 26, 27].

The exact pathogenetic mechanisms underlying spontaneous SAH in COVID-19 are not well understood. However, evidence points towards an inflammatory and hyper coagulative state, facilitated by SARS-CoV-2 induced endothelitis and microvascular dysfunction. Indeed, COVID-19 infection may cause injury to the vascular endothelium, making them vulnerable to rupture, especially at a pre-existing susceptible site (e.g. aneurysm) [28].

Conclusion

Atraumatic Non-aneurysmal sulcal subarachnoid hemorrhage is very rare. There are many causes for sSAH. In older patients, sSAH is due to CAA while in younger patients, reversible cerebral vasoconstriction syndrome (RCVS) is the most frequent etiology. Imaging studies help in the evaluation of sSAH. We herewith present a rare case of an isolated central sulcus hemorrhage which is very rare. Acute ischemic stroke co-occurring with sSAH has been infrequently reported. Nearly all cases of sSAH have been described to occur on the side with acute ischemic stroke, and it is unusual for sSAH to occur on the opposite side of the infarct territory. But in our case ischemic stroke on opposite side,

preceded sSAH by 3 years. There are many reports of SAH occurring in COVID-19 patients.

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