



Nontraumatic spontaneous bilateral intracerebral haemorrhage in a young patient: a rare case report

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Introduction and importance: Spontaneous bilateral intracerebral haemorrhage is a rare surgical occurrence, especially in young populations with poor prognosis. Hypertension is the leading cause but vascular malformations, infections and rare genetic conditions are also responsible.

Case presentation: Twenty-three-year-old male with no prior comorbidities presented to emergency with sudden onset loss of consciousness and 1 episode of seizure. No history of intoxication or trauma was given. Glasgow Coma Scale at presentation was E1V2M2. CT scan head revealed bilateral basal ganglia haematoma along intraventricular haemorrhage.

Clinical discussion: The patient was managed conservatively in the Neurosurgical Intensive care unit. Supportive management was provided. The patient's motor response was improving and a repeat CT scan showed a resolving haematoma. However, due to poor economic conditions, the patient party left against medical advice.

Conclusion: Spontaneous bilateral basal ganglia haemorrhage is a rare surgical emergency with no clear consensus on a management approach. This case highlights the importance of undiagnosed hypertension in causing intracerebral haemorrhage in poor economic groups.

Keywords: bilateral basal ganglia haemorrhage, case report, conservative management

Introduction

Spontaneous simultaneous basal ganglia haemorrhage (SSBGH) is a rare subtype of intracerebral haemorrhage with only a few cases reported worldwide^[1,2] It has a high mortality of about 33%^[1]. Hypertension (HTN) is the leading cause owing to the formation of microaneurysm (Charcot–Bouchard aneurysm) of lateral lenticulostriate arteries in older (> 50 years) patients with longstanding HTN^[3]. However, other etiologies also play a role in pathogenesis, especially in a young population such as metabolic etiologies, infection, vascular malformations, intoxication and so on^[4–8]. In light of the recent pandemic, few cases have been reported of covid-19 encephalitis with intracerebral haemorrhages^[9,10]. The patients usually present with features such as altered sensorium, headache and focal neurological

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HIGHLIGHTS

- Spontaneous bilateral intracerebral haemorrhage is rare surgical emergency with poor prognosis.
- Subtle behaviour changes may be only indicator in some cases.
- Recent advances such as stereotactic evacuation have provided promising results
- Due to its rarity, the therapeutic management remains a challenge.

deficit. CT scan is the imaging of choice in the acute setting with findings usually being hyperdense lesions in the basal ganglia region. This case report has been prepared in accordance with the SCARE 2020 guidelines^[11].

Case presentation

A 23-year-old man presented to our hospital emergency department due to sudden onset loss of consciousness (LOC) followed by abnormal body movement in the form of generalized tonic-clonic type, witnessed by friend 30 min before the initial evaluation. He had no other past medical or surgical history. He had no family history of diabetes; coagulopathy or family history of any neurological disease or cancer and no known psychosocial comorbidities. He was not taking any medication for any illness at the time of the onset of LOC. Social history was significant for smoking two cigarettes a day for 5 years. On examination, he had a Glasgow Coma Scale of 5 (eye-opening -2, verbal response -1, motor response -2). Cranial nerves II–XII were grossly intact, yet horizontal gaze was noted to be mildly abnormal. Pupils were

dilated, equal, round and mildly reactive to light. Deep tendon reflexes (DTRs) 2+/4+ in upper and lower extremities bilaterally and plantar reflex were B/L upgoing. The heart was found to be in sinus bradycardia. No heart murmur was noted, and blood pressure was 160/90 mm of hg.

The patient was intubated and put on mechanical ventilation. A plain CT scan revealed basal ganglia bleeding as manifested in hyperdense lesions. The bleed extended into the third ventricles, aqueduct and fourth ventricles Figures 1–4. The patient was shifted to the neurosurgical ICU and decongestants in the form of frusemide and 20% mannitol were given. Intravenous Antibiotics and IV Levetiracetam were also given. He was continued on ventilatory support, and subsequently tracheostomized on day 6. Nasogastric feed was also started gradually. His motor score gradually improved to localized response to pain. (M5) by the fourth day of injury. Supportive care was given. A subsequent scan after 3 days showed that the haemorrhage was resolving. All blood and radiological parameters including the coagulation profile were within normal limits. The patient was improving but because of his poor financial condition, patient party took the patient home against the medical advice.

Discussion

Intracerebral haemorrhage (ICH) is defined by the American Stroke Association (ASA) as rapidly developing clinical signs of neurological dysfunction attributable to a focal collection of blood within the brain parenchyma or ventricular system that is not caused by trauma. Intracerebral haemorrhage is the second most common cause of stroke second only to cerebral infarction and has a high case of fatality. Since 1980 knowledge of the epidemiology of stroke has increased with the increasing availability of brain-imaging techniques. Usually, the haemorrhage is solitary and unilateral. Unilateral intracerebral haemorrhage is a relatively

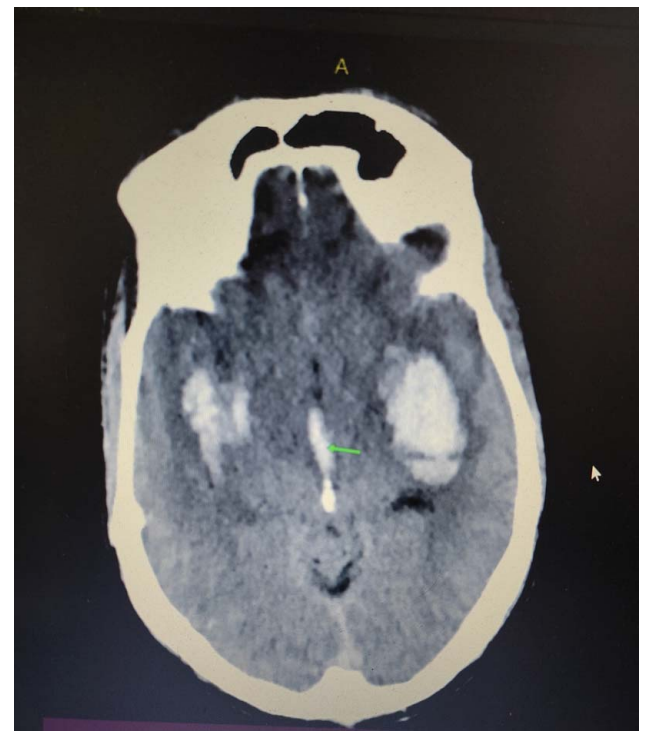


Figure 2. Extension of bleed into the third ventricle.

common occurrence, with an incidence of 24.6 per 100 000 person-year; however, a spontaneous bilateral ICH is exceedingly rare, with only 30–40 reported cases. However, the prognosis and

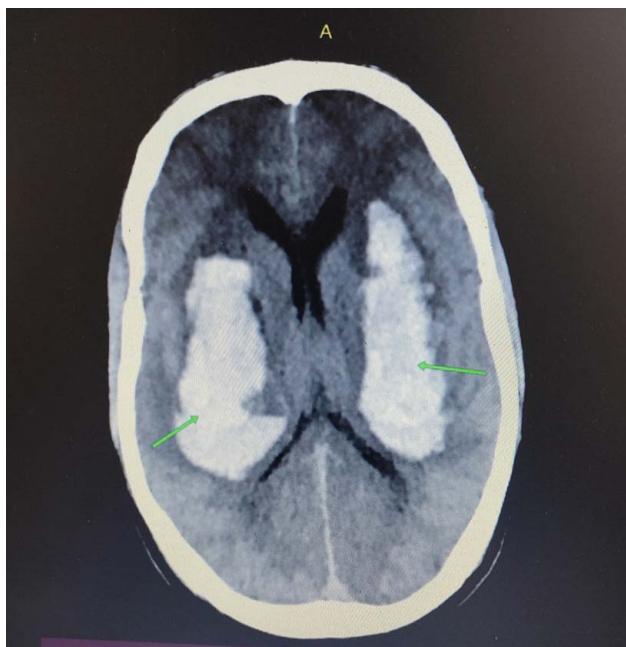


Figure 1. Bilateral basal ganglia haematoma.

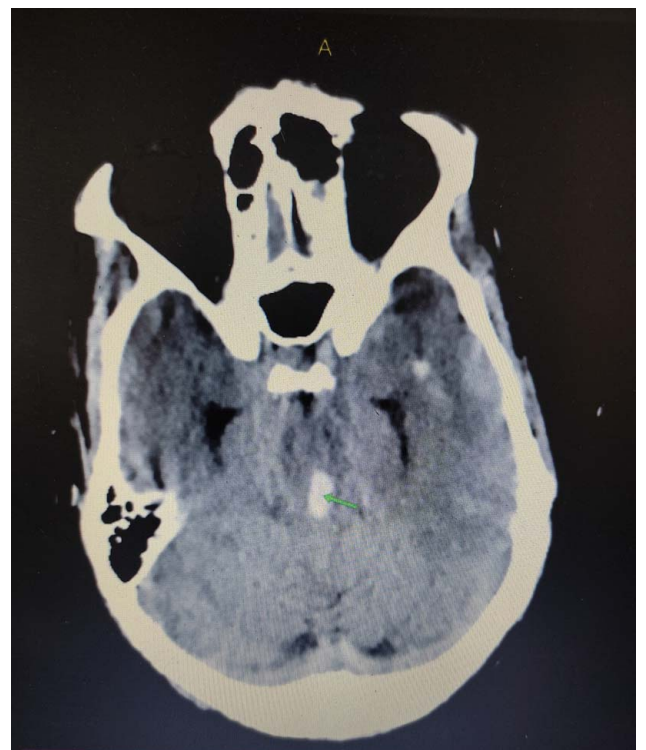


Figure 3. Bleed into the aqueduct.



Figure 4. Bleed into the fourth ventricle.

recovery are similar as compared to unilateral. The prevalence is more common in the Asian population as compared to the western population. Undiagnosed HTN along with lack of treatment among those diagnosed with HTN may be a contributing factor in poor economic regions of Asia.

The main contributors to spontaneous ICH are coagulopathies, vasculopathy, tumour masses, HTN, and tumour mass. Both unilateral and bilateral intracerebral haemorrhage can be brought on by risk factors such as a history of stroke, HTN, and advanced age^[12]. Cerebral amyloid angiopathy, which puts patients at risk for numerous spontaneous intracerebral haemorrhages, is part of the extended differential diagnosis for bilateral disorders of the basal ganglia and thalamus. Poisoning, metabolic, vascular, degenerative, inflammatory, viral, and neoplastic illnesses are other even more uncommon differential diagnosis categories^[13]. Our patient presented with no identifiable risk factors or past history that could direct us to an aetiology. This may be because lack of basic healthcare access leads to many hypertensive and other chronic conditions being undiagnosed. Another possibility may be the lack of awareness among the general population leading to an unenthusiastic attitude towards regular health checkup^[14].

The exact mechanism by which SSBGH occurs is not identified yet and various hypothesis have been postulated. One probable pathway is that the simultaneous rupture of bilateral microaneurysms on lenticulostriate arteries occurs by chance. Another more convincing explanation is that the initial haemorrhage causes specific hemodynamic conditions, like a reflex increase in blood pressure, which prompt the rupture of a second microaneurysm on the contralateral side in a short period of time^[15]. Basal ganglia are highly active metabolically and therefore intoxication with various poisons, metabolic disturbances, and neurodegeneration with brain iron accumulation all have symmetrical effects on the basal ganglia. Other systemic or metabolic diseases, degenerative diseases, and

vascular disorders can also have an impact on the basal ganglia and thalamus. Both deep grey matter structures may be affected by focal flavivirus infections, toxoplasmosis, and primary central nervous system lymphoma^[13]. Our patient gave a history of smoking which in itself is a contributory risk factor for HTN.

Our patient presented with LOC and abnormal body movements and it is consistent with other cases of SSBGH. Seizure is a relatively rare occurrence (6%) and there are no studies regarding whether the presence of a seizure implies poor prognosis or not^[11]. Sometimes patient may also present with subtle behavioural changes only with no apparent focal neurological deficit. Therefore, it is prudent for clinicians to consider spontaneous ICH in patients with mental status changes^[1,16]. The CT imaging of our patient revealed extensive haematoma extending up to the fourth ventricle. It is likely to be a precipitating factor in seizure and LOC in our patient.

There is a lack of consensus about the best approach towards the management of SSBGH. Although conservative management was the mainstay of the treatment of patients of SSBGH in most reported studies, there was a poor prognosis and recovery. Of the previously reported cases of multiple spontaneous simultaneous ICH, 10.5% died, 10.5% were considered severely disabled, 2.6% were left in a vegetative state, 10.5% walked with a cane, and 5.2% had a good recovery^[12]. Recent advances such as stereotactic evacuation are found to have reduced odds ratio for death and possible improvement in independent survival as demonstrated in a recent systematic review and meta-analysis^[17]. Comparing solitary ICH to simultaneous bilateral basal ganglia haematoma, almost all patients had worse results. Damage to the crossing and non-crossing fibres, the bilateral diaschisis phenomena, severely impaired consciousness, quadriparesis, and pseudobulbar palsy are the main causes. Statistical analysis revealed prognostic markers, including the admission Glasgow Coma Scale score, haematoma distribution, and overall haematoma volume. In other studies, the size of the haematoma was also taken into account as a predictive factor^[3,18]. Our patient was managed on a conservative approach, however, due to poor economic condition patient requested discharge against medical advice. Therefore, we were unable to assess whether the patient improved further as the patient is unlikely to come for regular follow-ups.

In light of the SARS-CoV-2 worldwide pandemic, it has recently been hypothesized that the COVID-19 infection can be neurotropic via the trans cribriform or the hematogenous pathway^[19]. It is still unclear why COVID-19 would increase the risk of bilateral ganglia haemorrhage. Despite this, the virus has been shown to predispose to a vulnerable, hypercoagulable condition, which increases the risk of ischaemic stroke and cerebrovascular catastrophe^[20]. It is hypothesized that the neurological impairments caused by bilateral basal ganglia haemorrhage and the etiopathology of COVID-19, whether through direct invasion or systemic inflammatory responses, may be caused^[10].

Conclusion

Bilateral basal ganglia bleeding is a rare occurrence. The most frequent risk factors are trauma and HTN. Various hypotheses have been proposed by different authorities, but the precise pathophysiological architecture that led to its genesis is still up for controversy. The most frequent risk factors for spontaneous bilateral ICH include advanced age, a history of HTN, and a past

stroke, but further testing is required to rule out further uncommon disease processes. Although a brain CT scan is the best initial test in an emergency, an immediate MRI scan should be done to fully describe the lesions for a conclusive diagnosis. Recent COVID-19 pandemics may have added another risk factor for Intracerebral haemorrhages. Although there is no consensus regarding the management of SSBGH, recent advances such as stereotactic evacuation of haematoma have resulted in a good prognosis Figures 1–4.

Ethical approval

NA.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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None.

Author contribution

P.S.Y., S.P., R.A., A.A., U.B., B.K.: involved in literature review, manuscript writing and proof reading. A.N.: corresponding author, involved in literature review, manuscript writing and proof reading. M.U., L.S.: involved in patient care, literature review, manuscript writing and proof reading. R.B.: involved in imaging interpretation and proof reading.

Conflicts of interest disclosure

The authors declared no relevant financial conflict or any other conflict of interest.

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References

- [1] Alhashim A, Hadhiah K, Al-Dandan H, *et al.* Spontaneous Simultaneous Bilateral Basal Ganglia Hemorrhage (SSBBGH): Systematic Review and Data Analysis on Epidemiology, Clinical Feature, Location of Bleeding, Etiology, Therapeutic Intervention and Outcome. *Vasc Health Risk Manag* 2022;18:267–76.
- [2] van Asch CJ, Luitse MJ, Rinkel GJ, *et al.* Incidence, case fatality, and functional outcome of intracerebral haemorrhage over time, according to age, sex, and ethnic origin: a systematic review and meta-analysis. *Lancet Neurol* 2010;9:167–76.
- [3] Takeuchi S, Takasato Y, Masaoka H, *et al.* Simultaneous multiple hypertensive intracranial hemorrhages. *J Clin Neurosci* 2011;18:1215–8.
- [4] McGee SM, McGee DN, McGee MB. Spontaneous intracerebral hemorrhage related to methamphetamine abuse: autopsy findings and clinical correlation. *Am J Forensic Med Pathol* 2004;25:334–7.
- [5] Ertl-Wagner B, Jansen O, Schwab S, *et al.* Bilateral basal ganglion haemorrhage in diabetic ketoacidotic coma: case report. *Neuroradiology* 1999;41:670–3.
- [6] Zinnanti WJ, Lazovic J, Housman C, *et al.* Mechanism of metabolic stroke and spontaneous cerebral hemorrhage in glutaric aciduria type I. *Acta Neuropathol Commun* 2014;2:1–15.
- [7] Kalbhenn T, Neumann LM, Lanksch WR, *et al.* Spontaneous intracerebral hemorrhage and multiple infarction in Williams-Beuren syndrome. *Pediatr Neurosurg* 2003;39:335–8.
- [8] Jang BH, Son SW, Kim CR. Fahr's disease with intracerebral hemorrhage at the uncommon location: a case report. *Ann Rehabil Med* 2019;43:230–3.
- [9] Khattar NK, Sharma M, McCallum AP, *et al.* Intracranial hemorrhage in a young COVID-19 patient. *Interdiscipl Neurosurg Adv Tech Case Manage* 2020;22:100878.
- [10] Daci R, Kennelly M, Ferris A, *et al.* Bilateral basal ganglia hemorrhage in a patient with confirmed COVID-19. *AJNR Am J Neuroradiol* 2020;41:1797–9.
- [11] Agha RA, Franchi T, Sohrabi C, *et al.* The SCARE 2020 Guideline: Updating Consensus Surgical Case Report (SCARE) Guidelines. *Int J Surg* 2020;84:226–30.
- [12] Seo J-S, Nam T-K, Kwon J-T, *et al.* Multiple spontaneous simultaneous intracerebral hemorrhages. *J Cerebrovasc Endovasc Neurosurg* 2014;16:104–11.
- [13] Hegde AN, Mohan S, Lath N, *et al.* Differential diagnosis for bilateral abnormalities of the basal ganglia and thalamus. *Radiogr Am J Roentgenol* 2011;31:5–30.
- [14] Aryal KK, Mehata S, Neupane S, *et al.* The burden and determinants of non-communicable diseases risk factors in Nepal: findings from a nationwide STEPS survey. *PLoS One* 2015;10:e0134834.
- [15] Kono K, Terada T. Simultaneous bilateral hypertensive putaminal or thalamic hemorrhage: Case report and systematic review of the literature. *Turk Neurosurg* 2014;24:434–7.
- [16] Shaheed TA, Glover N, Alboiny S. Nontraumatic spontaneous bilateral basal ganglia hemorrhage: a rare case report. *Cureus* 2020;12:10–3.
- [17] Akhigbe T, Okafor U, Sattar T, *et al.* Stereotactic-guided evacuation of spontaneous supratentorial intracerebral hemorrhage: systematic review and meta-analysis. *World Neurosurg* 2015;84:451–60.
- [18] Fogelholm R, Murros K, Rissanen A, *et al.* Long term survival after primary intracerebral haemorrhage: a retrospective population based study. *J Neurol Neurosurg Psychiatry* 2005;76:1534 LP–1538.
- [19] Baig AM, Khaleeq A, Ali U, *et al.* Evidence of the COVID-19 virus targeting the CNS: tissue distribution, host-virus interaction, and proposed neurotropic mechanisms. *ACS Chem Neurosci* 2020;11:995–8.
- [20] Goldberg MF, Goldberg MF, Cerejo R, *et al.* Cerebrovascular disease in COVID-19. *Am J Neuroradiol* 2020;41:1170 LP–1172.