Case Reports & Case Series (CRP)

Spontaneous subdural hematoma associated to Duret hemorrhage

William Alves Martins, MD a,⁎, Alice Becker Teixeira, MD a, Thomas More Frigeri, MD b, Eliseu Paglioli, MD, PhD b

a Department of Neurology, Hospital São Lucas, Pontifícia Universidade Católica do Rio Grande do Sul (PUCRS), Porto Alegre, RS, Brazil
b Department of Neurosurgery, Hospital São Lucas, Pontifícia Universidade Católica do Rio Grande do Sul (PUCRS), Porto Alegre, RS, Brazil

ABSTRACT

Subdural hematoma (SH) is a neurosurgical emergency, usually caused by head trauma. Non-traumatic causes include aneurysm or arterial–venous malformation rupture, coagulopathy and others. We report the case of a 66 year-old man who developed apparently unprovoked signs of increased intracranial pressure. Brain computed tomography scan showed an acute spontaneous SH, surgically treated. Throughout surgery, a ruptured cortical artery with intensive bleeding appeared and was cauterized. After surgery, patient remained comatose and a new CT demonstrated Duret hemorrhage at the brainstem. Acute spontaneous SH of arterial origin is rare and highly lethal, in which a good prognosis relies on early diagnosis and treatment.

© 2014 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/3.0/).

Introduction

Acute subdural hematoma (SH) is a common neurosurgical emergency, usually due to rupture of superficial cerebral or cortical bridging veins during severe head trauma. Non-traumatic causes include aneurysm or arterial–venous malformation (AVM) rupture, coagulopathy, cocaine abuse, metastatic cancer, Moya-Moya disease and others [1–4].

Acute spontaneous SH of arterial origin is a rare disorder, caused by disruption of a perisylvian artery without history of head trauma and evidence of AVM or aneurysm [1,2]. Evolution is sudden and dramatic neurological deterioration may be seen within minutes. There is no clear precipitant, but minor head trauma is thought to play a role in rupturing an adhered cortical artery, leading to massive subdural bleeding [3,4]. Prompt neurosurgical evacuation is crucial to a good outcome [5,6]. We report a case spontaneous SH of arterial origin associated to Duret hemorrhage after sudden rise in intracranial pressure and briefly review the current literature.

Case report

A 66-year-old male had been admitted at Hospital São Lucas for complementary investigation of a cholestatic syndrome due to pancreatic cancer. On the 15th day of admission, after coughing, he suddenly developed severe headache, vomiting, right-side weakness and rapidly evolved with altered mental status. At the time, systemic investigation with CT scans of thorax, abdominal and pelvic regions, along with bone scintigraphy, had been negative for metastatic involvement. No history of head trauma was evident and he did not display any prior neurological signs suggestive of metastatic disease. General inspection showed that he was jaundiced and mildly undernourished. Neurological examination showed anisosoria with a dilated (5 mm) left pupil, unresponsive to light. Glasgow coma scale (GCS) was 5/15 and he presented clear right-side weakness. A mild external deviation of the left eye could be seen, suggestive of IIIrd nerve palsy. After orotracheal intubation, brain CT scan showed no evidence of metastatic disease, but uncovered an acute SH in the left frontoparietal region with signs of active bleeding, causing significant midline shift and transtentorial herniation (Fig. A, B).

Laboratory tests were normal, except for mild normochromic anemia and conjugated hyperbilirubinemia. Coagulation studies did not show any abnormalities and he was only on prophylactic heparin, without any history of hemorrhagic diatheses. Platelet count was 318,000/μl, within normal range (150,000–400,000).

Surgery was immediately undertaken, using a left pterional craniotomy to access the hematoma. After opening the Dura mater, we encountered continuous arterial bleeding from a ruptured superficial cortical artery (Video). There was no sign of AVM or aneurism at the site and this artery was easily cauterized, immediately stopping the bleeding. After evacuation of the extensive SH, the patient was admitted to intensive care unit.

One day latter, subsequent CT scan showed almost complete resolution of SH, but displayed mild hemorrhage in the left parahippocampus and midbrain (Fig. 1C, D). Patient remained...
comatose and died nine days after surgical intervention, due to urinary tract septicemia.

Discussion

Acute spontaneous SH of arterial origin had been first described by Munro in 1934, but it was only seen as a unique syndrome by Tallala et al. in 1971 [2–4]. In 1988, Tokoro and colleagues proposed four diagnostic criteria: (a) no history of head trauma, (b) no cerebral cortex damage, (c) no evidence of ruptured aneurysm or AVM, and (d) identification of arterial hemorrhage during operation [1]. Following these criteria, about 91 cases have been reported in literature until now [6]. Our case satisfies Tokoro’s criteria for the diagnosis of acute spontaneous SH of arterial origin and highlights the sudden, acute and dramatic onset, probably induced by an increase in intracranial pressure following Valsalva-maneuver.

The pathophysiological mechanism behind this uncommon condition is controversial, but could involve: a) avulsion of a small artery branching off in a right angle; b) rupture of a corticodural bridging artery or c) adhesions between a cortical artery and the Dura mater [2–4]. Rather than lack of trauma, some authors state that to rupture these arteries would actually take minimum trauma [2]. Our patient’s cough could have generated a small increase in intracranial pressure, just enough to displace brain tissue within the cranium and disrupt an already adhered artery.

Acute spontaneous SH usually manifests as sudden unexplained coma, severe headache, vomiting, focal neurological deficits and seizure [1–4], mimicking stroke or subarachnoid hemorrhage [3]. Surgical evaluation should be urgent and a brain CT scan be performed as soon as possible after clinical stabilization, to guide further therapeutic measures.

Mortality rate ranges from 25% to 90% in reported series and may be associated with a lower GCS at presentation and delay in surgical treatment [3,6]. In a recent review, Coombs and colleagues found 91 cases of acute spontaneous SH of arterial origin reported on literature, corresponding for 61.5% of all acute spontaneous SHs with an overall mortality rate of 36.7% [6]. The mean age was 61 years old and it was twice more frequent in males [2]. Known risk factors include age over 50 years, alcohol abuse and systemic arterial hypertension [2–6]. Spontaneous and traumatic SHs actually share these risk factors. Some authors argue that patients with coagulopathy [3,4] should not receive the diagnosis of spontaneous SH, while others say that coagulopathy is only a contributor to a worse prognosis [2]. Our

Fig. Brain CT scan. A) Extensive SH in left convexity and mass effect before surgery; B) Transtentorial herniation (red arrow) and subdural hematoma in the temporal lobe (asterix); C and D) Post-operative CT scan showing hemorrhagic foci in left parahippocampal gyrus and midbrain (white arrows).
patient had a cholestatic syndrome, however he did not show any signs of coagulopathy or thrombocytopenia. The only anticoagulant drug in use was prophylactic non-fractionated heparin 5000 U twice a day, which probably did not play a role in his presentation.

Sudden development of SH with continuous intrahematomatous bleeding predisposes to rapid temporal lobe herniation and may explain the post-surgical appearance of Duret hemorrhage (DH) in our patient. Duret's phenomenon is actually a secondary hemorrhagic infarction of midbrain [7], resulting from compression and shearing of brainstem penetrating arteries by herniated parahippocampal gyrus [8]. Risk factors for development of DH include fast raising in intracranial pressure, old age, volume of the lesion and abrupt changes in intracranial pressure, such as produced by lumbar puncture or decompressive craniotomy [8,9]. The latter would cause sudden restoration of blood flow and reperfusion injury to ischemic brainstem. Nedergaard and colleagues studied the frequency of DT in acute vascular lesions [9]. In their study, DH developed in 45% in spontaneous cerebral hemorrhage, 36% in ruptured aneurysm, and 15% in ischemic stroke [9]. As several important axonal pathways are damaged in DH, it carries a poor prognosis [7], although some report only mild deficits, such as internuclear ophthalmoplegia [10]. Combination of acute SH of arterial origin and DH is extremely uncommon, despite its quick evolution to transtentorial herniation and has been reported only once [10].

Differential diagnosis in our patient comprises dural metastasis, cerebral venous thrombosis (CVT) and AVM or aneurism as the underlying etiology of SH. Dural metastasis causing or simulating SH is rare [11], mainly seen in patients with advanced malignancy, unlike our patient, who had limited neoplastic disease. Other possible cancer-related complication is CVT [12]. Studies show that 11% of all CVTs might be related to malignancy [13]. Nevertheless, CVT has only rarely been associated to SH [14], both of which are usually caused by intracranial hypotension, not fit for our patient presentation. Likewise, isolated SH is a rare presentation of AVM or aneurism [15,16]. Though our case did not allow assessment of vascular malformation (e.g. MRI and MRA), given the clinical acuity, we feel that visualization of a distal ruptured cortical artery during surgery without morphological anomalies or AVM, supports the diagnosis of acute spontaneous SH of arterial origin. We do not feel that the arterial bleeding was artificial as it was noted instantaneously as an arterial “pumper” through the subdural clot after gentle opening and elevation of the dura mater, as demonstrated in the surgical video.

Acute spontaneous SH of arterial origin is a rare and lethal disorder. Our case highlights the dramatic evolution of this condition, showing that immediate diagnostic approach and intervention are essential to give these patients appropriate care, since early surgical drainage is crucial to a good outcome.

Supplementary data to this article can be found online at http://dx.doi.org/10.1016/j.inat.2014.11.003.

References