CASE REPORT

Lenticulostriate artery aneurysm presenting as primary intraventricular haemorrhage

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SUMMARY

Isolated or primary intraventricular haemorrhage (PIVH) is rare and usually caused by hypertension, vascular malformations, aneurysms and moyamoya disease. Lenticulostriate artery aneurysm (LSA) is also a rare entity, and LSA aneurysm rupture causing primary intraventricular haemorrhage is extremely rare. There are only a few case reports. LSA aneurysms can occur medially or laterally, proximally or distally, though distal lateral LSA aneurysms are more common. They are usually associated with various vascular conditions. We report two rare cases of LSA aneurysms presenting as PIVH, one being medial and other being lateral distal LSA aneurysm. Both were normotensives, and one was associated with arteriovenous malformation.

BACKGROUND

Primary intraventricular haemorrhage (PIVH) is an uncommon type of intracranial haemorrhage accounting for only about 3% of all spontaneous intracranial haemorrhages, which refers to bleeding confined to the ventricular system within the brain without a recognisable parenchymal component. Hypertension is an important risk factor for PIVH, but arteriovenous malformations (AVMs), moyamoya disease and aneurysms are also important causes of PIVH, and yield of diagnostic cerebral angiography in the setting of PIVH is high. However, lenticulostriate artery aneurysms (LSA) which by itself is an uncommon entity, usually presents with intraparenchymal haemorrhages. LSA aneurysms presenting as PIVH are very rare with only few case reports in literature. We present two cases of LSA aneurysm presented with isolated intraventricular haemorrhages.

CASE PRESENTATION

Case 1

A 14-year-old girl presented to us with a history of sudden onset headache and vomiting. There was no history of trauma, loss of consciousness or seizures. On examination, her blood pressure was 110/70 mm Hg, pulse rate 84/min. Cranial nerves were intact and fundus was normal. There were no focal neurological deficits or meningeal signs.

Case 2

A 45-year-old female presented with acute onset headache and loss of consciousness for a day with gradual regaining of consciousness over the next 36 h. There was no history of trauma, vomiting or seizures. Her blood pressure was 130/80 mm Hg,

pulse rate 90/min. She was drowsy but was able to follow simple commands. She was moving all four limbs, and tone was normal. Tendon jerks were normal and plantars were flexor bilaterally.

INVESTIGATIONS

Case 1

The patient's CT head scan revealed blood in the ventricles and hydrocephalus without any evidence of intraparenchymal haemorrhage suggestive of primary intraventricular haemorrhage (PIVH) (figure 1A). However, incidental calcified AVM was also noticed. CT angiography brain did not reveal LSA aneurysm, though AVM was witnessed. Her blood haemogram and clotting profiles were within normal limits. She was taken up for digital subtraction angiography (DSA). DSA revealed right lateral distal LSA aneurysm (figure 1B,C).

Case 2

CT head scan revealed blood predominantly in lateral and the fourth ventricle, with mild ventricular dilatation (figure 2A). There was no evidence of intraparenchymal bleed. CT angiography brain revealed right medial LSA aneurysm. Further DSA was done which confirmed the presence of right medial LSA aneurysm (figure 2B,C). There was no evidence of moyamoya disease or AVMs.

DIFFERENTIAL DIAGNOSIS

AVMs; moyamoya disease; tumors with aneurysm of feeding arteries.

TREATMENT

Both the patients were treated conservatively, and they recovered gradually and completely.

DISCUSSION

Primary intraventricular haemorrhage (PIVH) is defined as bleeding within the ventricles of the brain without associated parenchymal or subarachnoid haemorrhage (SAH) demonstrated on non-contrast head CT.¹ It accounts for 2–4% of all intracranial haemorrhages.^{1 2} The aetiology of PIVH includes hypertension, AVMs, aneurysms, trauma, moyamoya disease and coagulopathy.¹ Aneurysms are second to AVM, as the bleeding source in cases of PIVH.² Aneurysms can arise from lenticulostriate artery (LSA). Lenticulostriate perforating artery aneurysms are rare.³ Parenchymal haemorrhages are more commonly associated with LSA aneurysm rupture,⁴ but isolated (primary) intraventricular

To cite: Srivastava T, Sannegowda RB, Sharma B, et al. *BMJ Case Rep*. Published online: [please include Day Month Year] doi:10.1136/bcr-2013-009968

Figure 1 (A) CT head scan showing blood in ventricles and hydrocephalus with calcified AVM in right frontal region, without any evidence of intraparenchymal hemorrhage suggestive of PIVH. (B) Digital subtraction angiography (right ICA injection), lateral view shows right distal lateral LSA aneurysm (arrow). (C) Anterior-posterior view showing right distal lateral LSA aneurysm (arrow). AVM, arteriovenous malformation; ICA, internal carotid artery; LSA, lenticulostriate artery; PIVH, primary intraventricular haemorrhage.

haemorrhages secondary to LSA rupture is extremely rare.⁵

Most LSA aneurysms reported are located in the distal course of the artery, within the basal ganglia. Proximal LSA aneurysms are most often middle cerebral artery (MCA)-LSA junction aneurysms, involving some part of the MCA trunk.⁶ LSA aneurysm rupture must be given diagnostic consideration in cases of PIVH.⁵ Catheter cerebral angiography should be considered when non-invasive imaging is unrevealing. Because of the relatively inaccessible location of these lesions for direct surgical or endovascular occlusion, conservative management may be a therapeutic option in patients with LSA aneurysm, and spontaneous resolution can be observed.⁵ Definitive treatment of LSA aneurysms involves both surgical and endovascular methods.^{4 7}

Aneurysms arising from LSA distal to their origin from the MCA are rare.³ Around 29 cases of aneurysm arising from LSA beyond its origin have been reported, and mostly present with intraparenchymal haemorrhage.^{4 8} LSAs are end arteries, small and fragile, and supply the basal ganglia and internal capsule. LSA aneurysms maybe saccular or fusiform, and commonly present with basal ganglia haemorrhage.^{3 8} They are usually associated with conditions like moyamoya disease, hypertension, vasculitis and AVMs.^{3 4 6 8} Treatment of LSA aneurysms is still a controversy because of small patient numbers. However, conservative treatment is appropriate for pseudoaneurysm with follow-up angiography, and surgery is considered in saccular aneurysms.⁹

Aneurysm could be true or pseudoaneurysms. Intracranial pseudoaneurysms encompass less than 1% of all intracranial arterial dilations.¹⁰ Pseudoaneurysms may develop as a result of direct (surgical or penetrating wound) arterial injury and infections,¹¹ and more frequently as a result of indirect (blunt) head trauma,¹² can be related to AVMs¹³ or pathologies like fibromuscular dysplasia (FMD).¹⁴ Arterial aneurysms associated with cerebral AVMs may be classified as intranidal, flow-related, or

unrelated to the AVM nidus. Intranidal aneurysms have a high correlation with haemorrhagic clinical presentation. Patients with flow-related aneurysms in association with an AVM may present with haemorrhage from either lesion,¹⁵ but in our cases, there was no history of trauma or infection, though AVM was present in one of the patients (case 1), and AVM was fully calcified and aneurysm was away from the nidus. As flow in AVM was not seen on catheter angiography, flow-related aneurysm was unlikely in our patient.

Fibromuscular dysplasia (FMD) is a non-atherosclerotic, non-inflammatory disease usually located at the cervical segment of internal carotid artery (extracranial) at C1-C2 level, rarely, FMD can have intracranial involvement with classical appearance of 'string of bead' pattern on catheter angiography.¹⁶ Though aneurysms at the Circle of Willis, along with pathology, like FMD, has been described with incidence varying from 7¹⁷ to 51%.¹⁸ In our cases, possibility of FMD was ruled out as 'string of beads' pattern was not seen on catheter angiography. The angiographic appearance of cerebral pseudoaneurysms is typically that of a poorly defined globular sac with delayed contrast filling and emptying.⁵ In our cases, both the patients had a well-defined aneurysmal sac with neck suggestive of true aneurysm. However, still definitive determination of whether our case represents a true or pseudoaneurysm is difficult, as it requires pathological analysis of the diseased vessel.⁵

Both our patients had single LSA aneurysm, though multiple aneurysms of LSA have rarely been reported. Gupta *et al*¹⁹ reported multiple aneurysms of LSA with left basal ganglionic bleed and showed spontaneous obliteration of these aneurysms on follow-up angiography. Other than deep intraparenchymal haemorrhage, LSA aneurysms can also present in various types either in isolation or in combination, such as SAH, intraventricular haemorrhage (IVH).⁹ LSA aneurysm presenting as pure

Figure 2 (A) CT head showing PIVH with mild ventricular dilation. (B) Digital subtraction angiography, lateral view (right ICA injection) showing right medial LSA aneurysm (arrow), and (C) anterior-posterior view showing right medial LSA (arrow). ICA, internal carotid artery; LSA, lenticulostriate artery; PIVH, primary intraventricular haemorrhage.

SAH is also rare. Eddleman *et al*³ and Bhat *et al*⁸ reported a case of pure SAH due to rupture of the proximal LSA fusiform aneurysm. Kochar *et al*⁴ described a rare case of ruptured fusiform LSA aneurysm presenting as SAH alone, arising from proximal M2 MCA branch, which was detected on superselective microcatheter angiography. However, our patient had isolated ventricular haemorrhage without any evidence of parenchymal or SAH.

Cases of LSA aneurysms with PIVH have been reported in the literature. Vates *et al*⁶ in 2001 described a case of lateral LSA aneurysm and PIVH in a patient of neurocytoma, with an aneurysm originating from a feeding artery. Kuroda *et al*²⁰ reported a case of PIVH due to distal LSA aneurysm rupture in a patient with moyamoya disease. Ellis *et al*⁵ reported a case of medial LSA aneurysm presenting with PIVH in an elderly hypertensive lady.

LSA aneurysms should be considered in young and non-hypertensive (80%) patients with deep intracerebral haemorrhage (ICH).⁹ ICH associated with hypertension is common, but may originate from a ruptured microaneurysm.^{9 21} Even in those over the age of 50 years, deep ICH can still be due to structural abnormalities and should not be dismissed as being a result of hypertension,⁹ and LSA aneurysms can be associated with hypertension along with various vascular lesions.^{3 4 6 8} LSA aneurysms may not be detected on initial angiograms because of their small size (5 mm) and deep location.⁹

Zhu *et al*²² in their study, found that angiographic yield (the frequency of positive angiography in a defined patient group) was significantly higher in patients with ICH at or below the median age of 45 years, than those above and without pre-existing hypertension than those with. In patients of the younger age group without pre-existing hypertension, angiographic yield was 48% in putaminal, thalamic, or posterior fossa ICH and 65% in lobar ICH. In the older hypertensive patients, the yields were 0% and 10%, respectively. However, in patients with isolated intraventricular haemorrhage, yield was high in both the age groups (67% vs 63%). In one of our patients (case 1), CT angiography showed AVM and aneurysm was not detected, but catheter angiogram revealed the LSA aneurysm.

To conclude, LSA aneurysms are rare, and presenting as PIVH is, further, a rare entity and may associate with other vascular lesions in the brain. One should consider catheter angiography in all the patients of PIVH to identify these rare aetiological causes.

Learning points

- Digital subtraction angiography should be considered in all the cases of PIVH to look for etiology, when non-invasive imaging is unrevealing.
- Though hypertension is the most common cause associated with PIVH, extensive evaluation including catheter angiography should be done to look for other causes.
- Lenticulostriate artery aneurysm (LSA) aneurysms are rare and usually presents with intraparenchymal bleed, but rarely these LSA aneurysm can present as primary intraventricular haemorrhage.

Contributors TS did the DSA procedure for these two patients and along with other coauthors involved in writing manuscript and search of literature.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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