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SHORT REPORT



Spontaneous neonatal subdural haemorrhage: always non-accidental injury?

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ABSTRACT

In any neonate or infant presenting with an acute onset of encephalopathy, and/or retinal haemorrhages with no history of major trauma and imaging showing subdural haemorrhage (SDH), the diagnostic priority is to exclude a non-accidental injury (NAI), given the association of these clinical features with the shaken baby syndrome. However, other causes of SDH may present in a similar manner in neonates and infants, in particular vascular lesions such as aneurysms. A four week old neonate presenting with seizures and retinal haemorrhages, was diagnosed with an acute subdural haemorrhage (ASDH) on imaging with significant midline shift needing surgical evacuation. As there was some blood extending into the left Sylvian fissure, further imaging was considered before surgical intervention. This showed a distally located middle cerebral artery aneurysm, which was successfully treated and the neonate made a remarkable recovery. We present a case of a neonate presenting with spontaneous acute subdural haematoma in the absence of classical diffuse subarachnoid haemorrhage, intracerebral haemorrhage or intraventricular haemorrhage, secondary to a ruptured middle cerebral artery aneurysm. The distribution of aneurysms in the neonatal age group is different to adults, with middle cerebral artery aneurysms and more distally located peripheral aneurysms being more common. Vigilance should be borne to exclude the aforementioned as causes for this presentation especially prior to undertaking surgical intervention.

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Introduction

The estimated incidence of acute subdural haematomas (ASDH) in infants is 20–25 cases per 100,000 children under the age of 1-year.¹ The diagnostic priority is to exclude physical child abuse and non-accidental injury (NAI), as this is the commonest cause of an ASDH following shaken baby syndrome.^{1,2} However, this leaves one in five patients with an alternative diagnosis such as vascular abnormalities, external hydrocephalus and coagulation abnormalities. As the following case illustrates, it is crucial to consider these potential, alternate aetiologies when diagnosing and treating neonatal presentations of ASDH. We present a 4-week-old term neonate presenting with a spontaneous ASDH secondary to a ruptured distal saccular middle cerebral artery (MCA) aneurysm who underwent aneurysm embolization followed by emergency craniotomy for ASDH evacuation successfully.

Case report

A four-week old female neonate was admitted to hospital with acute onset of left-sided focal seizures that progressed to status epilepticus. She was born at term (39-weeks gestational age) via normal vaginal delivery and was well from birth to the time of first seizure with no significant fetal or perinatal history. There was no history of trauma or recent infection and no family history of congenital or connective tissue disorders. After resuscitation, her examination revealed that she was moving all her limbs with a subtle weakness on her right side. The head circumference

was 38.2 cm and the anterior fontanelle was flat. Fundus examination revealed bilateral retinal haemorrhages predominantly peripapillary in nature isolated to this single layer.

An emergency non-contrast cranial CT scan demonstrated a significant left-sided fronto-temporo-parietal ASDH with a degree of mass effect and midline shift (Figure 1). Given the clinical findings, in the absence of trauma, a diagnosis of NAI was considered and necessary steps were taken by paediatricians to involve child protection teams, social services and police.

Owing to further deterioration a decision was made for surgical evacuation of a presumed traumatic ASDH. On reviewing the imaging, some of the subdural bleeding was observed to be extending into the Sylvian fissure, raising suspicion that the haemorrhage was extending and emanating from a possible underlying vascular lesion. This was in the absence of other more typical features of aneurysmal haemorrhage such as florid subarachnoid haemorrhage, intracerebral haemorrhage or intraventricular haemorrhage. Magnetic resonance angiography (MRA) and conventional catheter digital subtraction cerebral angiography (DSA) was performed, confirming the presence of a ruptured saccular aneurysm (11 × 11 × 11 mm in all dimensions) located distally on the opercular segment of the left MCA (M3) (Figure 2). Onyx embolisation of the aneurysm was performed followed by emergency craniotomy and evacuation of the ASDH under the same general anaesthetic.

Post-operatively the patient made an uneventful recovery and was discharged on the sixth post-operative day. The peripapillary retinal haemorrhages completely resolved by 4th day of admission. Six-year follow up demonstrates the patient achieving

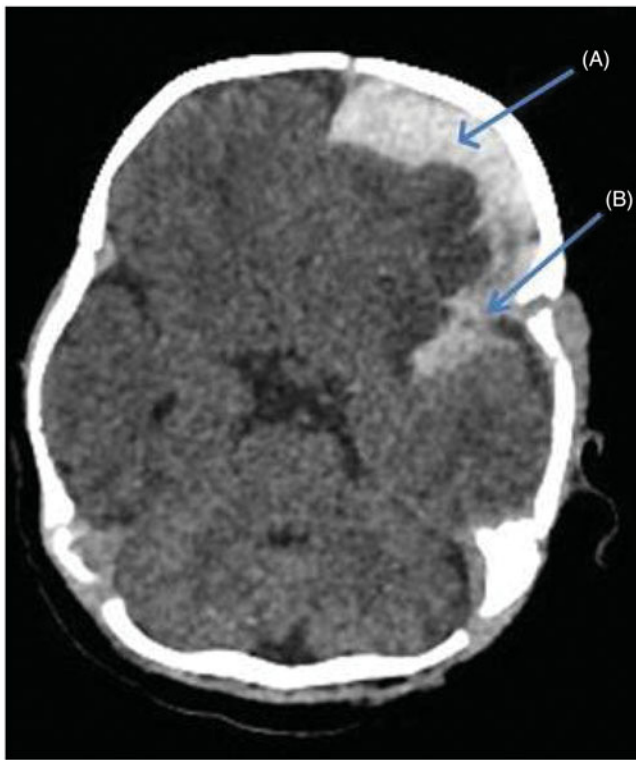


Figure 1. Axial non-contrast enhanced CT scan of the brain. A – Left sided acute subdural haematoma (ASDH) in the absence of diffuse, florid subarachnoid haemorrhage, intraventricular haemorrhage or intracerebral haemorrhage more classically observed with aneurysmal origin; B – Note the sliver extension of the ASDH into the left Sylvian fissure.

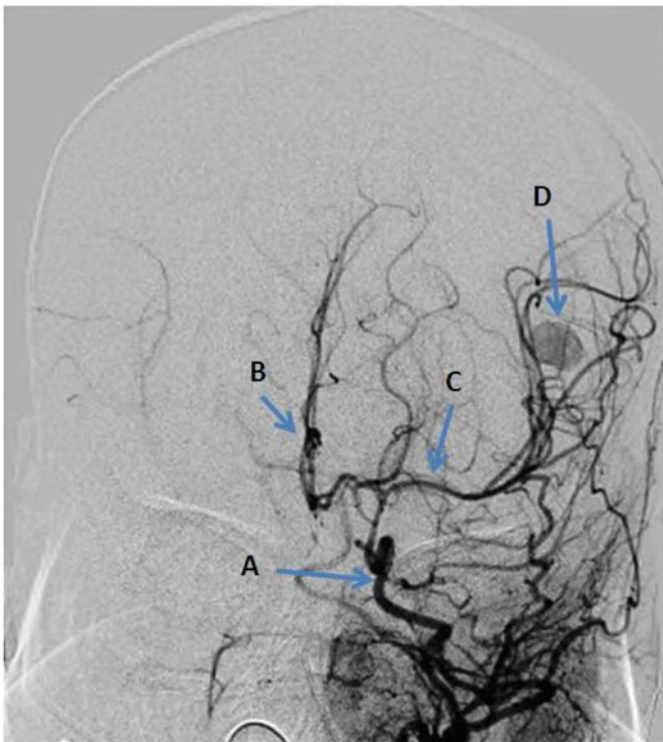


Figure 2. Cerebral angiography with coronal projection demonstrating the presence of a saccular aneurysm projecting from the opercular segment of the left distal middle cerebral artery (MCA). (A) Left internal carotid artery; (B) Left anterior cerebral artery; (C) Left proximal middle cerebral artery – sphenoidal M1 segment; (D) Aneurysm arising from distal aspect of left middle cerebral artery – opercular M3 segment.

her developmental milestones and remaining seizure free off medication. Surveillance imaging has demonstrated no further recurrence of the aneurysm. No risk factors associated with aneurysmal development were identified. The child safeguarding issue was immediately stopped as soon as the diagnosis was confirmed with vascular imaging.

Discussion

We report the unusual presentation and successful management of a ruptured left middle cerebral artery aneurysm presenting predominantly as an acute subdural haemorrhage in a 4-week old neonate. Intracranial aneurysms presenting in the first 4-weeks of life, are exceedingly rare. Our case appears to be only the 6th reported case in the literature of a neonate presenting with acute subdural haemorrhage secondary to aneurysm rupture.³

In any neonate or infant presenting with the triad of acute onset encephalopathy, an ASDH and/or retinal haemorrhages, with no history of major trauma, the diagnostic priority is to exclude NAI⁴ due to the significant causative correlation between physical child abuse or shaken baby syndrome and ASDH presentations. Although exceedingly rare, vascular lesions, albeit a unique subset differing significantly from those found in adults, may be missed under guise of NAI as a cause for ASDH in neonates.⁴ This could lead to a catastrophic outcome if surgical intervention is undertaken to evacuate the haematoma without being prepared for the possibility of encountering a vascular lesion or initially securing the aforementioned abnormality.

A recent systemic review documented 40 cases of neonatal aneurysms in 37 patients (excluding our case).³ The commonest mode of presentation was subarachnoid haemorrhage (SAH) followed by intracerebral haemorrhage (ICH) in 40%. The predilection for presentation as ASDH is rare although commoner than adults owing to differences in distribution and location of neonatal aneurysms. In a previous systematic review of aneurysms diagnosed within the first year of life, the majority were located in the MCA distribution and of these, distal location was commonest and therefore had a higher predilection to present with ASDH and ICH as opposed to pure SAH.⁵

These findings mirror the former literature review on neonatal intracranial aneurysms,³ which documented the commonest location to be the MCA (19/40; ~ 48%) with two-thirds of cases involving the distal intracranial vasculature, and the majority of MCA aneurysms being distal in location (17/19; ~ 89%). Although the presentation of an aneurysmal bleed predominantly as an ASDH in a neonate remains exceedingly rare – 3/40 cases were associated with ASDH and SAH with 1/40 being isolated ASDH – it is crucial to be mindful of this possibility with a spontaneous neonatal ASDH.

The implication of finding retinal haemorrhages adds further complexity as its implication in suspected NAI is controversial and understanding of this finding is in evolution. Multiple pre-retinal, intra-retinal, sub-retinal haemorrhages with extension to retinal edges with or without retinal splitting (traumatic retrolentostylosis) is highly indicative of shaken baby syndrome.⁶ A study of 123 children with SDH secondary to NAI demonstrated 83% to have retinal haemorrhages (of which 85% were bilateral) and missed by non-ophthalmologists in 29% of cases.⁷ However this finding is by no means pathognomonic.

Terson's syndrome is the finding of intra-ocular haemorrhage – which may be vitreous, subhyaloid, intra-retinal (IRH), pre-retinal (PRH) or sub-internal limiting membrane – in the

context of intracranial haemorrhage (SAH in 8–19.3%, intracerebral haemorrhage in 9.1%, traumatic brain injury in 3.1%).⁸ It is hypothesised to be associated with a rapid increase in intracranial pressure transmission through the optic nerve sheath and although commonly seen in adults, it has been reported in children as young as 7-months.⁹ Although a study of incidence of Terson's syndrome in children also revealed that in 57 patients with known intracranial haemorrhage from non-abusive cases, 55 (96%) did not have any evidence of vitreous/intra-retinal haemorrhages¹⁰ suggesting that this is much rarer in paediatric populations.

Furthermore, there is a well-documented association of retinal and fundal haemorrhages affecting multiple layers and in multiple areas in neonates especially associated with vaginal delivery as evidenced by the Newborn Eye Screen Test (NEST) study¹¹ and a further observational study on term newborns at a tertiary centre picked up retinal haemorrhages in 50% of patients, which was significantly associated with vaginal delivery and a shorter duration of the 2nd stage of labour.¹²

These issues certainly make the finding of retinal haemorrhages in the context of any neonate or infant presenting with ASDH a difficult conundrum. The systematic review from the Royal College of Paediatrics and Child Health¹³ provides the most upto date guidance on this issue and addresses most concerns, namely that:

1. Meta analysis provides a high association with retinal haemorrhage and abusive head trauma (AHT) (odds ratio = 15.3 with 95% CI 18.8–25.7)
2. Retinal haemorrhages are rare with accidental trauma and most associated with high impact head injury or crush injury
3. Whilst bleeding disorders are a recognised cause for retinal haemorrhages, no substantive evidence exists for the putative association of retinal haemorrhages with apparent life-threatening events, prolonged cough, seizures and cardiopulmonary resuscitation, although larger scale studies are warranted
4. With infants <42 days of age being investigated with suspected abuse with retinal haemorrhages, consideration should be given as to whether these have occurred as a consequence of delivery (with the commonest mode of delivery associated being either vacuum delivery on its own or with failed forceps delivery)

In our patient, the presence of highly focal peri-papillary haemorrhages restricted to a single layer and in the absence of a more multi-layered (intra-retinal haemorrhage [IRH] and pre-retinal haemorrhage [PRH]) and multi-locational pattern of haemorrhage, these findings may have been a consequence of delivery rather than shaken baby syndrome. The timing of resolution within a matter of days also is consistent with our patient's age – delivery associated haemorrhages appear to resolve within weeks. A study on the natural history of retinal haemorrhages suggests that a combination of multiple haemorrhages (IRH and PRH) in the context of trauma, PRHs in particular, persist for many weeks following the traumatic injury.¹⁴

Our suspicion that the haemorrhages in our patient were delivery related is also supported by the aforementioned study on incidence of Terson's syndrome in paediatrics¹⁰ and RCPCH systematic review in that in paediatric patients, the findings of retinal haemorrhages in the context of intracranial haemorrhage from causes not related to NAI are rare (only 2 cases associated with aneurysm and AVM were found in their systematic review). The only exception to this is in infants within the first 6-weeks

of birth where a high proportion of retinal haemorrhages may be present owing to delivery causes.

Nonetheless, the complexity of the finding of retinal haemorrhages in any infant or neonate suspected of NAI, serves to highlight the importance of all children undergoing ophthalmology examination by an ophthalmologist with pupillary dilatation with a standardised record of all retinal findings in cases of suspected abuse.¹³

Features on initial imaging that should raise suspicion of an underlying vascular malformation in the context of a spontaneous ASDH include presence of the Sylvian fissure. Specialist vascular imaging with non-invasive imaging in the first instance (CT-angiography and contrast-enhanced MRI, the use of contrast-enhancement improving sensitivity over time-of-flight MR-angiography) should be performed early in presence of any suspicious features especially if surgical intervention is being contemplated. Most neonates that remain clinically stable with spontaneous subdural haemorrhage do undergo routine MRI in non-emergent fashion to exclude vascular causes for the haemorrhage as well as assess for other features suggestive of NAI. Microsurgical treatment appeared to be the most common treatment modality overall with increased use of endovascular treatment in the past decade.

Conventional endovascular therapy utilises Guglielmi detachable coils (GDC) but a variety of factors have been associated with higher rates of incomplete occlusion and recanalization such as a wide neck (diameter >4 mm), dome-to-neck ratio <2, intra-aneurysm thrombus, aneurysm size (large and giant) and low coil packing density.¹⁵ Aneurysms with such complex radioanatomy have led to a variety of alternative techniques for endovascular obliteration such as stent-assisted coiling and balloon remodelling.¹⁶ The disadvantage is the need for antiplatelet therapy – a concern especially in the acute setting where acute surgical intervention may be required. The use of Onyx, a liquid embolic agent designed for endovascular treatment, can counteract some of the major difficulties encountered in treatment of wide-necked aneurysms with platinum coils alone: (1) it produces total or near-total filling of the aneurysm sac (2) it induces a stronger aneurysm neck neo-endothelialization reaction compared with regular platinum coils.^{17,18} In our patient, the presence of a wide necked aneurysm together with the need for surgical evacuation of the ASDH and the subsequent need to avoid antiplatelet therapy, led to the use of Onyx endovascular therapy to secure the aneurysm. Definitive intervention to secure the underlying vascular abnormality followed up rapid surgical evacuation of the haematoma can lead to a favourable outcome.³

Conclusions

To the best of our knowledge, this appears to be only the sixth case in the literature of a neonatal intracranial aneurysm presenting predominantly as a spontaneous ASDH, in the absence of more classical diffuse pattern of subarachnoid haemorrhage, intracerebral haemorrhage or intraventricular haemorrhage. The commonest location for neonatal intracranial aneurysms is the MCA with a significant proportion located peripherally on the distal vasculature and these therefore have a higher predilection to present with ASDH. Features that raise suspicion of an underlying vascular malformation in the context of an ASDH include the presence of associated ICH or extension of blood into the Sylvian fissure. A high degree of vigilance is required to

ensure vascular malformations are not missed as a cause for this presentation before undertaking any surgical intervention to avoid an on-table catastrophic outcome.

Disclosure statement

On behalf of all authors, the corresponding author states that there is no conflict of interest.

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