CASE REPORT

Intracranial vasospasm with delayed ischaemic deficit following epidermoid cyst resection

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ABSTRACT. Intracranial vasospasm following surgical removal of intracranial tumours is rare. To our knowledge there have been no previously reported cases of delayed vasospasm occurring post debulking of epidermoid cysts. We report a case in which vasospasm led to established cerebral infarction 2 weeks postoperatively. MRI and serial magnetic resonance angiography (MRA) reliably show initial multiple stenoses in the vertebral and internal carotid arteries followed by their spontaneous normalisation. MRA imaging is now of sufficient quality to enable it to be a safe and effective means of both looking for vasospasm and monitoring its resolution.

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Intracranial vasospasm leading to functional ischaemic deficit following surgical removal of intracranial tumours has been reported before but is not common. The exact pathogenesis remains unclear and is likely to be multi-factorial. We report a case in which delayed post-surgical transient vasospasm led to significant white matter infarction, presenting two weeks after surgical debulking of a recurrent epidermoid cyst. These findings were confirmed by serial magnetic resonance imaging (MRI), magnetic resonance angiography (MRA) and diffusion-weighted imaging (DWI) studies. To our knowledge, there has been one previously published case in which delayed vasospasm following dermoid cyst removal caused functional ischaemic deficit [1], but no previously published case reports of vasospasm occurring post debulking of an epidermoid cyst.

Case report

A 43-year-old male patient with known left cerebello-pontine angle epidermoid tumour underwent debulking surgery for progressive extension into the left temporal lobe and midbrain (Figure 1). Craniotomy and debulking for the same tumour had been performed 10 years previously without incident. On this occasion, surgery and immediate post-operative recovery were uneventful, and subsequent hospital discharge was prompt.

Two weeks after surgery, the patient presented with acute left hemiparesis, neglect, confusion and dysphasia. MRI demonstrated residual epidermoid tumour with a large post-operative pseudomeningocele, and multiple acute focal parenchymal infarcts in the right cerebral

hemisphere (Figure 2). MRA demonstrated multiple stenoses in the extracranial and intracranial segments of the vertebral and internal carotid arteries (Figure 3a).

Vasculitis and thrombophilia screens were negative. The patient was managed conservatively, and further imaging two weeks later demonstrated normalisation of the calibre of the affected blood vessels (Figure 3b). Following a period of rehabilitation, the patient was discharged home. Subsequent clinical review at 8 weeks demonstrated marked improvement of symptoms apart from persistent minor cognitive and speech problems. Repeat imaging at this time confirmed no underlying vascular abnormality (Figure 3c). The patient has also been referred to Cardiology for consideration of closure of an incidental patent foramen ovale (PFO) found on transoesophageal echocardiography.

Discussion

This case shares several features with a previous report of delayed symptomatic ischaemic deficit caused by vasospasm following the resection of an intracranial dermoid cyst [1]. Time and technology have moved on since this original report, and MRA series are now of sufficient quality and resolution to allow a reasonably confident diagnosis of intracranial vasospasm. In this case, the differential diagnosis included vasculitis or a shower of emboli. However, restoration of normal central vessel calibre within a relatively short timeframe in the absence of immunosuppressive therapy makes vasculitis unlikely. Although a PFO was found in our patient's case, the radiological features strongly conflict with cardiac emboli as the pathogenesis of his symptoms.

Vasospasm post-subarachnoid haemorrhage is fairly common and is usually demonstrated at computed tomography (CT), MRI, and catheter angiography in these cases. Although less common, symptomatic vasospasm

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Figure 1. Pre-operative axial T_2 series demonstrates a large epidermoid cyst in left temporal lobe, with moderate local mass effect and midbrain compression.

following intracranial tumour excision has been demonstrated in several small case series [2–5]. The underlying incidence of vasospasm in total is probably higher, but imaging of intracranial vessels is rarely performed in this patient group because the majority of patients are asymptomatic. Vasospasm occurs most commonly at 7–14

Figure 2. Diffusion-weighted axial imaging. Multiple foci of diffusion restriction in the right cerebral hemisphere are susceptibility artefacts from recent surgery in the left frontal cortex.

days following surgery [2, 3], but has been reported up to one month after surgical intervention [2].

Several mechanisms have been postulated in the pathogenesis of vasospasm following tumour excision [2–5]:

- 1. Accumulation of blood breakdown products in the basal cisterns
- 2. Hypothalamic dysfunction as a result of direct irritation or handling
- 3. Manipulation and damage to the blood vessel walls
- 4. Release of chemical substances from the resected tumour
- 5. Autoimmune or paraneoplastic response

Ruptured dermoid cysts have been reported to cause vasospasm leading to symptomatic ischaemic deficit in a handful of cases [6–8]. In one previously published case, delayed cerebral ischaemia manifested 36 h post-operatively as a consequence of vasospasm, presumably due to ruptured dermoid contents found at surgery [1].

Previous hypotheses to explain how ruptured dermoid cysts could cause vasospasm have included the release of the lipid contents of dermoid cysts, causing a chemical meningitis [6–8], and hyperkalaemia post tumour rupture [1, 6]. Chemical meningitis has been reported to occur in 20–40% of cases involving ruptured or incompletely resected epidermoid tumours [9, 10]. Thus, it seems that the irritant effect of the lipid metabolites might contribute to the vasospastic process.

Even though no formal evidence of vasospasm was obtained in our patient's case, serial imaging showing rapid normalisation of cranial vessels suggested that this was the likely aetiology of the clinical presentation. We concluded that the underlying cause of vasospasm was a chemical meningitis resulting from spillage of lipid epidermoid contents into the dural or meningeal spaces.

It has been suggested that corticosteroids given perioperatively, coupled with irrigation of the surgical site with hydrocortisone, could reduce the risk of chemical meningitis in operations involving epidermoid tumours [10]. Although vasospasm due to tumour removal has been treated successfully with hypervolaemic, hypertensive therapy [2, 4, 5], it appears not to be as beneficial when used in cases of vasospasm following dermoid cyst removal [1]. Intraarterial papaverine infusion and balloon angioplasty have been used in resistant cases with some success [2]. This approach is not, however, without significant risk of vessel rupture or further embolisation and is not routinely performed in our centre.

Our patient recovered without the use of any conventional treatment previously used for the management of vasospasm, and was subsequently well enough to be discharged home.

Conclusion

We present a unique case of vasospasm resulting in symptomatic ischaemic deficit that occurred as a complication in epidermoid cyst debulking surgery. This case demonstrates the need for both increased vigilance in the post-operative period and raised awareness of the

Figure 3. (a) Initial time-of-flight MRI angiogram (TOFMRA) at 1.5 Tesla. A surface-shaded selective reconstruction demonstrates critical long segment stenosis in the proximal segment of the right middle cerebral artery (MCA) (arrow) and multiple focal stenoses in the more distal MCA branches with generalised vasospasm. A terminal right carotid stenosis was also present (not demonstrated on this projection). (b) Two weeks after presentation, the same reconstruction from the same virtual viewpoint shows interval increase in the calibre of the proximal MCA, although it remains focally narrowed (arrow), as well as increase in the calibre of the more distal MCA branches. (c) Ten weeks after presentation, the same reconstruction from the same virtual viewpoint (but with TOFMRA at 3 Tesla) now shows no evidence of narrowing in the right MCA at any point. The right carotid stenosis (not shown) has also completely resolved.

possibility of delayed vasospasm leading to ischaemic damage. Complete excision of a tumour can reduce the risk of post-operative chemical meningitis but can be difficult to achieve due to adherence of cyst lining to brain parenchyma or blood vessels. Steroids might also be helpful in reducing this occurrence when used in the perioperative period.

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