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Acute Vasospasm following Transcallosal Resection of a Xanthogranulomatous Colloid Cyst of the 3rd Ventricle

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Word Count: 1817

ABSTRACT

We present the first case of a 57 year old man who developed severe, acute vasospasm following transcallosal resection of an unusual, xanthogranulomatous colloid cyst. The 16 year history of growth of this cyst may have resulted in its unusual pathology, and the subsequent vasospastic reaction to its excision. We discuss the potential pathological relationship between the inflammatory nature of the cyst, chemical meningitis and vasospasm, and what this implies about vasospasm in general. The severe, life-threatening vasospasm affected all four major vessels and required aggressive management by endovascular injection of nimodipine and angioplasty, with good recovery. The case illustrates a previously undescribed sequel of surgery for this condition, demonstrates an effective treatment and offers possible insights into the pathogenesis of vasospasm.

KEY WORDS

Xanthogranulomatous Colloid Cyst, Vasospasm, Neurosurgery, Interventional Neuroradiology

INTRODUCTION

Colloid cysts are rare intracranial tumours with an incidence of approximately 3.2 per million per year^[1], but are one of the commonest intraventricular tumours in man. Due to their propensity to cause chronic hydrocephalus and the reported risk of sudden death^[2], they are a common indication for operative intervention in relatively well patients. As a result, the drive to reduce complications and sequelae of surgery is high. The ideal method of resection of these tumours is a matter for debate, between endoscopic resection and transcallosal microsurgical resection^[3].

One of the important complications is that of aseptic meningitis^[4], potentially resulting from spillage of cyst contents. Here we report a case of vasospasm as a result of aseptic meningitis, following resection of a particularly unusual colloid cyst. This highlights the importance of limiting spillage of cyst contents at excision, may represent a common pathological process in the development of vasospasm, and provides a viable treatment option.

CASE REPORT

Presentation

A 57 year old man was admitted for elective resection of a long-standing colloid cyst of the 3rd ventricle. He presented to the neurosurgical clinic with a 4 month history of short-term memory impairment, episodic confusion and urinary incontinence resulting in his retirement from work, on a background of 20 months of mild decline in cognitive function.

He had originally presented to neurosurgical services 16 years previously, with episodic confusion, gait apraxia and headaches, secondary to hydrocephalus, related to a colloid cyst of the third ventricle. He underwent insertion of bilateral

ventriculoperitoneal shunts and since that time he had returned to a full active life working as a graphic designer.

His previous medical history included type 2 diabetes, diagnosed in the months prior to his second presentation to neurosurgical services, without any evidence of significant vascular sequelae. He currently took 75mg aspirin daily, which was stopped ten days prior to surgery, and 160mg gliclazide daily. He stopped smoking 20 years previously, drank minimal alcohol and lived independently at home with his wife.

On the day of the operation, he was disorientated in time and place and had poor short term recall, but was easily able to identify familiar people. He had no focal neurology on examination, no papilloedema and his systemic examination was unremarkable. His pre-operative imaging is shown in Figure 1.

Surgery

Excision of the cyst was performed via a bicoronal flap, via a right frontal approach. The third ventricle was accessed via a transcallosal approach. It contained a relatively thick-walled cystic structure (compared to a standard colloid cyst), containing thick mucous. This was aspirated without observed spillage of contents. The plane between the cyst and the surrounding structures was easily developed as the cyst 'peeled away' from brain tissue. There was no observed damage to local structures. Histology of the cyst demonstrated a columnar, mucinous epithelium, with multiple macrophages, multinucleated giant cells and cholesterol clefts. There were areas of homogenous colloid-like proteinaceous material, and surrounding reactive gliosis. These findings were consistent with a colloid cyst with xanthogranulomatous change.

Initial recovery and decline

Post-operatively, he was slow to wake from anaesthetic with no verbal response and a fluctuating conscious level with minimal withdrawal response to pain. An urgent CT-scan demonstrated good surgical excision of the cyst, a small volume of intraventricular air, and no acute haemorrhage. His extraventricular drain (EVD) was freely draining CSF. He remained ventilated and sedated overnight, but was successfully weaned off sedation and extubated the following morning. His EVD was clamped and removed at 48 hours. He made a slow but steady recovery post-extubation and at day 10 he was mobilising freely without focal neurological deficit. He was orientated to person and place, but displayed mild hypofrontal behaviour with initial aboulia progressing to disinhibition.

From the ninth post-operative day, his family felt that he was becoming more confused. On day 11, he began to complain of a bilateral throbbing headache. Over the next few hours he had an episode of incontinence before developing expressive dysphasia and limb weakness involving all four limbs, densely weak in the right arm and both legs, with less weakness in the left arm. His conscious level then declined over the next day to no verbal responses, with flexion of the left arm only to pain. An urgent CT scan approximately 1 hour after the onset of this event demonstrated no focal lesion and no increase in ventricular size. Aspiration of a patent shunt reservoir and subsequent lumbar puncture showed normal CSF pressures with a lymphocytic CSF containing 140 lymphocytes and 10 neutrophils, a protein of 1.37g and a non-suppressed glucose, with no organisms, and ultimately no growth on cultures, including fungal and mycobacterial culture. Initially, it was felt that his deterioration

was most likely to be due to either an acute ischaemic stroke or an unwitnessed seizure.

He proceeded to a MRI scan and MR-angiogram at the earliest opportunity. The MRA showed profound vasospasm occurring in all four major vessels at the point where they penetrated the dura, with ischaemia in the body of the corpus callosum, adjacent to the operative site (Figure 2), and smaller areas of restricted diffusion in the medial thalami and deep white matter of the cerebral hemispheres.

He was immediately transferred to Neurointensive care and given empirical ceftriaxone, vancomycin and dexamethasone twice a day, without improvement.

Definitive Treatment

The next day he underwent endovascular intervention via a femoral approach. The cerebral angiogram confirmed the findings on MR-angiogram, and demonstrated reduced perfusion across the major areas of stenosis. The areas of vasospasm were treated by local injection of 1mg nimodipine into each major vessel, followed by four vessel balloon angioplasty. The post-angioplasty angiogram showed a marked improvement in blood flow (Figure 3).

This was associated with a rapid improvement in his conscious level over 4 hours and improvement in both weakness and speech over the next few days. Upon discharge he mobilised freely without focal neurological deficit although he still exhibited episodic hypofrontal behaviour.

DISCUSSION

Colloid cysts are benign intracerebral tumours that originate from the roof of the 3rd ventricle. They present with symptoms of raised intracranial pressure

(headache, nausea, vomiting, memory dysfunction, gait disturbance, visual disturbance and incontinence). Occasionally they can cause sudden death, through either spontaneous rupture or acute obstruction of CSF flow^[2], and may present with cognitive change even in the absence of hydrocephalus^[5]. Management of the cysts can be conservative if less than 5mm in size, or may require CSF diversion, drainage of the cyst or transcallosal resection of the cyst, either under direct observation or via the endoscope.

Xanthogranulomatous inflammation is a term applied to a chronic inflammatory process characterised by cholesterol-laden macrophages. It has rarely been demonstrated within colloid cysts^[6], and may result from an immune response to cyst contents following asymptomatic, partial rupture of the cyst.

Xanthogranulomatous inflammation has been described in several tissues throughout the body including gall bladder, colon, bone and female reproductive tract as an independent idiopathic feature^[7], or as part of a systemic disorder such as Erdheim-Chester disease.

This gentleman suffered profound vasospasm in the postoperative period. This has not been described previously as a complication of simple colloid cyst excision, and has not been seen in direct association with lesions containing xanthogranulomatous inflammation. There are several potential mechanisms: subarachnoid bleeding; arteritis secondary to infectious meningitis; chemical meningitis secondary to spillage of contents of the cyst.

Subarachnoid bleeding is unlikely to be the cause in this case because there was no evidence of subarachnoid bleeding on the immediate postoperative CT. Experience with aneurysmal SAH shows that with a negative CT, even if bleeding had occurred the risk of subsequent vasospasm is minimal. Bacterial or fungal

infection was similarly unlikely as the lumbar puncture at the time of his deterioration was lymphocytic, without suppression of CSF glucose and no growth on multiple cultures.

We hypothesise that chemical meningitis with secondary vasospasm was the most likely cause. His CSF was consistent with aseptic meningitis and cytology showed multinucleate chronic inflammatory cells similar to those seen from the operative sample, which would not be expected in an acute meningeal reaction. It could be argued that a more rapid arteritic response to spillage of cyst contents might be expected if the xanthogranulomatous inflammation represents an immune response to cyst contents. However, the delay may represent a reaction to breakdown products of cyst contents or else a more chronic form of inflammation that takes time to develop, as is seen in granulomatous diseases. In addition, there was evidence of a decline from day 9, and this is well within the time period of vasospasm following subarachnoid haemorrhage.

The mechanism of vasospasm in this case may be analogous to vasospasm secondary to spillage of craniopharyngioma contents, especially as xanthogranulomatous inflammation has been reported as a feature of up to 30% of craniopharyngiomas^[8]. The contents of craniopharyngiomas have been documented to cause aseptic meningitis^[9], and have a direct vasospastic effect on vascular tissue both in ex vivo models^[10], and in vivo in a patient who had spontaneous rupture of a craniopharyngioma^[11]. This is a direct vasopressor response that is neither mediated by inflammation or endothelial damage in the vessel wall, although the specific vasospastic factor in craniopharyngioma fluid is not known.

Refractory vasospasm in the setting of subarachnoid haemorrhage has previously been treated with endovascular catheterisation of the afflicted vessels with

direct injection of vasodilators or balloon angioplasty^[12]. This is the first time we are aware of that this treatment has been used for four vessel vasospasm in the postoperative setting, and the first report of such a use following excision of a colloid cyst. Although this was successful, its prevention would be preferable. This occurred following transcallosal microscopic excision but the risk with endoscopic excision is unknown, but better visualisation of the cyst with pre-excision aspiration of cyst contents should be associated with reduced spillage. Increased post-operative CSF drainage might reduce exposure to cyst contents, however prolonged external CSF drainage by EVD or lumbar drain would have to be balanced against the increased risk of post-operative infectious meningitis/ventriculitis. Alternatively prophylactic dexamethasone for the reduction of the inflammatory response or nimodipine following excision of an inflammatory intraventricular cyst could prevent or reduced vasospasm, and any evidence of early vasospasm with fluctuating neurology, confusion or decreased conscious level could justify these interventions on a case by case basis, ideally confirmed by transcranial Doppler or vascular imaging.

CONCLUSION

Although this sequence of events is rare, it represents a relatively common intracranial vascular response. That vasospasm occurs in response to altered subarachnoid haemorrhage, craniopharyngioma contents and the unusual inflammatory cyst seen in this case, suggests either a common vascular response to multiple insults, or else a vasospastic factor common to all three entities. Our case does not demonstrate the specific mechanism, but provides a possible avenue of investigation. However, it demonstrates a viable therapeutic option for the successful

management of such severe vasospasm, and is the first demonstration of such in this setting.

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Legend for Figure 1

Pre-operative MRI. There is a large, heterogeneous cystic structure superior to the 3rd ventricle that is hyperintense on T1 and T2. The cyst displaces the floor of the third ventricles inferiorly, the corpus callosum superiorly, and the internal capsules and basal ganglia laterally. The ventricles are not enlarged.

Legend for Figure 2

MRI at post-operative day 13, performed 24 hours after the acute deterioration. The DWI demonstrates extensive restriction of diffusion within the corpus callosum indicative of acute ischaemia at the operative site. The MRA demonstrates normal vessel calibre within all four vessels extracranially, with acute vasoconstriction in both

carotids and both vertebral arteries at the point of penetration of the dura mater, indicated by the arrows.

Legend for Figure 3

Comparison of the left carotid and vertebral arteries pre- and post- injection of 1mg nimodipine and balloon angioplasty. There is a marked improvement in vessel calibre and blood flow in both vessels, with improved distal perfusion.