CASE REPORT

Incomplete cauda equina syndrome secondary to haemorrhage within a Tarlov cyst

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SUMMARY

Sacral perineural (Tarlov) cysts are benign, cerebrospinal fluid containing lesions of the spinal nerve root sheath. They are usually asymptomatic; however, a small proportion have the potential to cause compression of nerve roots and/or the cauda equina.

We report a case of a 61-year-old man who presented with acute onset back pain associated with bilateral radiculopathy. Between referral and consultation, the patient developed urinary dysfunction which resolved spontaneously. MRI revealed haemorrhage within a Tarlov cyst, resulting in compression of the cauda equina. Due to the considerable clinical improvement at the time of consultation, surgical decompression of the cyst was not considered to be indicated.

An interval MRI scan 8 weeks later demonstrated that the haemorrhage within the perineural cyst had spontaneously resolved and the patient remained asymptomatic at 5-year follow-up.

resolution of a symptomatic perineural cyst in the cervical spine has been described previously,⁷ to the best of our knowledge, this is the first report of spontaneous resolution of CES caused by haemorrhage into a sacral perineural cyst.

CASE PRESENTATION

A 61-year-old man presented to the emergency department with acute onset lower back pain and bilateral radiculopathy. Following initial assessment, the patient was discharged with analgesia and referred to our spinal surgical unit for an outpatient consultation.

In the 3 weeks that passed between referral and consultation, the patient developed symptoms of urinary hesitancy and altered awareness of the sensation of urinary flow. The patient's symptoms improved and by the time of the spinal outpatient consultation, his pain had become less severe and his bladder symptoms had resolved entirely. Therefore, a decision was made to continue with conservative treatment.

BACKGROUND

Sacral perineural (Tarlov) cysts are benign, cerebrospinal fluid containing lesions that develop within the nerve root sheath of spinal nerves.¹ They are a common incidental finding on MRI scans.² Although the majority are asymptomatic,² a small proportion of sacral perineural cysts have the potential to compress spinal nerve roots and/or the cauda equina.³

Cauda equina syndrome (CES) describes the constellation of signs and symptoms which most commonly results from compression of the nerve roots distal to the termination of the spinal cord. Frequently, the compressive pathology is a large herniated intervertebral disc that obliterates the spinal canal. It is not uncommon, however, for smaller discs to also cause symptoms of CES, due in part to a radiculitis.⁴ Disruption of bladder function is a hallmark feature of CES, the degree of which permits the subclassification into two clinical categories.⁵ CES with retention is characterised by insensate bladder paralysis, resulting in painless urinary retention and eventual overflow incontinence. In contrast, patients with incomplete CES (CESI) retain executive bladder control despite altered urinary sensation, loss of desire to void and/or poor urinary stream.

CES is a spinal surgical emergency, with delayed decompression typically resulting in irreversible neurological sequela.⁶ While the spontaneous

INVESTIGATIONS

An MRI scan was performed following the spinal consultation which demonstrated a cluster of perineural cysts arising from sacral nerve roots within the sacral canal (figures 1 and 2). The largest of these measured 1.9 cm and resulted in mild compression of the cauda equina. The cyst also contained a fluid level suggestive of recent intrasacral haemorrhage.

DIFFERENTIAL DIAGNOSIS

CES results most often from central disc protrusion, herniation or sequestration.⁴ Other less common causes include epidural haematoma, infection, primary and metastatic neoplasms and trauma.⁵

TREATMENT

Due to the considerable improvement in both our patient's pain and neurological symptoms at the time of spinal consultation, surgical decompression of the cyst was not considered to be indicated.

An interval MRI, 8 weeks after initial onset of symptoms (figures 3 and 4), demonstrated complete resolution of the intrasacral haemorrhage.

OUTCOME AND FOLLOW-UP

At 8 year follow-up the patient remained free of symptoms.

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Figure 1 Sagittal T2-weighted MRI scan showing haemorrhage within a symptomatic perineural cyst at the level of S1/2 taken at the time of consultation.

DISCUSSION

Sacral perineural cysts were first described by Tarlov in 1938.¹ The pathology is characterised by cerebrospinal fluid (CSF) accumulation between the endoneurium and perineurium.⁸ They arise at the junction of the posterior nerve root and dorsal root ganglion, and usually display microcommunication with the subarachnoid space.⁸ The aetiology of perineural cysts remains unclear. It has been postulated that cyst formation may be due to spontaneous or traumatic haemorrhage into the subarachnoid space, ischaemic degeneration, proliferation of arachnoid membranes or inflammation of the nerve root with subsequent oedema.^{8–10} Others have suggested that perineural cysts could be developmental or congenital in origin.^{11 12} It is widely agreed that cyst enlargement is due to hydrodynamic and pulsatile forces of CSF infiltrating the cyst, combined with a ‘ball-valve’ mechanism that permits influx while restricting efflux of CSF.^{13–15} Perineural

Figure 3 Sagittal sections of a T2-weighted MRI scan at 8 weeks follow-up showing spontaneous resolution of haemorrhage within the symptomatic perineural cyst.

cysts may become symptomatic if expansion of the cyst causes significant compression and/or irritation of surrounding nerve roots or erosion of adjacent bone structures.¹⁵

With the increased accessibility to MRI scans in recent decades, perineural cysts have become a relatively common incidental finding. Incidence is thought to be approximately 4%–5%, of which only around 13%–20% are symptomatic.^{2 16} In our patient, MRI demonstrated evidence of haemorrhage into one of the cysts which caused compression and inflammation of the adjacent sacral nerve roots and resulted in the clinical features of CESI.

Kong *et al* reported a case of a symptomatic perineural cyst secondary to spontaneous haemorrhage into the subarachnoid

Figure 2 Axial T2-weighted MRI scan showing haemorrhage within a symptomatic perineural cyst at the level of S1/2 taken at the time of consultation.

Figure 4 Axial T2-weighted MRI scan at 8 weeks follow-up showing spontaneous resolution of haemorrhage within the symptomatic perineural cyst.

space.¹⁷ The authors suggested that the haemorrhage resulted in increased hydrostatic pressure of CSF, which leads to the infiltration of CSF and haemorrhage into a pre-existing perineural cyst. The resultant expansion of the cyst caused the patient to suffer from lumbosacral pain. Additionally, Voyadzis *et al* noticed clinical deterioration in a patient following percutaneous drainage of a symptomatic perineural cyst, and suggested this could have been due to haemorrhage of the cyst wall.¹⁰

There is currently no consensus on the optimal management of CES caused by sacral perineural cysts. With regard to symptomatic perineural cysts more generally, surgical interventions such as laminectomy, fenestration and marsupialisation have been described.^{18–21} Particularly promising outcomes have been associated with the use of microsurgical techniques which have been reported to provide symptomatic relief in up to 47.8%–86.6% of cases.^{21,22} However, it is important to note that these figures arise from studies with small sample sizes and are often retrospective in nature.²¹ Therefore, the generalisability of these findings to all patients is limited. Surgical interventions also expose patients to postoperative complications such as infection, neurological deficits, CSF leakage, cerebral haemorrhage and ultimately, recurrence.^{3,21} Advanced patient age, a greater number of cysts on preoperative imaging and prolonged duration of preoperative symptoms have each been associated with poor outcomes.²¹

CT-guided percutaneous aspiration provides a minimally invasive technique and has also been advocated for the management of symptomatic sacral perineural cysts. However, studies have reported mixed outcomes. Paulsen *et al* observed immediate symptom relief which lasted for ≥ 3 months, although recurrence of cysts and their associated symptoms was common.² Patel *et al* and Murphy *et al* reported more favourable outcomes, such as long-term symptomatic relief, using CT-guided percutaneous aspiration in conjunction with an intracystic injection of fibrin glue.^{23,24} To the best of our knowledge there are no reports of CT-guided aspiration of perineural cysts causing CES.

In the UK, symptomatic perineural cysts are most commonly managed non-surgically, largely due to the lack of strong evidence in favour of surgical intervention. However, the evidence for less invasive treatment is also not favourable or strong. Mitra *et al* reported that intralaminar epidural steroid injection provided symptomatic relief in a single patient who experienced radiculopathy caused by a perineural cyst.²⁵ Whereas, Xu *et al* reported poor outcomes in two patients treated with medication and physical therapy for symptomatic sacral perineural cysts.²⁶ Both patients' symptoms worsened with time, and MRI revealed that the culpable cysts had increased in size within 5 years of initial diagnosis. Unfortunately, the small sample sizes mean that reliable inferences cannot be made from these studies.

In our patient, surgical intervention was not considered to be indicated as the clinical features of cauda equina compression had begun to resolve by the time of spinal consultation. Instead, the patient was observed and follow-up MRI at 8 weeks showed resolution of the haemorrhage. Subsequently, the patient made a full recovery, and at 8 years follow-up was pain free and reported normal bladder and bowel control.

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Learning points

- Sacral perineural cysts are benign lesions of the nerve root sheath of spinal nerves.
- Spontaneous haemorrhage within perineural cysts may result in compression and/or inflammation of adjacent nerve roots.
- In some cases, surgical decompression of symptomatic Tarlov cysts may not be indicated. However, we must stress the importance of clinical judgement, by a spinal specialist, in deciding the optimum management of patients on a case-by-case basis.

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