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Case report

Postoperative spinal adhesive arachnoiditis presenting with hydrocephalus and cauda equina syndrome

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Abstract

To our knowledge, the association between hydrocephalus and postoperative spinal adhesive arachnoiditis (SAA) has never been reported. Herein we describe an unusual case of a 45-year-old man with spinal adhesive arachnoiditis (SAA) who developed delayed-onset hypertensive hydrocephalus and cauda equina syndrome (CES) after multiple low-back surgeries. The patient's clinical presentation, imaging findings, surgical management, and the possible mechanisms are discussed in the light of the present literature.

Keywords: Spinal arachnoiditis; Adhesive arachnoiditis; Hydrocephalus; Cauda equina syndrome; Postoperative

1. Introduction

Spinal adhesive arachnoiditis (SAA) is a chronic non-specific inflammatory condition involving the leptomeninges and intrathecal neural elements [1]. A variety of etiologies have been proposed such as infectious, iatrogenic (oil-based myelography, epidural steroid injections, and spinal surgery), degenerative (spinal stenosis), traumatic, and idiopathic [2,3]. SAA has a clear preponderance for lumbosacral localization as reported in 86% of cases [4]. In the majority of cases, arachnoiditis is a coincidental finding without clinical consequences [2,4].

Symptomatic cases may present chronic low-back pain, motor and sensory loss, and reflex changes [2]. It is a chronic disease with late onset and only a minority of patients had progression of symptoms [2,4,5]. Herein, we report an unusual case of SAA presenting with hydrocephalus and cauda equina syndrome (CES) 8 years after complicated spinal surgery.

2. Case report

2.1. History

Our patient is a 45-year-old man who was operated on for lumbar disc hernia (L4-L5) in 1993 at another hospital. Three years later, he was reoperated for a recurrent herniated disc at the same level. This intervention was complicated by a cerebrospinal fluid (CSF) leakage and local infection with *Staphylococcus* requiring surgical closure of the dura and intravenous antibiotics. Since then, his postoperative course was uneventful. A magnetic resonance imaging (MRI) performed in 2002 for an episode of low-back pain was initially interpreted as a banal postoperative status with extradural fibrosis at the operation site. However, at that time, there was noticeably a conglomeration of nerve roots at the posterior thecal sac (Fig. 1). In July 2004 the low-back pain became progressively persistent but relievable medically. The pain was characterized by right-sided sciatica associated with hypesthesia in S1 territory. A control MRI performed in another hospital revealed an intradural mass with contrast enhancement at the level L4-5 and L5-S1 associated with an extradural fibrosis at L4-5 (Fig. 2), and this was diagnosed

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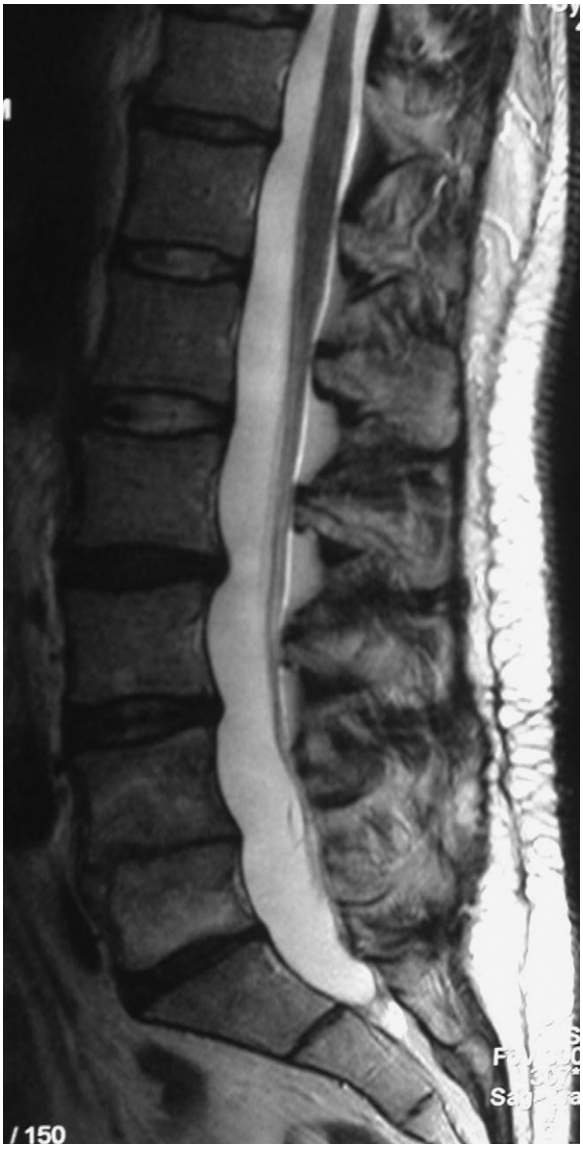

Fig. 1. Sagittal T2-weighted MR image of the lumbar spine in 2002 showing a clumping of nerve roots and an "empty-sac" appearance.

as a tumoral lesion. In September 2004 he was admitted at our hospital due to the onset of severe low-back pain with bilateral sciatica. A few hours later he developed severe headache and started vomiting, and then became stuporous. A computer tomography (CT) scan revealed the occurrence of hydrocephalus (Fig. 3).

2.2. Operation

In emergency, an external ventricular drainage (EVD) was performed. CSF samples were taken both for culture and chemical analysis.

2.3. Postoperative course

Immediately after the EVD, the patient regained normal consciousness status and the headache disappeared. However, the next day he developed sensory disturbances of the lower extremities and urinary retention suggesting CES. Consequently, we urgently performed a lumbar decompressive laminectomy at L4-S1. The dura had an inflammatory aspect and cultures were taken from the dura and the fibrotic extradural mass at the former operation site. After opening the dura, we noticed the presence of a severe arachnoiditis. There was no tumoral lesion. The cauda equina was embedded within an inflammatory mass. Surprisingly, no CSF was found intradurally at the levels between L4-S1. Cultures were obtained from the intradural inflammatory mass and we decided to perform a duraplasty. Postoperatively, the patient improved neurologically. High doses of corticosteroids were administrated in the postoperative phase. We inserted a ventriculo-peritoneal shunt (VPS) after temporary legation of the EVD aggravated our patient, due to persistent active hydrocephalus. This shunt was revised for proximal obstruction. Two weeks later the patient developed an intestinal inconvenience followed by meningitis with *Salmonella*. We removed his VPS and replaced it with EVD concurrently with intravenous antibiotics. Finally, the patient received a ventriculocardiac shunt which remained functional till the last follow-up.

2.4. Laboratory findings

Blood analysis at admission revealed normal C-reactive protein and white blood cell count. CSF cell count depicted a moderate increase of white blood cells (293 mm^{-3}), 85% consisted of neutrophils. CSF protein level was slightly elevated (69 mg/L, range 15–45 mg/L). CSF glucose was normal and lactate was elevated. CSF culture remained sterile including *Mycobacterium tuberculosis*. Samples obtained from the lumbar region intra- and extradurally were sterile. Tuberculin skin test was negative.

2.5. Follow-up

Three months after discharge the patient recovered completely neurologically and had no evidence of motor or sensory deficits. MRI showed a remarkable regression of the adhesive arachnoiditis (Fig. 4) 1 year postoperatively.

3. Discussion

The peculiarity of our case consists in the development of delayed-onset hypertensive hydrocephalus following postoperative SAA. A few authors reported the occurrence of acute hydrocephalus following myelography or spinal anesthesia [6–9]. They attributed that to the presence of contrast material in the cerebral subarachnoid space and consequent inflammation of the cranial leptomeninges [6,7]. In our case there was no history of Pantopaque myelography or epidural steroid injections. The patient had complicated spinal surgeries many years before clinical presentation of lumbosacral

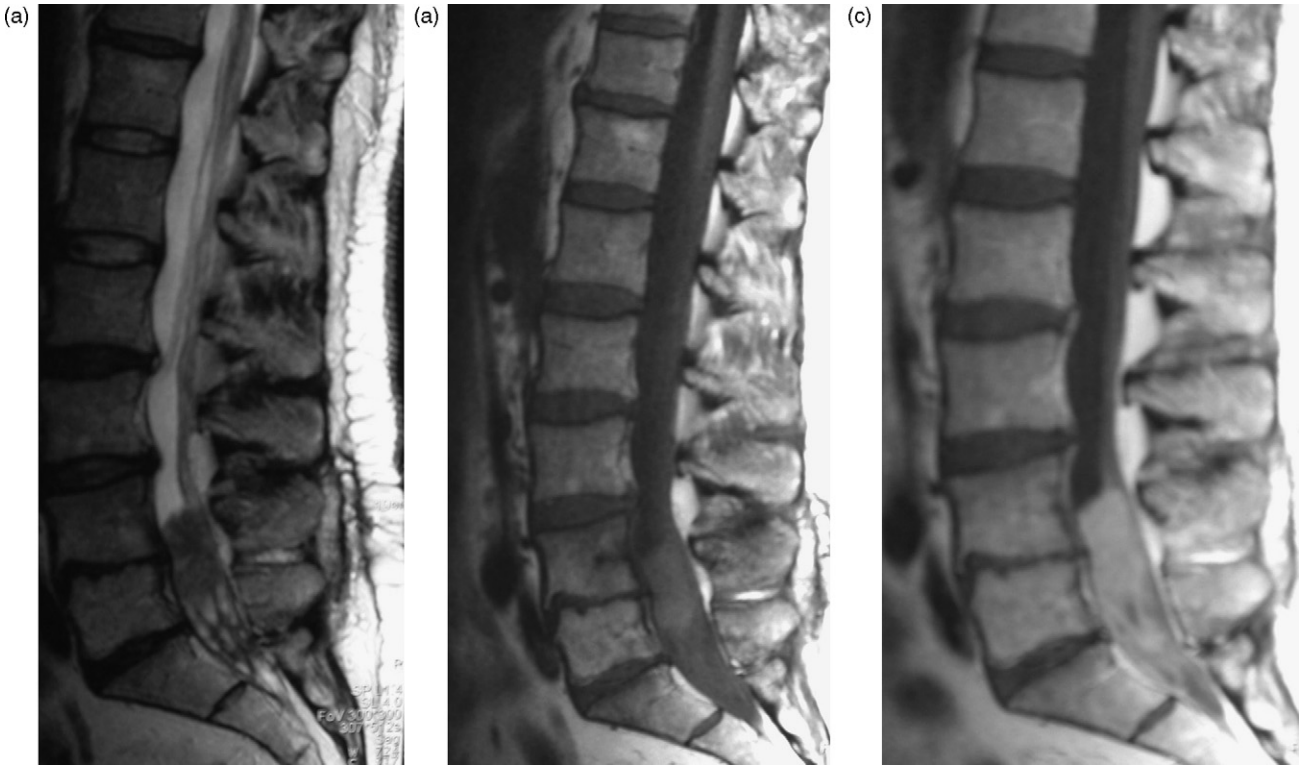

Fig. 2. (a) Sagittal T2-weighted MR image in 2004 revealing a soft-tissue mass and thickened root in the thecal sac. (b) and (c) Sagittal T1-weighted MR image 2004 before (b) and after IV gadolinium administration (c): showing the enhancement signal intensity of the intradural mass.

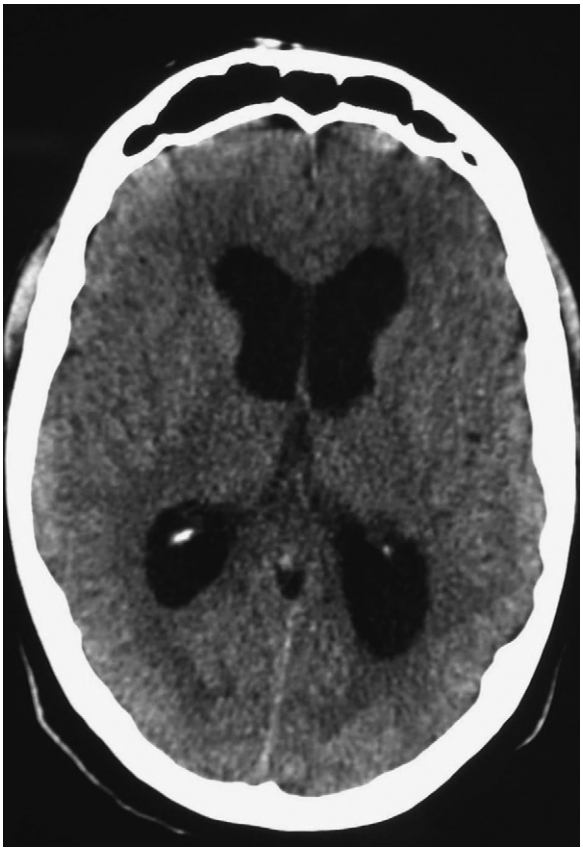

Fig. 3. CT-scan showing hydrocephalus.

SAA. MR imaging revealed a soft-tissue lesion with contrast enhancement initially diagnosed as a tumoral lesion, but actually corresponds to a group III of SAA according to the classification of Ross and Delamarter [10,11].

Recent cases of SAA without acute hydrocephalus reported in the literature are mostly iatrogenic resulting from multiple myelograms and surgeries [1,2,4,5,12,13]. At long-term follow-up ranging from 10 to 21 years, Guyer et al. [5] noted that 90% originally had intervertebral disc disease, Pantopaque myelography, and subsequent lumbar spine surgery.

The exact pathophysiological mechanism has still not been fully elucidated [14]. Three distinctive pathological stages of arachnoiditis were defined: arachnoiditis, adhesive arachnoiditis and arachnoiditis ossificans [1,12]. It has been hypothesized that the pathological process starts with fibrin deposition and a mild inflammatory response followed by collagen deposition and granulomatous reaction [2]. Burton [12] believed that it could result from an inflammatory focus (a herniated lumbar disc), foreign body (especially Pantopaque myelography) and autoimmune reaction. In our case proteinorachia was slightly elevated, and thus unlikely to be the causative factor for the hydrocephalus. Contamination of the CSF with blood during dural breaching could lead to increased intrathecal collagen synthesis and subsequent fibroproliferative reaction and arachnoiditis [15]. Spinal CSF reabsorption can be impaired in this way. Since SAA is able to mimic spinal cord tumors [3] we can imag-

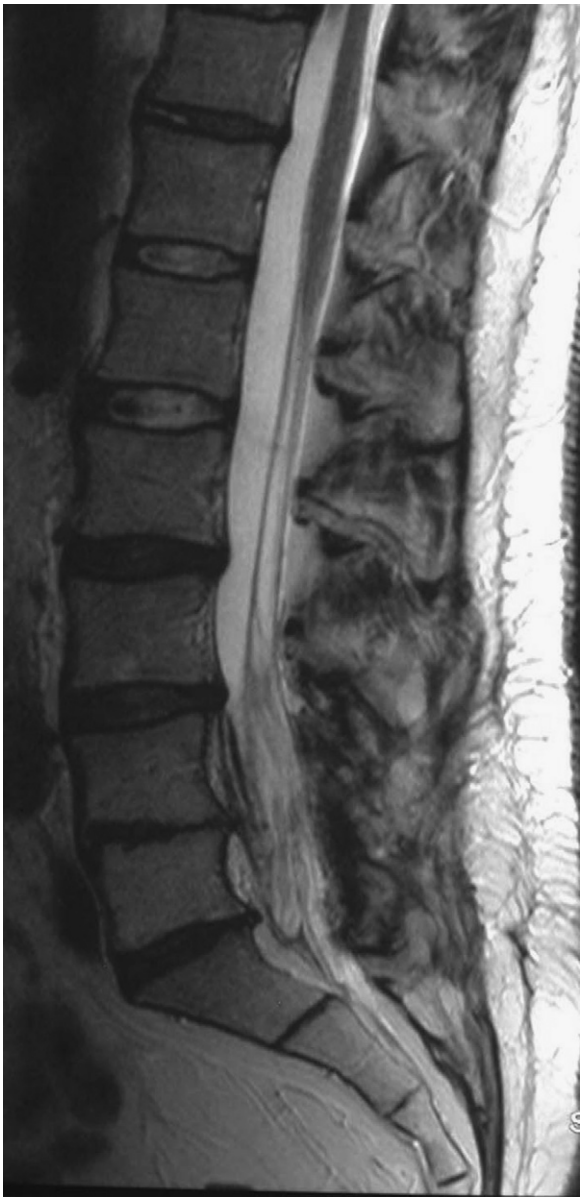

Fig. 4. Sagittal T2-weighted MR image 1 year after decompressive laminectomy, duraplasty and corticoid therapy. Note the reappearance of CSF at the L4L5 and L5S1 level.

ine that hydrocephalus could also result from a reduction in spinal compliance due to obstruction as described in patients with benign intraspinal tumors [16,17]. We assume that in our case the combination of multiple surgeries, infection and dural breaching led to SAA with pseudotumoral spinal obstruction. Subsequently, reduced spinal compliance and CSF reabsorption could possibly explain the development of hydrocephalus in our patient.

Clinical symptoms and signs are unspecific and patients typically present a history of chronic low-back pain and/or leg pain followed by motor loss, sensory loss, and reflex changes [1,2,5]. There is a considerable variability in the interval between the possible etiological event and the time of diagnosis (ranged from 14 days to 32 years) [4,18]. In most patients symptoms varied over time without clear evidence of progression [5,14].

MRI has an excellent correlation with CT myelography and has become nowadays the first choice imaging modality [11]. Ross et al. [11] divided the MR and CT myelography morphologic changes in three groups. In the first group, MRI shows large conglomerations of nerve roots residing centrally within the thecal sac. No peripheral dural thickening was seen in this group. In the second group, MRI demonstrated clumped nerve roots attached peripherally to the meninges with focal thickening of the meninges. In the third group, a soft-tissue signal appears within the thecal sac obliterating centrally the majority of the subarachnoid space. They reported a sensitivity of 92%, specificity of 100%, and accuracy of 99% [11]. The distinction between tumor and the third group of SAA may be impossible by MRI alone [10,11].

Arachnoiditis is a complex neurogenic pain condition and there is no specific clinical syndrome or correlation between imaging and clinical symptoms [1,2,5,18]. Medical and surgical treatments of arachnoiditis have been discouraging at long-term follow-up [4,5,18]. Microlysis of adhesions has been used, but only with moderate success at short follow-up and should be reserved for special cases [2,5,13,18]. Dorsal column stimulation gives sometimes symptomatic relief [1,2,5]. Actually, there is no definite cure or treatment plan for this rare condition [1,2].

4. Conclusions

We report the first case of postoperative SAA presenting with delayed-onset hypertensive hydrocephalus and CES without prior myelography or epidural steroid injections. The exact pathophysiological mechanism remains unclear. We propose that the combination of multiple surgeries, infection and dural breaching led to SAA and subsequent pseudotumoral spinal obstruction. This could reduce the compliance and CSF reabsorption of the caudal spinal canal and thus a possible mechanism of hydrocephalus.

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