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Case study

Idiopathic intradural dorsal thoracic arachnoid cysts: A case series and review of the literature

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ABSTRACT

Background: Spinal intradural arachnoid cysts (SIAC) are cerebrospinal fluid (CSF) filled sacs formed by arachnoid membranes and may be either idiopathic or acquired. Idiopathic cysts represent a separate entity and their aetiology remains uncertain. By far the most difficult differential diagnosis is distinguishing between idiopathic anterior spinal cord herniation (IASCH) and dorsal thoracic intradural arachnoid cysts (TIAC), due to their similarity in radiological appearance. Cine-mode (SSFP) is emerging as a novel technique in the diagnosis and operative planning of SIAC.

Method: Retrospective analysis of patients with idiopathic TIACs that were surgically managed at Royal North Shore Hospital and North Shore Private Hospital between November 2000 and November 2015.

Results: Ten patients were included in this study. Age ranged from 20 to 77 years with a mean age of 60 years and a female preponderance. The most common clinical features were progressive gait ataxia and lower limb myelopathy. Radicular pain tends to improve following surgery, however gait ataxia may not.

Discussion: While there are circumstances in which the distinction between dorsal thoracic intradural arachnoid cysts and idiopathic anterior spinal cord herniation are radiologically obvious, in cases where the appearances are less clear, cine-mode SSFP MRI imaging can provide an invaluable tool to differentiate these pathologies and lead the clinician towards the correct diagnosis and management. The mainstay of surgical management for dorsal TIACs is laminectomy and cyst excision or fenestration. Surgery for gait ataxia should be aimed towards preventing deterioration, while maintaining the potential for symptomatic improvement, whereas surgery for radicular pain should be curative.

1. Background and literature review

Spinal intradural arachnoid cysts (SIAC) are cerebrospinal fluid (CSF) filled sacs formed by arachnoid membranes and may be either idiopathic or acquired [1,2]. The acquired cysts are most commonly secondary to trauma, though any inflammatory insult inducing meningeal adhesion can contribute to their development [3]. Idiopathic cysts represent a separate entity and their aetiology remains uncertain [4]. We wish to report our unique series of idiopathic dorsal SIACs.

Spinal arachnoid cysts are classified into three types: type 1 are extradural cysts without nerve root involvement, type 2 are extradural cysts with nerve root involvement and type 3 are intradural cysts [5]. Type 3 represent a distinct clinical entity (SIAC), and are the focus of this study. SIACs can occur anywhere in the spinal canal, but are most common in the dorsal thoracic and cervical spinal canal [1,6,7]. The typical presentation is of slowly progressive myelopathy however SIAC as a cause for spinal cord compression is generally rare [1].

A variety of spinal cord pathologies can mimic SIACs radiologically, including other benign cystic lesions (neuroepithelial cysts or ependymal cysts), multiple sclerosis and idiopathic anterior spinal cord herniation (IASCH) [1,6,8]. By far the most difficult differential diagnosis is between IASCH and dorsal thoracic intradural arachnoid cysts (TIAC) [1,9]. Imaging evaluation is predominantly by

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magnetic resonance imaging (MRI) and computed tomography (CT) myelogram, each of which has drawbacks. The extremely thin membranes of the arachnoid cysts may not be visible on standard MRI sequences, and the aforementioned differential may have the exact same findings of anterior cord displacement with a prominent CSF-intensity space posteriorly [6,8]. CT myelogram may show the intradural filling defect more effectively, however this remains an invasive, painful procedure with a moderate radiation dose to the patient, and is quickly falling out of fashion [10] A 'scalpel sign' produced by the appearance of the enlarged CSF space on MRI and CT myelogram has been described, however this finding is not present for all TIACs, nor is it entirely specific [11].

Recent developments in MRI sequencing offer potentially more accurate methods for detecting SIACs [12]. Static balanced steady-state free precession (bSSFP) is a subset of gradient-echo steady-state MRI that generates fast and high-resolution images of the CSF space [13,14]. Cine-mode bSSFP MRI is a dynamic technique gated to the cardiac cycle and has superior temporal resolution [13,15]. Cine-mode bSSFP MRI currently plays a major role in the evaluation of CSF flow in cerebral aqueduct stenosis and has been shown to be effective at identifying CSF pulsation and arachnoid adhesions in patients with syringomyelia [13,16]. It is emerging as a novel technique in the diagnosis and operative planning of SIAC.

The literature on idiopathic SIACs is sparse and mostly limited to case reports and small case series. The benefits of operative management are controversial, and although most small case series have suggested an improved outcome post surgery, the evidence in favour of this result is limited [1,3,10,17,18]. Current surgical techniques include laminectomy or laminoplasty followed by durotomy and excision or fenestration of the cyst. Percutaneous aspiration is less invasive but has limited evidence [18].

We describe the clinical features of our patient cohort with idiopathic dorsal TIACs, our experience with their surgical management and demonstrate the value of static and cine-mode bSSFP MRI sequences in the diagnosis and follow-up of patients with this condition.

2. Methods

2.1. Study design

Retrospective analysis of patients with idiopathic TIACs that were surgically managed at Royal North Shore Hospital (RNSH) and North Shore Private Hospital (NSPH) between November 2000 and August 2016. Ethics were/was? approved on 29 January 2016 in accordance with National Health and Medical Research Council (NHMRC) guidelines.

2.2. Database search for patients

Surgical record databases were used to search for all patients who underwent an operation under the ICD10 procedure code 4031200 – removal of spinal intradural lesion. The primary investigator and another independent investigator reviewed the records and applied the selection criteria below. All patients who did not satisfy the inclusion criteria were excluded from the study. An in-depth chart analysis was performed on all patients who satisfied the inclusion criteria. Local PACS imaging databases at the RNSH and NSPH were used to evaluate the imaging on all patients who satisfied the inclusion criteria. Patients with TIACs who had SSFP MRI were analysed separately.

2.3. Inclusion criteria

All patients over the age of 15 with symptomatic idiopathic dorsal TIACs confirmed on histopathology.

2.4. Exclusion criteria

All patients with arachnoid cysts caused by trauma or spine surgery, located extradurally or in the cervical, lumbar or sacral spine, or age under 15.

2.5. Surgical technique

All cysts in this series were approached surgically via laminectomy and then either intradural fenestration or resection under a microsurgical intradural technique.

2.6. Outcomes

Post-operative outcomes were categorised by change in symptomatology at follow-up: resolution, partial improvement, no-change or deterioration. All patients received a follow-up MRI 2–3 months post-surgery to assess the decompression of the spinal cord and resolution of the TIAC.

2.7. Statistical analysis

Data were analyzed with univariate analysis through Microsoft Excel. Descriptive statistics used were mean and percentages.

3. Results

3.1. Clinical features

Ten patients met inclusion criteria and were included in the study. Age ranged from 20 to 77 years with a mean age of 60 years. There was a female predilection of 2:1. The duration of symptoms ranged from 6 months to 5 years with a mean of 27 months.

Thoracic myelopathy was the main presenting symptom with gait ataxia seen in nine (90%) patients. Three patients presented with thoracic radicular pain, one with thoracic back pain, one with sphincter disturbance, and another with overflow urinary incontinence in combination with gait ataxia (Table 1).

Hyper-reflexia was defined as exaggerated reflexes in the lower limbs compared with the upper limbs. Clonus could either be present on one side or both. The most common abnormal finding on clinical examination was hyper-reflexia, seen in six (60%) patients. Other abnormal signs included cross-adductor reflex, clonus, Babinski response, pyramidal weakness, sensory loss and proprioceptive loss (Table 2).

Table 1
Presenting symptoms.

Symptoms	Number of Patients (%) $n = 10$
Gait ataxia	9 (90)
Lower limb sensory disturbance	6 (60)
Radicular pain	3 (30)
Lower limb weakness	3 (30)
Overflow urinary incontinence	1 (10)
Sphincter disturbance	1 (10)
Thoracic back pain	1 (10)