Isolated splenial lesion in the corpus callosum in combination with urinary retention in a case of aseptic meningoencephalitis

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ABSTRACT

Introduction: Aseptic meningoencephalitis in rare instances can present radiologically as a reversible isolated lesion in corpus callosum (Reversible Splenial Lesion Syndrome or RSLS). Patients rarely develop temporary urinary retention, previously described as the meningitis-retention syndrome. Case Report: We report a case of a 28-year-old woman with aseptic meningoencephalitis with an unusual combination of RSLS and urinary retention. A complete resolution of neurological symptoms and urinary retention was noted within two months. Conclusion: In cases of aseptic meningoencephalitis, RSLS and urinary retention are good prognostic factors and are associated with complete recovery.

Keywords: Aseptic Meningoencephalitis, Meningitis-retention syndrome, Splenial lesion, Corpus callosum

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INTRODUCTION

Aseptic meningoencephalitis can rarely present with a solitary lesion in the centre of the splenium of the corpus callosum (RSLS). Its frequency is unknown, however it is likely to be under-detected due to the limited use of routine MRI’s in such patients [1, 2]. The condition is usually associated with an excellent prognosis and a complete recovery [3]. Acute urinary retention develops in rare cases of meningoencephalitis and has been previously described as the meningitis-retention syndrome [4, 5]. This could occur either due to immune mediated demyelinating injury or as an infectious lumbar sacral radiculitis (Elberg Syndrome) [5]. When urinary retention has been reported in cases of RSLS, spinal cord involvement has been demonstrated [4]. We report a case where the clinical and radiological findings suggest a central cause for the urinary retention.

CASE REPORT

A 28-year-old female presented with high grade fever (38.9 °C), bi-frontal headache and urinary retention three weeks post partum. She had associated night sweats,rigors and a cough productive of yellow sputum. Her past medical history was unremarkable and her last pregnancy had been uncomplicated. Her symptoms did not improve on oral antibiotics and she needed urinary catheterization.
Over the course of the next week, she continued spiking temperatures and developed new onset bilateral leg weakness, diplopia and poor concentration. Neck flexion was restricted due to pain and eye examination showed blurring of the disc margins with horizontal nystagmus on rightward gaze. There was increased tone in the legs with moderate pyramidal weakness and reduced rapid alternating movements of the feet. She was unable to stand due to severe ataxia. She was generally hyper-reflexic and the plantar responses were upgoing bilaterally.

Bloods revealed mild lymphopenia (0.6x10^9/L) and hyponatraemia (123 mmol/L) with normal inflammatory markers. Blood cultures, urinary cultures, chest radiograph and pelvic ultrasound were normal. Lumbar puncture showed a mild increase in the opening pressure at 29 cm water with lymphocytic pleocytosis at 146 white cells mm^-3 with 95% lymphocytes, elevated protein (0.76 g/L) and reduced glucose (1.9 mmol/L). Brain magnetic resonance imaging (MRI) showed an abnormal T2 high signal intensity in the splenium of the corpus callosum (figure 1).

The patient was treated with antibiotics, prednisone, acyclovir and anti-tuberculosis medications. Subsequently, haematological and CSF analysis returned negative for influenza, cryptococcal antigen, herpes simplex virus, varicella zoster virus, tuberculosis, listeria, enterovirus, HIV and syphilis.

Within two weeks, there was marked improvement of her neurological symptoms; however she had mild residual ataxia and nystagmus. Repeat MRI of the brain demonstrated an improvement in the amount of splenial hyperintense focus (figure 2). MRI of the spine performed at this stage was normal. Urinary retention was managed conservatively by catheterisation and resolved completely within the next two months along with her neurological symptoms. She remains asymptomatic at a two year follow-up.

Figure 1: A) Sagittal, and B) axial FLAIR (fluid attenuated inversion recovery) images show isolated hyperintense lesion in the splenium of the corpus callosum.

Figure 2: A) Sagittal, and B) axial FLAIR images show improvement in the amount of hyperintense focus in the splenium of corpus callosum two weeks later.

DISCUSSION

Reversible Splenial Lesion Syndrome (RSLS) has previously been described in association with a variety of clinical conditions such as epilepsy, anticonvulsant therapy, ischaemia, acute disseminated encephalomyelitis (ADEM) and mild encephalitis. In cases of meningocencephalitis, RSLS has been linked to a multitude of viral illnesses such as influenza A, mumps, Varicella-Zoster, adenovirus, rotavirus and O-0157 Escherichia Coli [2], however the aetiology is found only in a minor number of cases [6]. Appearance of the splenial lesion on imaging is usually preceded by a short prodrome of flu-like illness followed by onset of neurological symptoms within a few days. Splenial lesions usually present with symptoms of confusion, ataxia, seizures and hemispheric disconnection [1, 7]. MRI findings include hyperdensity on diffusion weighted images (DWI), T2-weighted images (T2WI) and fluid attenuated inversion recovery images (FLAIR). Hypodensity is noted on the apparent diffusion coefficient, iso-intensity on T1-weighted images (T1WI) and no contrast enhancement is seen on midline splenium [7, 8].

It has been postulated that the splenial lesion could arise from intramyelinic oedema due to separation of myelin layers. Another possible mechanism is the influx of inflammatory infiltrate with related cytotoxic oedema [4, 5]. Isolated involvement of the splenium could be due to direct viral invasion causing axonal damage, however pathological correlation is lacking. Ischaemia is unlikely due to the reversible nature of the lesion along with the absence of other lesions in similar vascular distributions [3]. There is often related hyponatraemia as seen in our case, which could be a cause of cytotoxic oedema, however its mechanism is unclear [9]. Differential diagnoses of the lesion include cerebral ischaemia, reversible
posterior leukencephalopathy syndrome, diffuse axonal injury, multiple sclerosis, acute disseminated encephalomyelitis (ADEM) and extra-pontine myelinolysis [3]. The above are unlikely in our patient due to the location and rapid reversibility of the lesion, and the absence of multiple lesions [4]. In general, RSLS is associated with an excellent prognosis and complete clinical recovery is often seen within a month of onset of symptoms [2, 3].

Urinary retention can occur in combination with CSF pleocytosis in cases of viral sacral myeloradiculitis (or ‘Elberg syndrome’) [4, 5, 10-12]. However lumbar sacral symptoms, typical of Elberg Syndrome, such as constipation, paraesthesia, and flaccid paralysis of lower limb muscles were absent in our patient making it an unlikely diagnosis. Additionally, HSV and VZV are the common pathogens involved in Elberg syndrome which had been excluded in our patient. Urinary retention can also develop in rare cases of aseptic meningoencephalitis and has been described previously as the meningitis-retention syndrome [8]. Retention typically develops a week after the onset of symptoms and tends to resolve spontaneously within a month [10]. However patients need to be catheterised in order to prevent over-distension bladder injury [8, 10].

ADEM is such a condition where central nervous system demyelination occurs secondary to an autoimmune mediated injury. Urinary dysfunction is common in ADEM and urodynamic studies in these patients have shown an under active detrusor muscle with hyperreflexia in the acute shock phase [8, 13]. Urinary retention has been previously reported in combination with RSLS on two occasions [7, 13]. Tasclar et al reported a case where diffuse myelitis was noted on radiological findings [7]. More recently, Kitami et al reported a case where the spinal MRI was normal, however sacral involvement was suspected based on clinical findings and lack of detrusor hyperreflexia on urodynamic studies [1, 12]. In our case, the neurological symptoms and examination were suggestive of an upper motor neuron lesion. MRI of the spine was normal nearly two weeks after the initial presentation and any changes of myelitis would be evident by then. RSLS could therefore represent as a mild variant of ADEM leading to improper detrusor contractility in its acute shock phase [8, 13]. However, further research is needed to look at central mechanisms as a cause of urinary retention and similarities between RSLS and ADEM.

In conclusion, aseptic meningoencephalitis in rare instances can present as a reversible isolated splenial lesion in corpus calosum on MRI. Urinary retention is an uncommon feature of RSLS, however it needs to be treated to prevent over-distention injury. Patients can be reassured that both the neurological symptoms and urinary retention are likely to completely resolve.

Author Contributions
Manmeet Saluja – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published.
Nicole Mcgrath – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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