

Case Report

Meningitis retention syndrome: a case report

Farhang Babamahmoodi¹, Roya Sheikholeslami¹, Tahoori Mousavi², Lotfollah Davoodi^{1*}

1. Antimicrobial Resistance Research Center, Department of Infectious Diseases, Mazandaran University of Medical Sciences, Sari, Iran.

2. Ph.D. Candidate in Molecular and Cell Biology, Student Research Committee, Molecular and Cell Biology Research Center, Faculty of Medicine, Mazandaran University of Medical Sciences, Sari, Iran.

*correspondence: **Lotfollah Davoodi**, Antimicrobial Resistance Research Center, Department of Infectious Diseases, Mazandaran University of Medical Sciences, Sari, Iran. E-Mail: Lotfdavoodi@yahoo.com

Abstract:

Urinary retention is a urologic emergency that is defined by an inability to voluntarily void urine, which can be acute or chronic. Benign inflammatory nervous diseases also cause acute urinary retention, in which patients lack apparent urethral outlet obstruction, but exhibit minor neurological and cerebrospinal fluid (CSF) abnormalities. The development of urinary retention in the context of meningitis and CSF pleocytosis without any lumbosacral radiculomyelitis is known as Meningitis Retention Syndrome (MRS). We report a 24-year-old male with complaints of urine dribbling, fever and also complete urinary retention a few hours later and nausea/vomiting, headache and flank pain. The general and genitalia examinations, routine laboratory tests, diagnostic ultrasound, and brain MRI were normal. But HSV Ab (IgG) and PCR HSV were positive in CSF analysis. Finally, Urologist performed the Urodynamic study and flaccid neurologic bladder was reported due the patient was treated with intravenous acyclovir 500 mg, every eight hours, with the diagnosis of MRS. MRS is a rarely described clinical entity with features of meningitis and acute urinary retention. The most common cause of urinary retention is benign prostatic hyperplasia. Other common causes include prostatitis, cystitis, urethritis, stones, and vulvovaginitis; receiving medications in the anticholinergic and alpha-adrenergic agonist classes; and cortical, spinal, or peripheral nerve lesions. Since MRS has a benign and self-remitting course, the effectiveness of immune treatments (steroid pulse therapy) remains unclear, although such treatments may shorten the duration of the disease. Management of the acute urinary retention is necessary to avoid renal injury due to hydronephrosis.

Keywords: urinary retention, Meningitis Retention Syndrome, MRS

Introduction:

Aseptic meningitis is serious inflammation of the meninges [1] and commonly defined as a syndrome characterized by acute onset of signs and symptoms of meningeal inflammation, cerebrospinal fluid (CSF) pleocytosis and the absence of microorganisms on Gram stain and/or on routine culture [2]. Aseptic meningitis is a common neurological disorder [3], but the exact prevalence of viral meningitis is not clear since it is under-reported. Among 2898 patients with such a diagnosis in the UK, 2005-2006, more than 10 times the number formally notified to the Health Protection Agency [4]. Non-bacterial agents including viruses, non-viral pathogens, and non-infectious conditions such as systemic lupus erythematosus, leukemia, lymphoma, nonsteroidal anti-inflammatory drugs (NSAIDs), and other chemicals [1] cause the aseptic meningitis. Urinary retention is a urologic emergency [5] and it means the inability to voluntarily void urine which can be acute or chronic [6]. Benign inflammatory nervous diseases also cause acute urinary retention, in which patients lack apparent urethral outlet obstruction, but exhibit minor neurological and cerebrospinal fluid (CSF) abnormalities. The development of urinary retention in the context of meningitis and CSF pleocytosis without any lumbosacral radiculomyelitis is known as Meningitis Retention Syndrome (MRS) [7].

Urinary retention as a manifestation of neurological disease is much less common than the symptoms of storage failure, namely urinary frequency, urgency and urgency incontinence [5] and only a few reports are available to date [8]. Causes of urinary retention are numerous and can be classified as obstructive, infectious and inflammatory, pharmacologic and neurologic disorders [6]. In neurologic etiologies, sacral herpes, a benign variant of Guillain-Barre' syndrome (inflammatory

radiculitis), typical ADEM, myelitis with leg weakness, typical multiple sclerosis (MS), Neuromyelitis Optica (NMO), herpetic brainstem encephalitis and chemical meningitis secondary to focal subarachnoid bleeding can be expressed [9]. The combination of aseptic meningitis or meningomyelitis and acute urinary retention has been recently acknowledged [8]. Furthermore, MRS has been reported in association with Herpes Simplex-2 (HSV2) and other organisms like mycobacteria and Listeria [7]. A diagnosis is not easily made when a combination of aseptic meningitis and acute urinary retention is confirmed, and such a diagnosis should be established both urologically and neurologically in order to rule out other common causes of neurogenic urinary retention by a detailed history, brain/spinal MRI, and nerve conduction study [9]. In the majority of cases, patients admitted with MRS have meningismus containing fever, headache, a stiff neck, nausea, and vomiting. However, patients might have a variety of symptoms ranging from asymptomatic pleocytosis in the cerebrospinal fluid (CSF) with normal to increased protein content and normal to mildly decreased glucose content to a serious neurological deficiency except urinary retention [1,10]. The underlying pathophysiology of MRS remains unclear [11]. CNS lesions that affect the spinal cord or the brain can cause detrusor areflexia. Lower urinary tract (LUT) dysfunction –is believed to be related to pyramidal tract involvement, and most probably reflects the severity of the spinal cord lesion [9]. But the responsible site of lesions for urinary retention in MRS remains unclear, except for areflexic detrusor at the time of urinary retention [3].

Case Report:

A 24-year- old married male was attended our clinic with the following conditions from 5 days before admission: complaints of urine dribbling, fever and chills and then complete urinary

retention a few hours later, followed by nausea and vomiting, headache and flank pain in the next day. The Patient was admitted to a medical center and antibiotic therapy and fixed catheterization were conducted. Three days later, after catheter removal, he suffered urinary retention and referred to an infectious central hospital. The patient had a headache and brief abdominal pain on admission. General and genitalia examinations were normal. The patient reported a history of trauma to the head 18 months ago for which he was admitted to hospital for a week with the diagnosis of brain contusion and was discharged without any complications. No leukocytosis was found in preliminary tests. Urine analysis Test, Complete Blood Count (CBC), Wright tests, erythrocyte sedimentation rate (ESR), Liver function tests (LFTs or LFs) and the serum levels of Blood urea nitrogen (BUN), C-reactive protein (CRP), fasting blood sugar (FBS), prostate-specific antigen (PSA), and Calcium, phosphorus levels were normal, sonography of the abdomen and pelvis showed no abnormality. On the fourth day of hospitalization, due to continuing headache and MILD stiff neck, lumbar puncture (LP) was performed with suspicion of meningitis. Cerebrospinal fluid (CSF) findings are shown in Table 1.

Brain MRI was performed to rule out space occupying lesions and diseases of the CNS system associated with urinary retention, but no any pathologic findings were reported. Finally, the urodynamic study was performed by a urologist for the patient and flaccid neurologic bladder was reported. The patient was treated with intravenous acyclovir 500 mg, every eight hours, with the diagnosis of meningitis retention syndrome (MRS). A headache improved completely and neurological examination was normal. Three days later the fixed catheter was removed and drainage of residual urine was performed with a temporary catheter for a week periodically. After three weeks of treatment, the

patient had gained complete control of urination and he did not have any problem in the three-month follow-up.

Discussion:

Aseptic meningitis is serious inflammation of the meninges [1] and commonly defined as a syndrome characterized by acute onset of signs and symptoms of meningeal inflammation, cerebrospinal fluid (CSF) pleocytosis and the absence of microorganisms on Gram stain and/or on routine culture [2]. Non-bacterial agents including viruses, non-viral pathogens, and non-infectious conditions such as systemic lupus erythematosus, leukemia, lymphoma, and nonsteroidal anti-inflammatory drugs (NSAIDs) and other chemicals [1] cause the aseptic meningitis. Meningitis-retention syndrome (MRS) is a rarely described clinical entity with features of aseptic meningitis, and urinary retention [12]. Acute urinary retention is extremely rare in children, young adults, and women and the prevalence of this disorder are not known [9]. The most common cause of urinary retention is benign prostatic hyperplasia. Other common causes include prostatitis, cystitis, urethritis, and vulvovaginitis; receiving medications in the anticholinergic and alpha adrenergic agonist classes; and cortical, spinal, or peripheral nerve lesions [6] like Spina bifida occulta/tethered cord syndrome and Fowler's syndrome. Inflammatory neurologic diseases also cause acute urinary retention, in which patients lack apparent urethral outlet obstruction, but exhibit only minor neurological and cerebrospinal fluid (CSF) abnormalities [9]. Herpes zoster is a common painful VZV infection that also presents with urinary retention as a sacral autonomic manifestation when it occurs in the sacral dermatomes (anogenital area) [13].

We present an interesting case of aseptic meningitis further complicated by urinary retention. Few such cases have been published

during the past few years. This combination of symptoms has thus far been termed MRS and no clear associations with other disorders have been made. It is necessary to define MRS in a more precise manner in order to differentiate it from other similar conditions. In most cases reported as MRS, there are no encephalitic signs, distinguishing it from ADEM. Moreover, excluding occasional splenic lesions, MRI of the brain revealed no abnormalities, thereby distinguishing this condition from demyelinating diseases. Lack of leg numbness and paresthesia also helps to differentiate MRS from Guillain-Barre syndrome, polyneuropathies, and conditions affecting the lower motor neurons [9]. In the majority of the published cases, CSF analysis revealed lymphocytic pleocytosis, elevated protein levels, and mildly decreased glucose levels [12] like our patient, with similar results including positive serum HSV Ab and CSF PCR FOR HSV with negative bacterial smear and culture. Myelin basic protein (MBP) suggestive of central nervous system demyelination for the differential diagnosis, a detailed history, brain/spinal MRI, and nerve conduction study are necessary. However, MRI was performed in our patient, which did not reveal any specific pathology. The mechanism of urinary retention of past cases was investigated by urodynamic studies, their brain/spinal/lumbar plexus MRI scans and nerve conduction studies [9]. The results of urodynamic studies showed that all patients examined in other cases had detrusor areflexia, which leads to an inability to contract the bladder on voiding, and two patients had an unrelaxing sphincter such as our case, which the urodynamic study, indicated neurogenic flaccid bladder. Few cases of acute urinary retention with CSF abnormality alone have been reported with no other neurological involvement (i.e., no upper or lower motor neuron signs). These cases can be subdivided into those associated with infectious diseases, such as HSV-2 and

infectious mononucleosis, and those without any evidence of infection [10].

Most patients are young, healthy adults and present with neurological symptoms [12]. Based on the mechanism of urinary retention, these disorders can be divided into two subgroups: disorders of the peripheral nervous system (e.g., sacral herpes caused by herpes simplex virus or varicella zoster virus, with unilateral sacral pain, sensory signs and often skin rashes in the same area); and disorders of the central nervous system, meningitis retention syndrome (MRS), with fever, headache, stiff neck, and minor pyramidal signs, like our patient who had nausea, vomiting and headache, fever and acute urinary retention, at the same time. All these features indicated a central nervous disorder in urinary retention. But in contrast to other cases reported, our patient presented with a stiff neck during the course of the illness and not at the onset and symptoms associated with meningitis such as disturbance of consciousness, hypoesthesia, weakness, and disturbed reflexes of the lower extremities, fecal incontinence, tetraparesis, and diplopia were absent. Several hypotheses have been put forward to explain the detrusor hypofunction and urinary retention in MRS including spinal shock secondary to meningeal irritation, inflammation of upper motor neurons of the spinal cord, direct viral invasion, or development of post-infectious acute disseminated encephalomyelitis (ADEM). Our patient had no symptoms or signs of encephalitis or myelitis and had normal CNS imaging [7]. Whereas brain and spinal cord lesions typically appear in ADEM, such change is not observed in MRS [9]. Since MRS has a benign and self-remitting course (i.e., a duration of 2–10 weeks), the effectiveness of immune treatments (e.g., steroid pulse therapy) remains unclear, although such treatments may shorten the duration of the disease. Management of the acute urinary retention is necessary to avoid bladder injury due to overdistension [10].

Conflicts of Interest

The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.

References:

1. Ntziora F, Alevizopoulos A, Konstantopoulos K, Kanellopoulou S, Bougas D, Stravodimos K. Aseptic meningitis with urinary retention: a case report. *Case reports in medicine*. 2011; 2011, 3.
2. Tapiainen T, Prevots R, Izurieta HS, Abramson J, Bilynsky R, Bonhoeffer J, et al. Aseptic meningitis: case definition and guidelines for collection, analysis and presentation of immunization safety data. *Vaccine*. 2007; 25(31): 5793-802.
3. Tateno F, Sakakibara R, Sugiyama M, Takahashi O, Kishi M, Ogawa E, et al. Meningitis-retention syndrome: first case of urodynamic follow-up. *Internal Medicine*. 2011; 50(12): 1329-32.
4. Rice P. Viral meningitis and encephalitis. *Medicine*. 2013; 41(12): 678-82.
5. Fowler C. Short Commentary on “Acute urinary retention due to benign inflammatory nervous diseases” by Sakakibara et al. in *J Neurol* (2006) 253: 1103–1110. *Journal of neurology*. 2006; 253(8): 1102.
6. Selius BA, Subedi R. Urinary retention in adults: diagnosis and initial management. *American family physician*. 2008; 77(5): 643-50.
7. Krishna A, Devulapally P, Ghobrial I. Meningitis retention syndrome. *Journal of Community Hospital Internal Medicine Perspectives*. 2012; 2(1).
8. Tascilar N, Aydemir H, Emre U, Unal A, Atasoy HT, Ekem S. Unusual combination of reversible splenic lesion and meningitis-retention syndrome in aseptic meningomyelitis. *Clinics*. 2009; 64(9): 932-7.
9. Sakakibara R, Kishi M, Tsuyusaki Y, Tateno A, Tateno F, Uchiyama T, et al. “Meningitis-retention syndrome”: A review. *Neurourology and urodynamics*. 2013; 32(1): 19-23.
10. Sakakibara R, Yamanishi T, Uchiyama T, Hattori T. Acute urinary retention due to benign inflammatory nervous diseases. *Journal of neurology*. 2006; 253(8): 1103-10.
11. Sakakibara R, Uchiyama T, Liu Z, Yamamoto T, Ito T, Uzawa A, et al. Meningitis-retention syndrome. *Journal of neurology*. 2005; 252(12): 1495-9.
12. Basoulis D, Mylona M, Toskas P, Tsilingiris D, Fytili C. Meningitis-Retention Syndrome. *International neurourology journal*. 2015; 19(3): 207.
13. Gayral L. Meningo-radiculite de la queue de cheval et herpes. *Encephale*. 1953; 42(3): 274-82.

Tables and Charts:**Table 1:** The results of the CSF analysis

Variable	Result		Variable	Result
WBC	750			
PMN	5%		GRAM SMEAR	NEGATIVE
Lym	95%		HSV PCR	positive
RBC	50		Leptospirosis PCR	Negative
GLU	65		CMV PCR	Negative
PROT	129		VZV PCR	Negative
LDH	94		Culture	Negative