



# Urinary retention occurring one week after spinal anesthesia: a case of Elsberg syndrome

## Rétention urinaire survenant une semaine après une rachianesthésie: un cas de syndrome d'Elsberg

Kiyoshi Fujii, MD · Katsuyuki Moriwaki, MD, PhD · Tsuyoshi Torii, MD, PhD ·  
Ken Hashimoto, MD, PhD · Kazuhisa Shiroyama, MD, PhD ·  
Minoru Tajima, MD · Mikako Sanuki, MD · Shigeaki Kurita, MD, PhD

Received: 21 July 2014 / Accepted: 9 September 2014 / Published online: 16 September 2014  
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### Abstract

**Purpose** We describe a case of urinary retention caused by viral sacral myeloradiculitis (Elsberg syndrome) that occurred one week after spinal anesthesia. The differential diagnosis of urinary retention after spinal anesthesia is discussed.

**Author contributions** Kiyoshi Fujii managed the patient with the neurologist, Tsuyoshi Torii. Kiyoshi Fujii wrote his case report with Katsuyuki Moriwaki and other coauthors. Katsuyuki Moriwaki, Kazuhisa Shiroyama, Minoru Tajima, Mikako Sanuki, and Shigeaki Kurita reviewed the case. Katsuyuki Moriwaki explained to the patient the clinical importance of publishing this case report in the Journal and obtained the patient's consent. Tsuyoshi Torii made the diagnosis of Elsberg syndrome, treated the patient, and discussed the differential diagnosis of acute urinary retention after spinal anesthesia. Ken Hashimoto managed the patient and wrote his accounts particularly on cauda equine syndrome. Kazuhisa Shiroyama described the difference in presentation of urinary retention in Elsberg syndrome compared with that of ordinary postoperative urinary retention. Minoru Tajima surveyed the literature on viral myeloradiculitis and discussed the diagnosis of Elsberg syndrome. Mikako Sanuki described the detailed time course of the case and discussed the differential diagnosis of Elsberg syndrome from anesthesia-related complications. Shigeaki Kurita reviewed the literature on the concept of Elsberg syndrome and the clinical diagnosis of the syndrome and discussed these issues.

K. Fujii, MD · K. Moriwaki, MD, PhD (✉) ·  
K. Hashimoto, MD, PhD · K. Shiroyama, MD, PhD ·  
M. Tajima, MD · M. Sanuki, MD · S. Kurita, MD, PhD  
Department of Anesthesiology, Critical Care and Pain Medicine,  
National Hospital Organization, Kure Medical Center, Chugoku  
Cancer Center, Aoyamacho 3-1 Kure-shi, Hiroshima 737-0023,  
Japan  
e-mail: moriwaki@kure-nh.go.jp

T. Torii, MD, PhD  
Department of Neurology, National Hospital Organization,  
Kure Medical Center, Chugoku Cancer Center, Kure-shi,  
Hiroshima 737-0023, Japan

**Clinical features** A 76-yr-old male patient presented for operative removal of a right testicular hydrocele under spinal anesthesia. Anesthesia and surgery were uneventful, and he was discharged on the fifth postoperative day. Two days after discharge, he developed intermittent anal pain and voiding difficulty and was readmitted to hospital on the tenth postoperative day. He subsequently developed urinary retention, incontinence of feces, and difficulty in defecation. Magnetic resonance imaging showed no epidural hematoma, abscess, or other lesions in the spinal column, cauda equina, or spinal cord. Neurological examination showed dysesthesia in the perineal region and loss of the anal reflex and bulbocavernosus response, which indicated sacral (S4-5) radiculopathy or a lesion of the conus of the spinal cord. A cerebrospinal analysis showed slight elevation of protein without pleocytosis. After neurologic consultation, herpetic sacral myeloradiculitis was suspected and intravenous acyclovir was administered along with large doses of methylprednisolone and immunoglobulin. The symptoms gradually resolved, and the difficulty in voiding resolved 19 days after initiation of the treatment. The patient was discharged 23 days after the start of the treatment without any other complications.

**Conclusion** This case suggests that Elsberg syndrome is important in the differential diagnosis of urinary retention after spinal anesthesia and should be discriminated from other anesthesia-related complications.

### Résumé

**Objectif** Nous décrivons un cas de rétention urinaire causée par une myéloradiculite sacrée d'origine virale (syndrome d'Elsberg) qui est survenue une semaine après une rachianesthésie. Le diagnostic différentiel de la rétention urinaire après rachianesthésie est discuté.

**Caractéristiques cliniques** *Un homme âgé de 76 ans s'est présenté pour l'ablation chirurgicale d'une hydrocèle testiculaire droit sous rachianesthésie. L'anesthésie et la chirurgie se sont déroulées sans incident particulier et le patient a reçu son congé au cinquième jour postopératoire. Deux jours après le congé, il a présenté une douleur anale intermittente et des difficultés d'évacuation; il a été réhospitalisé au dixième jour postopératoire. Il a ensuite développé une rétention urinaire, une incontinence fécale et des difficultés de défécation. L'imagerie par résonance magnétique n'a mis en évidence aucun hématome péri-dural, abcès ou autre lésion de la colonne vertébrale, de la queue de cheval ou de la moelle épinière. L'examen neurologique a constaté une dysesthésie de la région périnéale et une perte du réflexe anal et de la réponse bulbo-caverneuse, indiquant l'existence d'une radiculopathie sacrée (S4-5) ou une lésion du cône de la moelle épinière. Une analyse du liquide céphalorachidien a montré une discrète augmentation des protéines sans pléiocytose. Après consultation d'un neurologue, une myéloradiculite sacrée d'origine herpétique a été suspectée et de l'acyclovir a été administré par voie intraveineuse avec de fortes doses de méthylprednisolone et d'immunoglobulines. Les symptômes ont disparu progressivement et les difficultés d'évacuation avaient disparu 19 jours après l'instauration du traitement. Le patient a quitté l'hôpital 23 jours après le début du traitement sans autres complications.*

**Conclusion** *Ce cas suggère que le syndrome d'Elsberg est important dans le diagnostic différentiel de la rétention urinaire après une rachianesthésie et qu'il doit être distingué des autres complications liées à l'anesthésie.*

Postoperative urinary retention (POUR) is a common problem among patients who undergo anesthesia and surgery.<sup>1,2</sup> Unlike simple POUR, we report a case where a patient developed delayed urinary retention attributed to viral sacral myeloradiculitis (Elsberg syndrome) one week after spinal anesthesia.<sup>3-5</sup> This syndrome, not readily familiar to anesthesiologists and surgeons, appears to be an important differential diagnosis to discriminate in the adverse sequelae of spinal anesthesia.

### A case report

A 76-yr-old male patient (height 165 cm; weight 65 kg) was scheduled for the removal of a right testicular hydrocele under spinal anesthesia. Other than hypertension, he had no other significant history (including no benign prostatic hypertrophy). For

anesthesia, preservative-free 0.5% isobaric bupivacaine 3.4 mL (Marcaïne™ Spinal 0.5% isobaric, Astrazeneca, Sweden) without intrathecal opioids was injected through the L3/4 interspace using a 25G Quincke spinal needle without paresthesia. Anesthesia and the planned surgery proceeded uneventfully, and the urinary catheter was removed on the first postoperative day. The patient experienced no prolonged POUR and was discharged on the fifth postoperative day. Two days after discharge, he began to feel intermittent anal pain and complained of voiding difficulty. He was readmitted under the care of the urology department on the tenth postoperative day and gradually developed severe urinary retention, incontinence of feces, and difficulty in defecation (i.e., dyschezia) in the following days. Urinary retention was treated with intermittent urinary catheterization and an alpha-1 blocker (Urapidil). Although nonsteroidal non-inflammatory drugs somewhat improved the pain, his symptoms remained. As an epidural hematoma was a suspected cause of the urinary retention, anesthesiology and neurology consultations were requested seven days after the onset of urinary retention. On the day of consultation, the patient was afebrile and showed no sign of meningitis. A neurological examination showed perineal dysesthesia along with loss of the anal reflex and bulbocavernosus response, which indicated bilateral sacral (S4-5) radiculopathy or a lesion of the conus of the spinal cord. He had no infectious skin lesions in the urogenital region and showed no sensory abnormalities or motor weakness in the lower extremities. A magnetic resonance imaging (MRI) using gadolinium diethylenetriaminepentaacetic acid showed no epidural hematoma, abscess, or other lesions in the spinal column, cauda equina, or spinal cord. A cerebrospinal fluid (CSF) analysis on the day of consultation showed lymphocytes  $3 \text{ mm}^{-3}$  (normal range: 0-5), protein  $44 \text{ mg} \cdot \text{dL}^{-1}$  (normal range 10-40), and glucose  $70 \text{ mg} \cdot \text{dL}^{-1}$  (normal range: 50-75), which indicated slight elevation of protein without pleocytosis. With a provisional diagnosis of viral (i.e., herpetic) sacral myeloradiculopathy, the patient was given acyclovir 250 mg *iv* every eight hours for seven days coupled with methylprednisolone 1,000 mg *iv* as pulsed steroid therapy for three days and large doses of immunoglobulin (25 g) for five days. The symptoms gradually resolved, and dyschezia disappeared eight days after initiation of the treatment. After additional pulsed steroid therapy for three days, the patient's voiding difficulty disappeared completely 19 days after the initiation of therapy. He was discharged 23 days after the start of the treatment without further complications. Serum antibodies of herpes simplex virus Type 1 (HSV-1) and Type 2 (HSV-2) and varicella-zoster virus (VZV) from samples taken on the day of neurologic consultation were

elevated. The VZV-IgG antibody index, measured using enzyme immunoassay, was 12.0 (normal range: < 2), and based on a neutralization test, antibodies of HSV-1 and HSV-2 were 32 times normal (normal < 4). Antibodies of HSV-1, HSV-2, and VZV were negative in the CSF. Antigen-specific IgG and immunoglobulin (IgM) antibody titers of HSV-1 and HSV-2 in the serum at 46 days after the onset of the symptoms were >128 times and < 0.8 times the normal range, respectively.

## Discussion

When urinary retention occurs after neuraxial anesthesia, the cause may logically be attributed to adverse sequelae of anesthesia.<sup>1,2</sup> In the present case, the patient had at least five days of intact urinary function after surgery until the onset of urinary retention. In addition, anal pain, hypoesthesia in the sacral dermatome, and incontinence associated with the urinary retention were also delayed in presentation until the seventh postoperative day. Therefore, given the delayed presentation after an otherwise full recovery immediately after the spinal anesthetic, it was unlikely that the spinal needle caused direct damage to spinal nerve roots or the spinal cord. Epidural abscess or hematoma was not seen in the MRI, leaving the possibility that the delayed neural damage was caused by local anesthetic neurotoxicity as a potential anesthetic complication. Nevertheless, findings of a slight elevation of protein without pleocytosis in the CSF and symptoms indicated viral (i.e., herpetic) sacral myeloradiculitis (Elsberg syndrome) in our case. The diagnosis of Elsberg syndrome was made based on the clinical presentation, in conjunction with the elevated antibody titers of HSV-1 and HSV-2 and VZV in the serum, and good response to antiviral therapy.

Viral (i.e., herpetic) sacral myeloradiculitis is rare but an important differential diagnosis of acute urinary retention.<sup>3–5</sup> Elsberg syndrome is defined as urinary retention with sacral herpes and sacral radiculitis caused by HSV or VZV.<sup>4,5</sup> It has been reported that other viruses, including cytomegalovirus, Epstein-Barr virus, and enteric cytopathogenic human orphan virus, also cause similar sacrum myeloradiculitis resulting in urinary retention.<sup>3–5</sup> In a broad sense, therefore, the term, Elsberg syndrome, is also used in cases with those viruses.<sup>4,5</sup>

Viral titers in the serum and CSF are useful to determine the specific causal virus;<sup>3–5</sup> however, the antibodies for HSV and VZV in CSF were negative in our patient. Previous studies have reported that they are not necessarily positive in sacral radiculitis with such viruses.<sup>3,5</sup> Some reports outline the use of polymerase chain reaction testing to identify the causative virus in the CSF,<sup>3,4</sup> but this was not available for our case.

Although we did not identify which specific virus was responsible for the radiculomyelitis in our patient, HSV-2 infection has been reported as responsible for significant neurological morbidity more than any other virus.<sup>6</sup> In addition, sacral dorsal ganglia are preferentially affected by HSV-2;<sup>6</sup> therefore, it seems reasonable to assume HSV-2 infection was the causative virus in our case. The primary infection of HSV-2 often accompanies painful vesicular genital skin lesions;<sup>3</sup> however, such skin lesions are sometimes absent in cases of recurrent HSV-2 infection,<sup>4</sup> as in our case. Elevated HSV-specific IgG antibody titer and normal IgM antibody titer measured 46 days after the onset of urinary retention could support the recurrence of HSV.

A report by Yoshida *et al.* outlined urinary retention occurring several days after spinal anesthesia.<sup>7</sup> They described acute urinary retention associated with sacral herpes-zoster in a patient who underwent a Cesarean delivery under combined spinal-epidural anesthesia.<sup>7</sup> The onset of urinary retention occurred on the sixth postoperative day – similar to that of our patient – with epidural hematoma again being ruled out with MRI. Up until that time, their patient had an uneventful postoperative course.

Another differential diagnosis of urinary retention after spinal anesthesia to consider is cauda equina syndrome (CES) due to neurotoxicity of local anesthetics after spinal anesthesia.<sup>8,9</sup> In addition to urinary retention, the neurological symptoms can include fecal incontinence, perianal hypoesthesia, and possibly a negative MRI, as reported by Kato *et al.*<sup>8</sup> As for MRI findings, however, in one case report of a patient with transient neurologic symptoms, enhancement of the mildly thickened cauda equina and lumbosacral nerve roots on a post contrast study was described as being attributed to lidocaine neurotoxicity.<sup>10</sup> MRI in viral myeloradiculitis may also show varying degrees of root or lower spinal cord edema with enlargement and hypersensitivity on T2-weight images in some patients,<sup>4</sup> though this was normal in our patient. Therefore, MRI findings themselves cannot discriminate abnormalities in cauda equina and spinal roots in Elsberg syndrome from those in CES due to neurotoxicity of local anesthetics.<sup>10</sup> To discriminate CES due to neurotoxicity of local anesthetics from Elsberg syndrome, CSF examination and measurement of viral antibody titers both in the CSF and serum may give crucial information.

Recurrence of HSV-1 infection has previously been described in a parturient receiving intrathecal and epidural morphine for pain management.<sup>11</sup> Although no report has suggested any reactivation of HSV-2 and VZV after neuraxial anesthesia,<sup>11,12</sup> the possibility that spinal anesthesia triggered a viral reactivation in the sacral

dorsal root ganglia cannot be ruled out in our patient. Nevertheless, other factors, such as emotional or surgical physical stress, may contribute to decreased immunity to viruses.<sup>11</sup> Therefore, the mechanism of viral reactivation in the sacral ganglia after spinal anesthesia remains speculative.

Prompt administration of the acyclovir antiviral agent, large doses of corticosteroid, and immunoglobulin therapy were associated with relief of the symptoms in our patient. Although viral myeloradiculitis is thought to be self-limiting and generally has a good prognosis, serious complications, such as necrotizing myelitis and death, have been reported.<sup>3,4</sup> Early diagnosis and aggressive treatment appear key to avoiding such severe complications.

**Funding** No funding was obtained for this work.

**Conflicts of interest** None declared.

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