

Case Report

Man-in-the-barrel syndrome: Case report of ventral epidural abscess and review of the literature

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Abstract

Background: Man-in-the-barrel syndrome (MBS) is an uncommon clinical condition for which patients present with bilateral brachial diplegia but intact lower extremity strength. This syndrome is typically attributed to a cranial/cortical injury rather than a spinal pathology.

Case Description: A 62-year-old diabetic male presented with bilateral upper extremity paresis attributed to a ventral cervical epidural abscess diagnosed on magnetic resonance imaging. Emergent cervical decompression resulted in slight improvement of upper extremity strength. However, he later expired due to sepsis and respiratory compromise.

Conclusion: Establishing the correct diagnosis via clinical examination and proceeding with appropriate management of MBS attributed to a cervical epidural abscess is critical to achieve a good outcome.

Key Words: Brachial diplegia, man-in-a-barrel syndrome, ventral cervical epidural abscess

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INTRODUCTION

First reported by Mohr *et al.* in 1969, man-in-the-barrel syndrome (MBS) is uncommon and characterized by bilateral brachial diplegia but intact lower extremity function.^[4] Upper extremity weakness is greater proximally than distally, whereas motor function remains intact in lower extremities. Most cases of MBS are attributed to intracranial pathology. Here, however, we present a patient with a cervical epidural abscess who presented with MBS.

CASE REPORT

Clinical data

A 62-year-old diabetic male presented with left upper extremity weakness. Two days after admission, he

developed proximal bilateral upper extremity motor deficits but intact lower extremity strength. Motor compromise was characterized by shoulder abductors 1/5; elbow flexors 1/5; elbow extensors 3/5; wrist extensors 4+/5; finger flexors; and abductors 4+/5 (Medical Research Council grading scale) bilaterally. Deep tendon reflexes were 1+ in both upper and lower extremities,

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and were accompanied by bilateral Babinski's and Hoffmann's signs without clonus. He also complained of and demonstrated focal tenderness in the posterior cervicothoracic spine.

Laboratory and radiographic evaluation of cervical epidural abscess

Laboratory examination showed leukocytosis (24.8 K/uL), elevated C-reactive protein (38.4 mg/dL), and an elevated erythrocyte sedimentation rate (106 mm/h). The magnetic resonance image (MRI) of the cervical spine showed a ventral cervical epidural abscess extending from C2 to C6 [Figure 1a and b]. The patient underwent an emergent cervical laminectomy (C3–C6) to decompress the cervical cord. Purulent fluid was expressed ventrally using a nerve hook. A C3–C6 cervical fusion was then undertaken utilizing standard lateral mass fusion techniques and a surgical drain was left in place.

Postoperative course

Postoperatively, the patient exhibited slight improvement in his proximal upper extremity motor strength; 2/5 in the

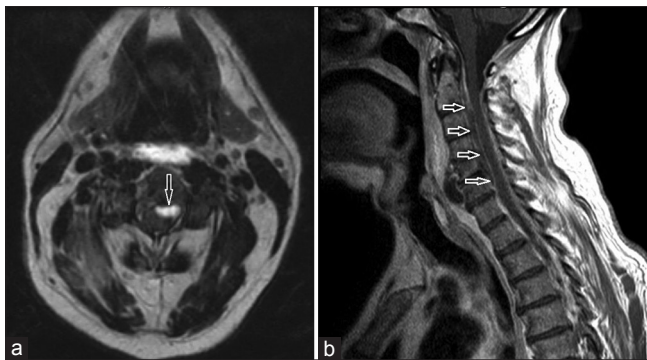


Figure 1: (a) Axial MRI cervical spine T1 with contrast showing a contrast-enhancing ventral epidural mass consistent with an abscess or phlegmon. There are no subacute or chronic intramedullary changes in the cervical cord that would otherwise suggest a chronic pathology. (b) Sagittal MRI cervical spine T1 with contrast showing a ring-enhancing ventral epidural mass from C3–T1 consistent with abscess or phlegmon. The ventral epidural mass exhibits mass effect on the cervical cord with effacement of the dura

shoulder abductors and elbow flexors. Surgical cultures identified: *Streptococcus anginosus*, *S. constellatus*, and *Staphylococcus epidermidis* (methicillin sensitive). The initial broad-spectrum antibiotic therapy was narrowed down to intravenous cefazolin as recommended by the infectious disease team for a total of 6 weeks. Unfortunately, the patient became hemodynamically unstable due to sepsis/aspiration pneumonia on postoperative day 9 and expired.

DISCUSSION

Three clinical syndromes describe isolated upper extremity weakness; cruciate paralysis of Bell, MBS, and anterior spinal artery (ASA) syndrome. In each of these syndromes, the symptoms are largely permanent and irreversible.

The mechanism of neurological injury varies according to the underlying pathology. ASA syndrome and cruciate paralysis of Bell are typically related to vascular and cervical cord injuries.^[6] Alternatively, MBS mostly occurs secondary to cerebral/cortical injury and usually results in isolated bilateral upper extremity plegia. It was first described by Mohr *et al.* and was attributed to bilateral brain infarcts in border zones (e.g. watershed infarcts of middle and anterior cerebral artery distributions, central pontine myelinolysis, and decussation of the pyramids) secondary to cerebral hypoxia.^[4] According to the literature, only 1 in 11 patients with MBS secondary to cerebral hypoperfusion survived.^[6]

There are less than 10 cases of MBS reported due to spinal cord pathology [Table 1],^[1,2,5,6,8,9] where it is attributed to injury of the anterior horn cells resulting from trauma and/or vascular insults (e.g. vertebral artery dissection or thrombosis).^[2] Berg *et al.* offered an atypical variant of the anterior spinal artery syndrome where incomplete pial collaterals spared the lower extremity motor neurons but selectively damaged the upper motor neurons resulting in upper extremity plegia.^[2]

Table 1: Reports describing cases of Man-in-the-Barrel syndrome secondary to spinal pathology

Author & Year	Age (y), Sex	Presentation	Etiology	Treatment	Outcomes
Soubrier, 1995 ^[6] , *	Unknown*	UE weakness	Spinal epidural abscess	Unknown*	Unknown*
Renard, 1997 ^[3] , *	Unknown*	Unknown*	Unknown*	Unknown*	Unknown*
Berg, 1998 ^[2]	64, F	UE weakness	VA dissection	Anticoagulation	Same exam; improved pain
Strupp, 2000 ^[7]	56, F	Vertigo, double vision, UE weakness	Thromboembolism, PFO	Anticoagulation	Stable
Ben Sassi, 2009 ^[1]	53, M	UE weakness	PICA & ASA infarct – thrombogenic state	Anticoagulation	Dead (mesenteric infarction)
Rodriguez-Vico, 2010 ^[4] , *	Unknown*	Unknown*	Unknown*	Unknown*	Unknown*
Yoon, 2013 ^[8]	42, M	UE weakness, neck extension	Neck extension, cervical spondylosis	Steroids	Good
Present case	62, M	UE weakness	SEA	Cervical decompression, fusion + Abx	Dead (aspiration/sepsis)

*The full articles couldn't be obtained despite extensive literature search/or if found, were not in English and the extracted information was limited

In the case presented here, MBS was caused by a cervical spinal epidural abscess (SEA). The pathophysiology resulting in MBS deficit included both compressive trauma and vascular ischemia. On the other hand, most patients with spinal MBS have vascular dissection, thromboembolism, or spondylosis (e.g., as noted here). Although most patients with spinal MBS do well, our patient expired from pneumonia/sepsis.

Our treatment rationale was focused on the goal of rapid cord decompression along with source control and reducing the infectious burden. We elected instrumented fusion given the multilevel laminectomy, which was concerning for mechanical instability and delayed spinal cord injury through loss of the posterior tension band and resultant kyphosis of the cervical spine. The use of metal in vertebral osteomyelitis and nonsterile spinal procedures is controversial. In the past decade, increasing research on the subject of the biocompatibility of metallic implants in the setting of pyogenic vertebral infections have led to increased use where it has been considered as a safe option.^[3]

CONCLUSION

Here, we reported the case of a 62-year-old male who presented with MBS secondary to a ventral cervical epidural abscess. Despite prompt intervention, the patient died due to medical comorbidities.

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Conflicts of interest

There are no conflicts of interest.

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