Muscle weakness of the lower limbs after epidural anesthesia in a pregnant woman with undiscovered systemic lupus erythematosus

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To the Editor: A 29-year-old nulliparous woman (weight 68 kg, height 159 cm) at 31 ± 1 gestational weeks was admitted to the West China Second Hospital, complaining of lower limb edema for 3 months and dyspnea for 1 week, which had become more severe over the last 2 days. She was confirmed pregnant at a local hospital after experiencing amenorrhea for 40 days. All routine tests were normal until she was found to be anemic without symptoms at 18 gestational weeks. Her hemoglobin (Hb) was 77 g/L without thalassemia. After oral intake of a polysaccharide iron complex for 4 weeks, her Hb increased to 81 g/L with edema in her lower limbs. Urine testing was positive for protein (+) and occult blood (3+). At 30 gestational weeks, she felt chest distress, palpitations, and fatigue and could not maintain a supine position at night. A routine blood examination showed an Hb level of 67 g/L, and the transthoracic echocardiography showed mild mitral stenosis with moderate regurgitation and mild pericardial effusion. She was then admitted to our emergency hospital with orthopnea and edema. Her physical examination showed a heart rate of 120 beats/ min and blood pressure of 110/70 mmHg. A wet rale could be heard at the left lung. Laboratory tests showed that her Hb was 73 g/L, brain natriuretic peptide (NT-BNP) 10,300 pg/mL, troponin 0.098 µg/L, myohemoglobin 146.4 µg/L, glutamic-pyruvic transaminase 98 IU/L, glutamic oxalacetic transaminase 100 IU/L, albumin 25.7 g/L, urea nitrogen (BU) 11.64 mmol/L, creatinine (Cr) 138 mmol/L, and potassium 5.63 mmol/L. The urine test was positive for protein (3+). An ultrasound test showed left pleural effusion. After multiple department consultations, the pregnancy was terminated to improve the patient's situation.

After entering the operating room, the patient was routinely monitored. An epidural catheter was inserted through the lumbar 1 to the lumbar 2 intervertebral disc. A test dosage of 3 mL of 1% lidocaine was injected into the

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Quick Response Code:	Website: www.cmj.org
	DOI: 10.1097/CM9.000000000000665

epidural space. Next, 6 and 3 mL of 2% lidocaine were injected. The cesarean section was uneventful. Transfusion, dieresis, and other support methods were used to maintain electrolyte homeostasis during the operational and post-operational periods. The patient could lay down in the supine position without dyspnea on post-operative day (POD) 1, but laboratory tests indicated that her situation had worsened (NT-BNP: 24,600 pg/mL, albumin: 22.3 g/L, BU: 14.38 mmol/L, Cr: 212 mmol/L, and potassium: 4.85 mmol/L). She complained of weakness of the bilateral lower limbs on the morning of POD 1 (15 h after epidural anesthesia). A neurologic examination demonstrated 3 of 5 for strength and numbness in the bilateral lower limbs. She had normal sensations of touch, pin prick, and temperature. Magnetic resonance imaging (MRI) of the lumbar spine was unremarkable.

A full immunologic investigation on the day of hospital admission revealed positive antinuclear antibodies (ANA +), immunoglobulin G (IgG): 15.10 g/L, immunoglobulin A (IgA): 1.75 g/L, complement C3: 0.38 g/L (normal range 0.79–1.52), complement C4: 0.06 g/L (normal range 0.16– 0.38), anti-nRNP/Sm antibody (++), anti-Sm antibody (+), and anti-dsDNA (+++) on the afternoon of POD 2. A rheumatologist was consulted. The patient complained of a facial skin rash and a frostbitten rash on both hands for 10 months, and she had lost her hair in the past with no joint swelling. According to her history and lab tests, she was confirmed to have systemic lupus erythematosus (SLE) and was immediately intravenously administered methylprednisolone and hydroxychloroguine. After the corticosteroid and hydroxychloroquine infusion, she experienced a mild improvement in strength. On POD 5, when she transferred from the obstetric department to the nephrology department, the strength in her lower limbs was 4 of 5. On POD 7, her strength recovered to 5 of 5. She experienced serositis, nephritis, seizures, psychosis, and

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Chinese Medical Journal 2020;133(5)

Received: 29-08-2019 Edited by: Li-Min Chen

lung infections on the following days. Although all possible treatment methods were used, including continuous renal replacement therapy, antibacterial therapy, and plasmapheresis, her condition continued to deteriorate, and she died on POD 38.

Neuraxial blockade is first choice of anesthesia for pregnancy, regardless of whether it is for natural delivery or cesarean section. Complications of neurological deficits, such as reduced limb function, lasting numbness, and paralysis are rare but devastating. The common causes, including physical injury, epidural hematoma, and infection, are primarily considered first, but neurotoxicity from local anesthetics and transient neurologic syndrome (TNS) must also be considered. Neurologic complications may occur early if related to traumatic catheter insertion or can occur later in the post-operative course if caused by catheter-related spinal space-occupying lesions such as epidural hematoma or epidural infection. In this patient, physical trauma was excluded since she did not complain of pain or paresthesia when the catheter was inserted. Epidural hematoma was also excluded because she exhibited no back pain or signs on the MRI. Infections were also excluded since the patient presented no fever, back pain, nuchal rigidity, or leukocytosis. Although 2% lidocaine can lead to morphologic damage in rats, clinical reports in humans are lacking. TNS was also excluded because of the absence of back pain.

Systemic diseases that can damage the nervous system are easily overlooked when neurologic deficits occur after epidural anesthesia. SLE is an autoimmune multisystemic disease that occurs predominantly in fertile-aged women. Owing to medical advances, the number of patients with SLE who become pregnant has increased worldwide, and most pregnancies are successful. The neuropsychiatric involvement of SLE, especially of the peripheral nervous system (PNS), has been scarcely studied^[1,2] despite its association with significant morbidity and a worsened quality of life.^[2] This is the first report of PNS-SLE in pregnancy. According to existing research, [2] the most common clinical presentations of PNS-SLE are distal axonal sensory or sensory-motor polyneuropathy with acute or subacute onset, which are similar to the symptoms of neuraxial blockade complications. In most cases, the onset is dramatic with sudden weakness in different nerve territories. In this case, no identical symptoms manifested before lidocaine was infused into the epidural space. Whether the local anesthetics aggravated the damage to local vessels or induced the nerve lesion directly is difficult to determine. Martinez-Taboada and colleagues^[3] described 2 SLE patients with severe mononeuritic multiplex secondary to necrotizing vasculitis of small- and mediumsized vessels. One patient presented with neurologic involvement at the disease onset, and the other later in

the disease after discontinuing steroid and chloroquine treatment.

Distinguishing pregnancy-associated signs and symptoms from those of SLE can be difficult. Fatigue, mild arthralgia, hair loss, dyspnea, headaches, edema, anemia, and thrombocytopenia are common ambiguous manifestations.^[4] In this case, the obstetrician failed to pay attention to the facial skin rash and frostbitten rash on both hands for 10 months, obvious hair loss, fatigue, and anemia and proteinuria found in the second trimester, and the missed diagnosis eventually led the patient's death. Hence, involvement of and assessment by experienced physicians is important during pregnancy with multiple symptoms.

What we can learn from this case is that: (1) if lesions involve multiple organs during pregnancy, experienced physicians should be consulted as soon as possible and (2) if post-epidural numbness or weakness of the extremities occurs, common complications and systemic diseases should be considered simultaneously, especially those that can damage the nervous system.

Declaration of patient consent

The authors have obtained all appropriate patient consent forms. In the form, the husband of the patient has given his consent for the clinical information of the patient to be reported in the journal. The husband of the patient understand that their name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflicts of interest

None.

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How to cite this article: Wu L, Jiang XQ, Xiong YQ, Lin XM. Muscle weakness of the lower limbs after epidural anesthesia in a pregnant woman with undiscovered systemic lupus erythematosus. Chin Med J 2020;133:621–622. doi: 10.1097/CM9.000000000000665