

CASE REPORT

Postoperative Paraplegia as a Result of Undiagnosed Primitive Neuroectodermal Tumor, Not Epidural Analgesia

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Postoperative paraplegia is a rare complication after epidural analgesia and often occurs with spinal hematoma or cord injury. We present the case of a 16-year-old girl who suffered from a tumor mass in the neck and abdomen who underwent gynecologic operation. Preoperatively, liver metastasis was found by computed tomography. Pathologic findings revealed that the abdominal mass was an ovarian dermoid cyst. After the operation, the patient complained of paraplegia while receiving epidural analgesia for postoperative pain control. A peripheral primitive neuroectodermal tumor in the thoracic and lumbar spines with spinal cord compression was later detected using magnetic resonance imaging. Learning from this case, we suggest that when a patient is preoperatively diagnosed with tumor metastasis, back pain and soreness, spinal cord compression from tumor metastasis should be excluded before epidural analgesia is implemented. [*J Chin Med Assoc* 2007;70(10):456–459]

Key Words: epidural analgesia, paraplegia, primitive neuroectodermal tumor, spinal metastasis

Introduction

Epidural analgesia is commonly used to provide postoperative pain relief and is seldom associated with complications such as paraplegia. We present here the case of a girl who was initially diagnosed with ovarian cancer with liver metastasis who suffered from postoperative paraplegia while receiving epidural analgesia. An emergency magnetic resonance imaging (MRI) examination was performed and, surprisingly, an undiagnosed peripheral primitive neuroectodermal tumor (PNET) with intraspinal extension was detected. It turned out that the irreversible paraplegia was caused by tumor compression, not by the epidural analgesia maneuver *per se*. In this article, we review the literature and focus on the differential diagnosis of postoperative paraplegia and its significance in our case.

Case Report

A 16-year-old girl (46 kg in weight, 161 cm in height) had suffered from a full abdomen and a poor appetite for about 2 weeks, and came to our hospital for evaluation. Physical examination showed a firm distended abdomen with palpable right inguinal lymph node and left neck mass of about 6 cm in diameter. Laboratory data revealed elevated tumor markers as follows: lactic dehydrogenase of 1,428 U/mL, α -fetoprotein of 9.4 ng/mL, cancer antigen-125 of 154 U/mL and estradiol of 74.1 pg/mL. Ultrasonography displayed a left heterogeneous adnexal mass of about 15 cm in diameter, and abdominal computed tomography (CT) detected a cystic tumor in the middle and lower abdomen that measured about 18 cm in diameter, with metastasis to the liver and lymph nodes. Chest X-ray

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E-mail: d94845002@ntu.edu.tw • Received: August 21, 2006 • Accepted: May 7, 2007

was unremarkable except for spinal scoliosis. Ovarian cancer was then suspected and a debulking operation was scheduled.

In the operating room, the patient was intravenously given 100 µg fentanyl, 210 mg thiamylal, and 80 mg succinylcholine after preoxygenation. General anesthesia with tracheal intubation was maintained with 2–3% sevoflurane in oxygen and atracurium (0.2–0.5 mg/kg·hr, intravenously). The operation was uneventful, and the tumor mass was excised. Before emergence from anesthesia, epidural catheterization was implemented for postoperative patient-controlled analgesia (PCA). The epidural catheter was smoothly inserted via a midline approach at T12–L1, 3 cm in the epidural space and 5 cm beneath the skin. The epidural space was located using the loss-of-resistance technique. Neither blood nor cerebrospinal fluid was found during the procedure of epidural catheterization. The PCA formula was 100 mL of 0.5% bupivacaine and 750 µg fentanyl in 500 mL of 0.9% normal saline. The PCA pump gave a continuous dose of 3.5 mL, bolus dose of 3.5 mL, lockout time of 12 minutes, and a 4-hour limit of 40 mL. The patient's vital signs were within normal range during the 4.5 hours of the operation. The endotracheal tube was extubated after the effect of muscle relaxant had been reversed.

During the period of recovery, blood pressure, heart rate and SpO₂ (<40% FiO₂) were around 124/90 mmHg, 114 beats/min and 99%, respectively. Although the patient could still move her legs under epidural PCA, progressive numbness and weakness in her lower limbs were noted. Before she left the recovery room, a sensory block up to the level of T7 dermatome

was noted. Under suspicion of suffering from the side effects of epidural PCA, the epidural catheter was removed and the PCA program was terminated. The total PCA dosage was 31.8 mL (31.8 mL of 0.1% bupivacaine and 47.7 µg fentanyl). However, the symptoms had not improved 3 hours later.

A neurologist was consulted and emergency MRI was performed to rule out spinal hematoma or cord injury. Some left paravertebral soft tissue from T1 to T7 was found, which had caused spinal cord compression with extension into the neuroforamen of T4–6. A mass over the anterior paravertebral area of the L-spine and bone metastasis of T4–5 were also noted (Figure 1). Pathology revealed metastatic PNET with dermoid cyst of ovary. We retraced the patient's medical history and found that low back pain had persisted for about 2 months and right leg weakness for 3 days. Spinal cord compression of T4–6 could have been the major cause of postoperative paraplegia, not epidural PCA. Surgical intervention to relieve the spinal cord compression was not suggested because of multiple metastases and tumor invasion to the aorta. The patient received palliative treatment with chemotherapy and local radiotherapy. There was no improvement in her neurologic condition 6 months after the episode.

Discussion

Paraplegia is a rare but devastating complication after epidural anesthesia or analgesia. When postoperative paraplegia occurs, the anesthetic method, surgical procedure and preexisting disease should all be considered.

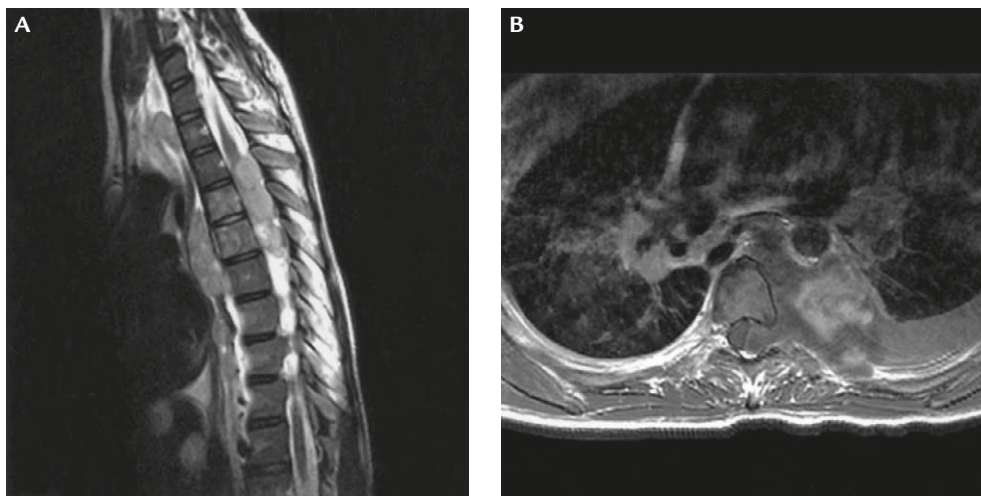


Figure 1. Magnetic resonance imaging of the thoracic spine: (A) sagittal T2; (B) axial T2. After enhancement over T4 and T5 vertebral bodies, bone metastasis was considered. Multiple lobulated masses about 5 cm in diameter are seen over left paravertebral soft tissue from T1 to T7 with extension into the neuroforamen of T4–5 and T5–6, which gathered over the left epidural space.

Paraplegia may result from spinal hematoma, intracord injection, chemical toxicity or epidural air after regional anesthesia.¹⁻⁴ Surgery-related paraplegia from reduced perfusion to the spinal cord has occurred in patients undergoing thoracotomy.^{5,6} Preexisting diseases, including spinal metastasis,^{2,7-10} thoracic disk herniation,¹¹ and transient anterior spinal artery syndrome,¹² should be considered in postoperative paraplegia. In this case, the postoperative paraplegia was related to peripheral PNET, a preexisting disease with multiple metastases. Unfortunately, it was not diagnosed before the gynecologic operation was performed. The possible reasons for the misdiagnosis are manifold. First, the symptom of back pain had been neglected because of the coexisting scoliosis in this patient. Although tumor metastasis to her neck had been noted, a full-scale bone scan was not performed to exclude the possibility of bone metastasis. Neurologic symptoms of weakness and soreness in her legs had developed before the operation, but the patient did not mention them to the doctors. Otherwise, it would have been an alert to doctors to initiate a detailed neurologic examination preoperatively. Lastly, a thorough neurologic examination should have been done before regional anesthesia or epidural analgesia was initiated.

Postoperative paraplegia may also result from spinal metastases from colon cancer,^{2,7} prostate cancer,^{8,10} or endometrial cancer.⁹ The reported cases in the literature were similar to ours and were also unrecognized preoperatively. Furthermore, spinal cord compression from epidural metastases is associated with lung cancer, breast cancer, lymphoma, myeloma, prostate cancer, or sarcoma,¹³ but rarely related to peripheral-type PNET. The first case of PNET was reported in 1975, and afterwards, only a few cases of peripheral PNET (including another 1 in Taiwan) have been reported.^{14,15} Peripheral PNET usually occurs in children or young adults, with rapid progression to the spinal cord, bone, liver, and lungs. Patients with peripheral PNET may complain of back and radicular pain and leg paresis. Treatments include surgery, radiotherapy and chemotherapy, but there is a high incidence of local recurrence and high mortality.¹⁴

Kararmaz et al have suggested that the preexisting pathology of the spine should be considered as a differential diagnosis of acute postoperative paraplegia.⁹ But in our case, a more interesting question could be, "Does epidural analgesia worsen spinal cord compression?" de Medicis and de Leon-Casasola believed that epidural analgesia might have worsened the cord compression in their patient who experienced reversible paraplegia caused by T11 vertebra bone metastasis after cessation of lumbar epidural infusion.² In our

case, cord compression was at the level of T4-6 and far from T12-L1, where the epidural catheter was inserted. In addition, postoperative paraplegia in our patient continued to proceed even after the epidural catheter had been removed.

It is also possible that continuous infusion through an epidural catheter may change the compliance of the epidural space and might produce compression forces on the spinal cord when the finite epidural space is surrounded and invaded by a large tumor. If changed compliance of epidural space can explain the exacerbation of spinal cord compression, an irreversible paraplegia may occur after epidural PCA due to the low compliance of epidural space compressed by the tumor. We therefore suggest that the use of epidural PCA should be cautious or even avoided in patients with preoperative evidence of malignancy with multiple metastases or epidural spinal cord compression. To our knowledge, this is the first case of postoperative paraplegia associated with peripheral PNET in a patient receiving epidural PCA for postoperative pain control. What we have learned from this case is that peripheral PNET may be easily overlooked and masked by other coexisting diseases (in this case, ovarian dermoid cyst and scoliosis). In gynecologic patients with back pain, numbness or weakness in the lower extremities, careful neurologic evaluation should be performed before epidural PCA is considered.

References

1. Tryba M. Epidural regional anesthesia and low molecular heparin: Pro. *Anesthesiol Intensivmed Notfallmed Schmerzther* 1993;28:179-81. [In German]
2. de Medicis E, de Leon-Casasola OA. Reversible paraplegia associated with lumbar epidural analgesia and thoracic vertebral metastasis. *Anesth Analg* 2001;92:1316-8.
3. Moen V, Dahlgren N, Irestedt L. Severe neurological complications after central neuraxial blockades in Sweden 1990-1999. *Anesthesiology* 2004;101:950-9.
4. Mayall MF, Calder I. Spinal cord injury following an attempted thoracic epidural. *Anaesthesia* 1999;54:990-4.
5. Short HD. Paraplegia associated with the use of oxidized cellulose in posterolateral thoracotomy incisions. *Ann Thorac Surg* 1990;50:288-9.
6. Mato M, Perez A, Otero J, Marquez C, Mora R, Torres LM. Paraplegia after left thoracotomy in a patient carrying an epidural catheter. *Rev Esp Anesthesiol Reanim* 2001;48:89-92.
7. Hirlekar G. Paraplegia after epidural analgesia associated with an extradural spinal tumor. *Anaesthesia* 1980;35:363-4.
8. Graham GP, Dent CM, Mathews P. Paraplegia following spinal anaesthesia in a patient with prostatic metastases. *Br J Urol* 1992;70:445.
9. Kararmaz A, Turhanoglu A, Arslan H, Kaya S, Turhanoglu S. Paraplegia associated with combined spinal-epidural anaesthesia caused by preoperatively unrecognized spinal vertebral metastasis. *Acta Anaesthesiol Scand* 2002;46:1165-7.

10. Bessac B, Levy R, Chauvin M. Paraplegia after spinal anesthesia. *Ann Fr Anesth Reanim* 1992;11:218–20.
11. Matsuura JA, Makhoul RG, Posner MP, Smith J, Litwack RS. Intradural herniation of a thoracic disc causing paraplegia coincident with epidural anesthesia. *Anesth Analg* 1997;84:922–3.
12. Crystal Z, Katz Y. Postoperative epidural analgesia and possible transient anterior spinal artery syndrome. *Pain Practice* 2001;1:372–3.
13. Spinazze S, Caraceni A, Schrijvers D. Epidural spinal cord compression. *Crit Rev Oncol Hemat* 2005;56:397–406.
14. Fabre E, Guillevin R, Chretien F, Le Guerinel C, Duffau H. Peripheral primitive neuroectodermal tumor of the cauda equina in an elderly patient. Case report. *J Neurosurg Spine* 2006;5:68–71.
15. Juan CC, Hung GY, Chen YJ, Lee PC. Coexistence of peripheral primitive neuroectodermal tumor and tetralogy of Fallot. *J Chin Med Assoc* 2006;69:134–7.