Delayed Incomplete Paraplegia After En Bloc Spondylectomy of Thoracic Metastasis, A Case Report

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Case report

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Abstract

Background: Delayed paraplegia after total en bloc spondylectomy (TES) is exceptionally rare, and from the best of our knowledge, no such cases were reported yet.

Case presentation: We herein report a case of delayed paraplegia which happened after TES of a solitary thoracic metastasis from renal clear cell carcinoma. After thorough evaluation, the patient received TES to expect a long-term local control. In operation, the patient suffered a short period of low blood pressure (mean synthetic pressure < 90 mmHg) which happened during vertebral prosthesis implantation. After operation, the neurological status was just as preoperative level, however, the patient suffered incomplete paraplegia 10 hours later. His symptoms and clinical examinations were consistent with infarct of anterior spinal artery and ischemia syndrome of spinal cord. The patient was given hyperbaric oxygen therapy and rehabilitation exercises after the tragedy, and he got recovery 2 months later, and showed much better than preoperative status 12 months later. Possible reasons for the delayed paraplegia were discussed, and we carefully concluded that intraoperative hypotension and manipulation both contributed to the happening of anterior spinal artery infarction.

Conclusions: Based on this case and literature review, keeping blood pressure above daily level and other precautionary measures for spinal cord ischemia were recommended.

Introduction

Vertebral metastasis is a big challenge for clinicians. Surgery is considered when spinal cord function can be saved and long-life expectancy is expected [1]. For solitary vertebral metastasis, TES provides the best chance to achieve long-term local control, preserve spinal cord function and improve quality of life [2,3]. Generally, two steps are required to achieve TES: a posterior en bloc laminectomy, then en bloc corpectomy with an oncological wide margin. Finally, a vertebral prosthesis is implanted to reconstruct the spine [2]. Even though, it is a aggressive procedure with possible serious complications, such as blood transfusion, big vessels injury, nerve injury, et al [4]. From the current literature, spinal cord is circumferentially decompressed, and spinal column is shortened after TES, which does not influence much on blood supply of spinal cord, neurological status was generally well preserved [5,6], even though neurological deteriorations immediately after surgery were reported [7], delayed paraplegia was not reported. Here we presented a case with solitary thoracic vertebral metastasis from renal clear cell carcinoma. The patient suffered incomplete paraplegia 10 hours after TES. The reasons for that tragedy were carefully discussed in this paper, and a couple of minutes of low blood pressure during operation was deemed to be the main causing factor. While low mean systemic blood pressure was reported to be one of the reasons for cerebral and spinal cord ischemia [8,9], to the best of our knowledge, there are no reports of delayed paraplegia after TES of thoracic metastasis.

Case Report
A male patient, 67 years old, 10 years ago, his left kidney was partially excised due to renal clear cell cancer. No radiotherapy and chemotherapy were given after surgery. He complained progressively weakness in both lower extremities in the past 3 months. The patient declined any other previous medical history or allergy.

Physical examinations: Neurological status was C on the left, D on the right according to the American Spinal Injury Association Impairment Scale [10]. In details, superficial feeling deficit was found below the nipple horizontal level (T4 innervation). Strengths of major muscles of lower limbs were obvious weak, iliopsoas: left /, right /; quadriceps: left and right /; tibialis anterior: left /, right /; Patella reflex and Achilles reflex were overactive in both sides. Babinski sign was positive at both sides.

Medical images: from the anterior posterior (AP) film of full spine X-ray, left pedicle and pars cannot be seen clearly at T4 level (Fig 1a, b). Osteolytic lesion of left pedicle, vertebral body and pars at T4 in both sagittal, coronal and axial transverse images of thoracic spine CT scans can be found. A soft tissue mass without obvious boundary and calcification can be seen, and the tumor mass already invaded into spinal canal, left side of spinal cord was seriously compressed (Figure 1c, d, e). Relatively low signals were found in the T1 and T2 weighted MR images of T4 vertebrae, while complex signals can be seen in fat tissue suppressed images. Spinal canal at T4 level were occupied by tumor tissues and spinal cord was seriously compressed in the left (Figure 1 f, g, h, i). From positron emission tomography (PET/CT) images (Figure 2), solitary metastatic tumor was highly suspected at T4 vertebrae and its appendix, probably from left kidney, however, no primary and other metastatic lesions were found. Needle biopsy was done under fluoroscopy, and pathological findings confirmed the metastasis was from renal cell carcinoma.

**Operation**

According to Tomita stages and evaluation [11], the patient was scored 3/10, which suggested wild or marginal excision was needed to achieve a long-term local control. Before operation, preoperative embolization was executed to embolize the feeding arteries of spinal metastases, and no neurological deterioration was found after that procedure. Then T4 vertebral TES was undertaken through posterior approach, and 4cm length of T4 ribs were sacrificed as well as two pairs of T3 and T4 nerve roots. In details, pedicles screws were implanted 3 levels up and below the targeted level respectively, then 4 cm of T4 ribs adjacent to thoracic spine were separated and removed. Then paravertebral soft tissues were bluntly dissected to lateral side of vertebral bodies, and two hands were gently put around the targeted vertebrae and fingers touched in the front. After two protective plates were put on each side, right pedicle was cut off with a fretsaw, while left pedicle was eroded by metastasis and broken, thus the total vertebrae annex was removed. Both sides of T3 and T4 nerve roots were cut off and ligated at the ventral side of dura, so as to retract spinal cord and reduce the tension. After temporary rods were implanted on both sides to certify stabilization, T4 vertebrae was isolated by removing the up and below intervertebral discs and cutting off the anterior and posterior longitudinal ligaments, then the vertebrae was removed in one block. After cartilage of endplates was cleared, an appropriate size of titanium mess cage was
implanted with allograft, and spinal reconstruction was finished. Dura sac was intact and spinal cord compression was not found.

Besides, it's worth mentioning that during the period of vertebral prosthesis (WEGO, China) installation, blood pressure decreased to 70/40mmHg for 5-7 minutes. Then, blood pressure increased to normal level shortly after giving norepinephrine. After operation, allergic skin rashes were found on the skin of the whole body. We suspected the low blood pressure in operation was resulting from the allergic reaction to the prophylactic antibiotics (second-generation cephalosporins, Cefuroxime Sodium), because blood pressure decreased just shortly after the second time use of the drug.

**Post operation**

Immediately after surgery, major muscles’ strength of lower limbs was same as preoperative level. However, 10 hours later, muscle strength of lower extremities progressively got weaker, finally reduced to 0/Ⅳ, whereas, superficial feeling (temperature and pinprick sensation) below T4 innervation was intact (American Spinal Injury Association, ASIA B). Concerned with the patient's history of intraoperative hypotension, spinal cord ischemia was considered due to hypoperfusion, and albumin and suspended red blood cells were supplemented. 3 days later, the power of major muscles did not show any improvements. Postoperative pathological results confirmed the diagnosis of bone metastasis from renal clear cell carcinoma.

After discharge, he was suggested to have hyperbaric oxygen treatment (23% O2) in a rehabilitation department of a local hospital. 10 days after surgery, that was 5 days after treatment of hyperbaric oxygen (90 minutes each day) the muscle strength of right lower limb recovered to the preoperative level (ASIA D), however, the left side recovered poorly (ASIA C). However, the sensation and movement of lower limbs improved day by day, and he can walk without assistance for 400 meters 12 months later. Until the end point of follow up, that was 40 months after operation, no local recurrence and new metastasis reoccurred, defecation and urination were as usual, while muscle tone was slightly elevated (ASIA D).

**Discussion**

From his previous renal cancer history, T4 vertebral metastasis were highly suspected, which was furtherly verified by needle biopsy pathological results. Symptoms, clinical examinations, and medical images were all consistent with that solitary metastasis from renal carcinoma, which caused neurological deficits below the lesion site. About the surgery indications for vertebral metastasis, palliative decompression surgery can be suggested in the treatment of acute spinal cord compression for relieving symptoms and improving quality of life [11,12]. For more invasive surgery, e.g., en bloc resection of tumor, should be only considered when solitary lesion site is present and optimistic prognosis is expected [13,14]. To determine the most reasonable treatment plan for the patients with metastasis, Tomita's Scoring System are widely used to assess the patient's prognosis [11] [15]. According to the scoring system, grade of malignancy of the primary tumors, visceral and bone metastases are overall considered.
About this patient, he was scored 3/10, which indicated he probably had a long-life expectancy, and the aim of surgery was long term local control, to achieve this, TES is the most promising method.

What was confusing was that the muscle power of lower limbs started to get paralyzed 10 hours after operation. Though, no similar delayed paraplegia were reported before, the most likely reasons for paraplegia after TES are spinal cord injury, epidural hematoma formation, and spinal cord ischemia [16-18]. In operation, special attention was paid to protect spinal cord, all procedures close to it were very careful, and no injury of spinal cord was noticed, which was verified by the fact that neurological status immediately after operation was just as the preoperative situation, and intraoperative spinal function monitoring may help identify spinal cord injury in time [19]. Concerning the discharge from drainage tube was about 1000ml on the first day after operation, so it was obviously unobstructed. Furtherly, no spinal cord compression was found in emergent CT scan images. Therefore, epidural hematoma and other spinal cord compressive factors were excluded. Therefore, spinal cord ischemia may be the main reasons for postoperative paralysis. It is well known that blood supply for spinal cord in thoracic segment is mainly from anterior spinal artery with poor collateral circulation [20], and the blood supply is easy to be influenced by atherosclerosis, operative maneuvers, blood pressure and vasoconstriction. In this case, a pair of segmental arteries had to be embolized before operation to reduce blood loss, which may reduce blood supply to the target spinal cord but should not be significantly, and no neurological deterioration was found in this case after that procedure. For TES, preoperative embolization was reported to be safe and can reduce intraoperative blood loss significantly, however, paraplegia arising from the procedure was not reported [20-21]. Except for one pair of embolized segmental arteries, operators confirmed no damage to other blood supplies. Besides, blood pressure also plays an important role in blood supply of spinal cord [8], when we think of the issue of low blood pressure (70/40mmHg) did happen after the use of cefuroxime sodium during vertebral prosthesis implantation, which was a delicate moment for spinal cord, because manipulation, low blood pressure and preoperative embolization may together reduce the blood supply to spinal cord. Now therefore, it is highly suspected that on the basis of atherosclerosis, antibiotics or other medicines caused anaphylactic low blood pressure, at the same time, operations may interrupt the circulation of spinal cord, they both together led to thrombosis of anterior spinal artery. Even worse, norepinephrine and fluid supplementation were given to elevate blood pressure, when blood pressure got elevated, ischemia-reperfusion injury may furtherly damage spinal cord, and Riluzole was substantiated to reduce this injury [22]. Physical examinations found incomplete paraplegia, that was complete loss of power but without sensory abnormalities and without sphincter dysfunction, which were consistent with syndrome of anterior spinal artery infarction (pseudo-polymyelitic form), because of ischemia of anterior horns [23]. Several top experts in surgical treatments of vertebrae metastasis were consulted about this case, for example, Boriani, Jianru Xiao, etc., who all agreed that it may be caused by intraoperative hypotension. Therefore, blood pressure must be kept at an appropriate range during TES of thoracic metastases.

Allergic rashes were found after operation, and cefuroxime sodium may be the most likely allergen, even though preoperative skin testing was negative. That is because degradation of cephalosporins are
multiple and unstable antigenic fragments which are not fully understood. Without validated reagents and reference standard for cephalosporin skin testing, false-negatives or positives are inevitable [24,25].

**Literature review**

After TES, the maximum rate of neurological deterioration was reported to be 29%, and preoperative neurological deficits, postoperative cerebrospinal fluid leakage, and surgical site infections were risk factors for poor activities of daily living (ADLs) after surgery [26]. However, to the best of our knowledge, no cases of delayed paraplegia after TES were reported. Except for epidural hematoma formation, the interruption of blood supply to spinal cord should be fully alert. For this case, the feeling deficit after surgery was termed conductive tract dissociative sensory disorder, which are typical symptoms of embolization of anterior spinal artery [23]. Neurosurgeons always think that appropriately lowered blood pressure can reduce bleeding and improves the surgical field, which may be true [27]. However, it is a dilemma in this case, low blood pressure was deemed to be the main reason for symptoms of spinal cord ischemia, and a couple of minutes of low blood pressure may result in spinal shock. Therefore, elevating mean systemic blood pressure is an effective method of increasing blood flow to spinal cord [8]. From our experiences, mean systemic blood pressure should be kept above 100mmHg during perioperative period. Intraoperative monitoring of somatosensory evoked potentials (SSEP) during surgical procedures may help identify ischemia of spinal cord in time. Continuous monitoring amplitude and latency of SSEP, and an increase of 10% latency was reported to be a sign of reduction of spinal cord perfusion [19].

Before operation, preconditioning hyperbaric oxygen can induce tolerance of spinal cord against ischemia and attenuate early apoptosis of spinal cord cells by upregulation of antioxidant enzymes, which was verified in rabbits and rats [28-29]. To speak of the rehabilitation measures, hyperbaric oxygen treatment may play a role in function recovery of spinal cord after ischemia [30]. About this case, hyperbaric oxygen treatment was started 3 days after operation, 90 minutes each day, and obvious improvements of both lower limbs’ power could be seen (from ASIA B to D), while it still needs more data to evaluate its effectiveness on spinal cord ischemia. Although there is widespread controversy, methylprednisolone is often used in acute spinal cord injury [31]. For this case, 30mg methylprednisolone per Kg of body weight was given intravenously within 15 minutes once neurological deterioration was found, and the medicine was used as reported in literatures [31]. Besides, dehydrate agents, neurotrophic and neuroprotective agents can be considered to protect and facilitate rehabilitation of spinal cord function, but there are short of consistent clinical evidence [22,32-33]. Although often overlooked, compensation collateral circulation, including the system of epidural arcades in the immediate setting, and paraspinal collaterals in the long run, may play a role in functional recovery of spinal cord [34]. Thrombus recanalization, even rare, may be another reason for the recovery of spinal cord function [35]. Therefore, spasmolytic, anticoagulants and other drugs which improve circulation also can be used in appropriate time.

**Conclusion**
Delayed paraplegia after TES is rare, herein, we presented a case who suffered incomplete paraplegia 10 hours after this surgery for solitary thoracic metastasis. In this manuscript, we analyzed the possible causes of this complication, and we found the patient’s symptoms were consistent with anterior spinal artery infarct syndromes. Therefore, a period of low systemic blood pressure caused by allergy of cephalosporins was supposed to be the main reason. So, we suggest keeping systemic blood pressure above 100mmHg during TES may help reduce incidence of spinal cord ischemia. From this case, and furtherly through literature review, we carefully concluded that hyperbaric oxygen may facilitate spinal cord function recovery from ischemia injury.

Declarations

I, (Dr. Feng Wei), certify that this manuscript is a unique submission and is not being considered for publication, in part or in full, with any other source in any medium.

Ethics This work got approval from hospital ethics committee, and we have obtained consent from the patient using The Lancet patient consent form.

All data and materials are available when asked.

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Declarations of interest The authors declare that they have no competing interests.

Author contributions Chuanchao Du: Conceptualization, Methodology, Writing-Original Draft, Submission. Shanshan Liu: Validation, Formal analysis, Visualization. Fei Jia: Visualization, Software. Xiaoguang Liu: Writing, Review & Editing, Supervision. Feng Wei: Writing, Review & Editing, Supervision.

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Figures
Figure 1

Evaluations of preoperative images. a. from the AP film of full spine, left pedicle cannot be seen clearly at T4 level (red arrow). Osteolytic lesions of left pedicle, partial vertebral body and pars at T4 are shown in sagittal (c), coronal (d) and axial view (e) of CT scan images. A tissue mass without obvious boundary and calcification occupied spinal canal, left side of spinal cord can not be seen (e, irregular domain delineated in red). Relatively low signals were found in the T1 (f) and T2 (g) weighted MR images of T4 vertebrae (red arrow), while complex signals can be seen in fat tissue suppressed images (i). Spinal canal at T4 level were occupied by tumor tissues and spinal cord was seriously compressed (i, irregular domain delineated in red).

Figure 2

Preoperative PET/CT images. Solitary metastatic tumor was shown at T4 vertebrae and its appendix, no primary and other metastatic lesions were found (red arrow).
Figure 3

Intraoperative photos and AP and lateral X-ray films. The spine was reconstructed with vertebrae prosthesis and internal fixation system (posterior view). X-ray films showed the pedicles screws and prosthesis were well located.