Rare Huge Epidural Hematoma Associated with Re-fracture of Kummell Disease: A Case Report and Literature Review

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Case report

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Abstract

Background Kummell disease combined with huge spinal epidural hematoma is a very rare phenomenon, and its potential pathogenesis and natural course remain unclear.

Case description We describe a rare case of Kummell disease with huge spinal epidural hematoma. A 75-year-old male was diagnosed with osteoporotic vertebral compression fractures and was treated conservatively. After suffering minor trauma again 8 days ago, he presented unbearable low back pain and activity restriction. Lumbar MRI showed that L1 vertebral had re-fracture and intervertebral vacuum cleft, and a huge spinal epidural hematoma extending from T12- L1. Due to the patient had no neurological deficits and unbearable low back pain, percutaneous vertebroplasty was performed, and pain was relieved significantly. The follow-up MRI showed that the hematoma almost disappeared 7 days after the operation.

Conclusion Although extremely rare, there is a possibility of spinal epidural hematoma after re-fracture of Kummell disease, and it could be further confirmed via MRI and pathological examination. Timely operation is recommended, and the results are usually favorable.

Introduction

With the advent of the global population's aging, more osteoporotic vertebral compression fracture occurs, often accompanied by severe acute low back pain and activity restriction1. Clinically, the symptoms of most patients can be relieved gradually after several weeks of conservative treatment2, but Kummell disease is a rare type of osteoporotic vertebral compression fracture, which gradually appears vertebral collapse and kyphosis, and its occurrence may be related to vertebral ischemic necrosis3. Huge spinal epidural hematoma (SEH) combined with Kummell Disease is extremely rare. The pathogenesis and natural course of SEH combined with Kummell Disease remain unclear. In this case study, we describe a Kummell patient with huge SEH and review the literature to explore its potential pathogenesis, natural course, and treatment.

Case Description

A 75-year-old male patient experienced a persistent low back pain and movement restriction after a slight fall 3 months ago. He was transferred from a traditional oriental hospital to the emergency room of our hospital. Here, X-ray examination showed that osteoporotic compression fracture of L1 (Fig. 1A). He was treated conservatively such as bed rest, analgesics, and oral calcium supplements. However, he was admitted to our hospital with suffered minor trauma again 8 days ago and complained of severe and unbearable pain in the lower back and impaired mobility. On examination, the patient presented with significant tenderness and percussion in the L1 spinous process and paraspinal, especially during flexion and extension. Neurological examination was negative and laboratory examination results were within the normal range. The patient reported no history of heart disease, cerebrovascular disease, or
hypertension. X-ray examination showed compression fracture of L1 vertebra and the compression degree was more than 50% (Fig. 1B), CT demonstrated that fracture of L1 characterized by vertebral vacuum cleft (Fig. 1C-D), MRI of the thoracolumbar spine revealed that collapse of L1 vertebra, vacuum cleft filled with fluid, and a huge soft tissue of ventral epidural of spinal cord extended from T12 to L1 on T1-weighted images showed low signal intensity, while T2-weighted images showed high signal intensity (Fig. 1E-F). The patient had severe osteoporosis with a T-score on the bone marrow densitometry of -3.80. So, he was considered as re-fracture of Kummell Disease. Since the intractable pain unrelieved by conservative treatment, a unilateral percutaneous vertebroplasty and intraoperative biopsy were performed, and then approximately 5 ml of bone cement carefully injected into the vacuum space of the vertebral body under X-ray examination. After the operation, the pain was relieved immediately, and the patient could walk independently with a lumbar brace. Postoperative X-ray showed that the bone cement diffused and filled well in the L1 vertebral body (Fig. 1G-H), and pathological examination demonstrated bone marrow tissue (Fig. 2). Postoperative MRI revealed that the hematoma almost completely disappeared on at 7 days follow-up (Fig. 1L-K).

**Discussion**

SEH usually occurs immediately after spinal trauma or surgery\(^4\). However, Kummell disease with huge SEH is a unique complication, which has been rarely reported since it was first reported in 2008\(^5\). It occurs more commonly in the conservative treatment of thoracolumbar osteoporotic vertebral compression fracture with or without neurological deficits\(^6\).

The pathogenesis and natural course of Kummell disease associated with SEH is still unclear. Several hypotheses have been used to explain it. One hypothesis\(^7\) is that there is a connection between the intravertebral cleft and epidural space in Kummell disease, so that during weight bearing by nonunion or re-fracture with dynamic mobility, the fluid including hemorrhage may be under pressure and pushed into the epidural space, resulting in SEH. Another theory\(^8\) is that local pooling of thin-walled epidural venous plexus may be ruptured due to a brief increase in venous pressure caused by nonunion or dynamic re-fracture. In our case, the patient suffered from the aggravation of low back pain caused by minor trauma again and MRI suggested evidence of re-fracture, so we believe that the formation of SEH may be caused by re-fracture.

Whether the patients of Kummell disease with SEH have neurological deficits remains to be elucidated. Kummell disease is a nonunion of fractures characterized by pseudoarthrosis and vacuum cleft\(^9\). We believe that whether the patient has neurological deficits may be related to the segment of Kummell disease, the size of the hematoma and the relative position of the hematoma to the conus medulla.

The treatment of Kummell disease with SEH depends on the following factors: the patient's general ability to withstand surgery, the presence of severe back pain, kyphosis, and the presence of neurological deficits. For patients of Kummell disease with huge SEH when their neurological intact, or the whole body is unable to tolerate surgery such as hematoma removal, percutaneous vertebroplasty, as a minimally
invasive treatment, not only can effectively alleviate pain, restore vertebral body height, reconstruction of spinal stability\textsuperscript{10}, but also fill the vacuum cleft with bone cement, thus blocking the connection between the vacuum cleft and the epidural space, helps to spontaneous absorption of the hematoma. For patients with neurological deficits, the prognosis is related to the degree of preoperative neurological impairment and the time to operation\textsuperscript{11}. Therefore, timely and adequate decompression is the key to achieve great surgical results. Kim et al\textsuperscript{12} adopted anterior thoracolumbar vertebrae resection and bone grafting, and posterior bone cement enhanced screw fixation to increase spinal stability. Oda et al\textsuperscript{5} performed vertebroplasty combined with posterior decompression, which provided satisfactory vertebral reconstruction and posterior decompression through a single posterior approach. In this case, the patient had no neurological deficits, and we performed percutaneous vertebroplasty. The patient recovered well immediately and the reexamination 1 week after operation showed that the hematoma almost completely subsided.

**Conclusions**

Although extremely rare, there is a possibility of SEH related to re-fracture of Kummell disease, and it could be further confirmed via MRI and pathological examination. Timely operation is recommended, and results are usually favorable.

**Abbreviations**

SEH, spinal epidural hematoma; MRI, magnetic resonance imaging; CT, computed tomography

**Declarations**

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**Authors’ contributions**

DZC, DJH, and WTW designed the study. JPD, LL, and XKX collected the data. WTW, JFW, and CYG performed the operation. LLB summarized the case data and wrote the paper. All authors read and approved the final manuscript.

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**Availability of data and materials**

All data generated or analyzed during this study are included in this article.
Ethics approval and consent to participate

This study was approved by the ethics committee of Xi’an Honghui Hospital, and written informed consent was obtained from the patient to publish the details of his case.

Consent for publication

A written, signed informed consent to publish all data and any accompanying images was obtained from the study participant.

Competing interests

The authors report no conflicts of interest in this work.

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