




CASE REPORT

Magnetic Resonance Imaging Negative Spine Trauma Followed by a Delayed Intravertebral Vacuum Cleft–Kümmell's Disease: A Case Report and Literature Review

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Background: Kümmell's disease (KD) is a rare clinical entity characterized by delayed post-traumatic vertebral body collapse, in which an intravertebral vacuum cleft (IVC) is formed. The patient sustained a minor spinal trauma, and their spine X-ray examinations were negative. However, after an asymptomatic period of months, they developed recurrent back pain because of the progressive vertebral collapse of the affected spine. However, no imaging examination could confirm the magnetic resonance imaging (MRI)-negative initial spinal trauma followed by delayed vertebral collapse with IVC.

Case Presentation: We report a case of KD occurring in a 66-year-old postmenopausal patient whose lumbar MRI on sagittal planes were immediately performed following the initial trauma (a ground-level fall) and revealed that the vertebral integrity or connectivity was not interrupted and the marrow signal was even. After an asymptomatic period of 8 months, the back pain reappeared and progressively exacerbated. The wedge-shaped change in the T11 vertebra with an IVC was confirmed by lumbar computed tomography and MRI. Finally, KD was diagnosed following extensive routine hematological and biochemical workups. Percutaneous kyphoplasty was performed to relieve her back pain by restoring her vertebral stability.

Conclusions: We confirmed that the OVF was not the first step in the KD sequence, and the IVC - KD - could from an initial MRI-negative spine trauma.

Key words: Avascular necrosis; Computed tomography; Delayed; Intravertebral vacuum cleft; Kümmell's disease; Magnetic resonance imaging

Introduction

Kümmell's disease (KD) is a rare clinical scenario first described by Kümmell in 1891, which sustained a minor spinal trauma, was essentially asymptomatic for months or years, and eventually developed a progressive and painful spinal kyphosis. The advent of X-rays confirmed that spinal kyphosis was the result of delayed vertebral collapse.

However, some authors subsequently thought that the fracture following minor spinal trauma was missed because of the poor quality of the radiographic studies. Subsequent studies confirmed that the initial X-rays were normal and vertebral collapse appeared only on the delayed films.^{1,2}

The key characteristics of KD diagnosis include a history of negative X-ray, "mild" spinal trauma, and a

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Received 23 March 2022; accepted 16 September 2022

recurrence of intolerable back pain from progressive kyphosis following an asymptomatic period of months.¹ Radiological signs of intravertebral vacuum cleft (IVC) are highly associated with KD.^{2–5} If KD is radiologically diagnosed, it would also represent delayed vertebral collapse.

Post-traumatic bone necrosis is the most supported pathogenetic hypothesis for developing KD,^{6,7} although the negative X-ray performance of the vertebra following trauma cannot confirm this standpoint. Other scientists believe the undisplaced fracture following a mild spine trauma is the initial factor for delayed vertebral collapse.⁸ However, since there are no imaging investigations between the time from the traumatic event to eventual vertebral collapse that can confirm either of these theories, the first step for delayed vertebral collapse remains unclear.

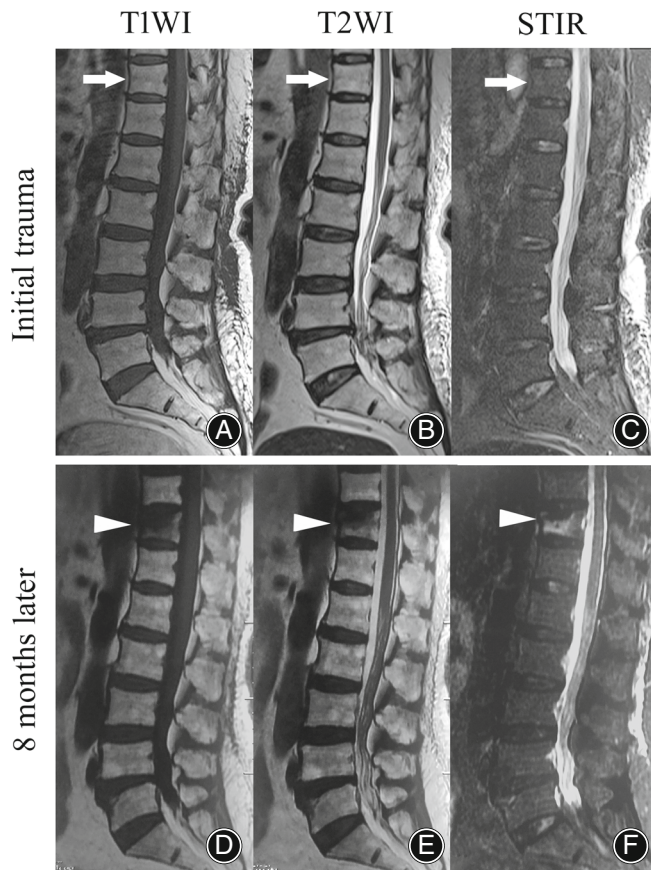


Fig. 1 Lumbar MRI on sagittal planes taken immediately after the trauma and 8 months later. The integrity and connectivity of the vertebra following the initial trauma are not interrupted, and the marrow signal is even. (A–C, arrows) After asymptomatic 8 months, the pain is onset again, all MRI sequences show the wedge-shaped changes of T11 vertebra, and the abnormally confined hypointensity area underneath the upper endplate (D–F, arrowheads), indicating the formation of IVC. **T1WI** T1-weighted image, **T2WI** T2-weighted image, **STIR** short-tau inversion recovery

Magnetic resonance imaging (MRI) is recommended for detecting the presence of an undisplaced fracture line or bone bruises within the vertebral body, which are difficult to visualize on conventional X-rays. To date, the literature has not reported on the initial negative MRI after mild spine trauma, followed by delayed IVC with a recurrence of back pain. We present a case of a 66-year-old female with KD, where negative MRI after spine trauma progressed to delayed IVC.

Case Report

This study was approved by the Ethical Committee of Tianjin Medical University General Hospital (No. IRB2022-WZ-060) and the patient provided written informed consent.

A 66-year-old female with no previous medical history complained of back pain in a local hospital after a ground-level fall. She requested an MRI examination to rule out osteoporotic vertebral fracture (OVF). Lumbar MRI demonstrated all the vertebral integrity or connectivity was not interrupted, and the marrow signal was even (Figure 1). She was told to receive conservative treatment, and her back pain subsided after bed rest for 1 week.

Eight months after that trauma, she developed back pain and returned to the local hospital. During the interim between the initial trauma and subsequent presentation, she resumed daily activities with no back pain or history of spinal trauma. She was sent home with conservative treatment

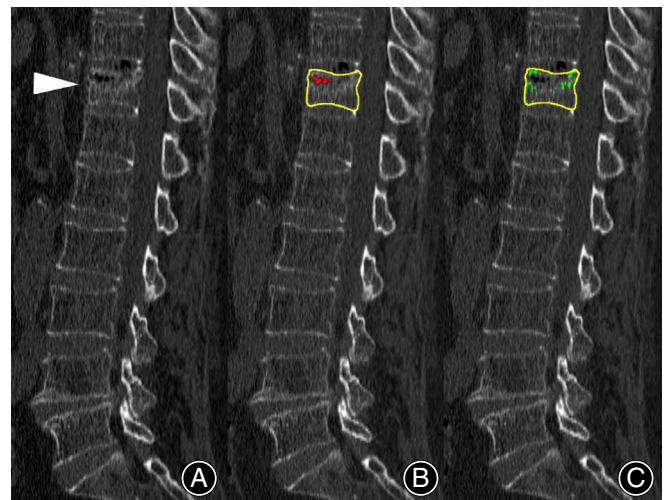


Fig. 2 Sagittal lumbar CT image analyzed by ImageJ. The original image of the T11 vertebra (A, arrow) analyzed by ImageJ with a CT value of -1000 HU indicates the IVC filled with gas (B, red color area) in the T11 vertebra (yellow outline). The same original image analyzed by ImageJ with a CT value of 300 – 1000 HU shows no continuous mineralized callus formation (C, green color area) inside or outside of the IVC, indicating the IVC nonunion. **HU** Hounsfield Unit, **IVC** intravertebral vacuum cleft

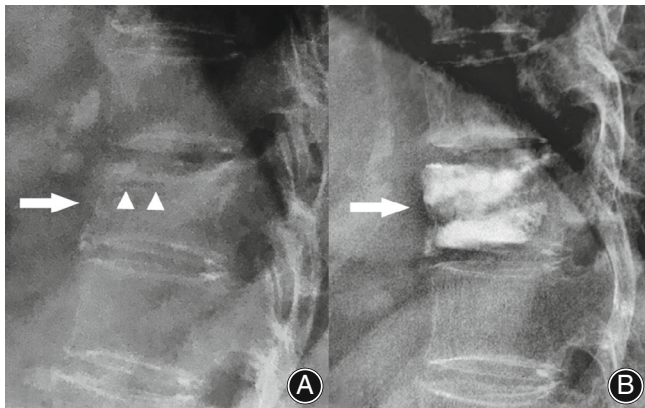


Fig. 3 Lateral view of spinal X-rays centered on T11 vertebra pre and post-operation on 3 days. The pre-operation X-rays indicate the IVC (A, arrowheads) within the wedge-shaped changes of T11 vertebra (A, arrow). The post-operation X-rays (B, arrow) show the restoration of T11 vertebral height, filling with bone cement without leakage.
IVC intravertebral vacuum cleft

of bed rest after an unremarkable workup. However, she was referred to us for further treatment 1 month later due to progressive back pain. Clinical examination revealed percussive pain of T11 spinous process accompanied by an apparent paravertebral spasm. Abnormally confined hypointensity area underneath the upper endplate was detected on all sagittal lumbar MRI sequences (Discovery MR750, General Electric, Milwaukee, WI, USA) of the T11 vertebra, including sagittal T1-, T2-weighted, and short-tau inversion recovery (STIR) images, indicating IVC formation (Figure 1). Lumbar computed tomography (CT) (Optima CT660/GE Medical Systems, USA) was performed to further determine the nature of IVC content and callus formation in and out of the IVC. The sagittal CT images were analyzed using ImageJ (Wayne Rasband National Institutes of Health, USA, Java1.8.0_112 [64-bit], <https://imagej.nih.gov/ij/>) with a CT value of -1000 Hounsfield Unit (HU) and $300-1000$ HU,⁹

showing the IVC with gas collection but no continuous mineralized callus formation (Figure 2).

After extensive workup, including routine hematological and biochemical parameters, KD was diagnosed. She was admitted to our hospital and underwent percutaneous kyphoplasty treatment. Her back pain was significantly relieved postoperatively. Postoperative X-rays on the 3rd day indicated restoration of the vertebral height of T11 vertebra filled with bone cement without leakage (Figure 3). Three months and one year after discharge, the patient was ambulated with no pain and performed portions of her housework.

Discussion

The case we presented here sustained a minor spinal trauma with negative MRI, after an asymptomatic period for 8 months, developed a recurrence back pain, as well as presented an IVC of the T11 on CT and MRI.

Illustrating the KD Definition

The advent of radiographic technology verified Hermann Kümmell's initial description of KD in 1891 that the patient underwent a minor spinal trauma essentially asymptomatic for weeks to months, then developed painful, progressive kyphosis. However, many would not accept imaging examination attributed to mild, essentially asymptomatic trauma.^{3,10} The modern concept of KD refers to a clinical scenario in which patients suffer from painful kyphosis because of delayed vertebral collapse after an X-ray negative minor spinal trauma.^{3,11} However, the literature on this subject remains limited and the cases of KD with initial X-ray-negative spine trauma since 1950 are documented in Table 1.

In this report, the patient fulfilled the criteria of KD-delayed post-traumatic vertebral collapse. She sustained a negative MRI minor spinal trauma, after an asymptomatic period for 8 months, developed a recurrence of back pain from the mild vertebral collapse of the affected spine.

TABLE 1 Literature on KD with initially X-ray-negative spine trauma since 1950

Studies	Age (y)/sex	Level	Initial imaging studies	Asymptomatic period	Treatment	Prognosis (back pain)
Steel ²¹ , 1951	23/male	T10	X-ray	2 months	Taylor back brace	Relieved
Nicholas ²² , 1981	56/female	T11	X-ray	2 months	Thoracolumbar brace	Relieved
Brower ³ , 1981	71/male	T12	X-ray	3 weeks	-	-
Hermann ²³ , 1984	45/female	L1	X-ray	4 weeks	-	-
Malzer ¹⁴ , 1992	60/male	T11	X-ray	1 year	Anterior interbody fusion	Relieved
Van ¹⁰ , 1993	75/male	T11	X-ray and bone scans	9 weeks	-	-
Bhalla ²⁴ , 1998	71/female	T12	X-ray	5 months	-	-
Osterhouse ²⁵ , 2002	79/male	L2	X-ray	6 weeks	-	-
Ma ⁵ , 2010	75/female	T12	X-ray	4 months	Vertebroplasty	Relieved
Formica ¹¹ , 2016	67/male	L1	X-ray	6 months	Ponteostomies and arthrodesis	Relieved

Special Radiographic Sign of KD

Radiological studies have confirmed a significant correlation between IVC and delayed post-traumatic vertebral collapse.^{6,12} In addition, the IVC with gas collection was in an advanced stage of KD,^{2,12,13} indicating impaired bone union. Histological examinations have also confirmed that impaired bone regeneration is associated with the vertebral collapse in KD.^{7,14,15} Commonly, the clinical symptoms in OVF patients gradually decrease with fracture union.¹⁶ The patient suffered from progressive back pain that was refractory to conservative treatment. The T11 hypointensity area on sagittal MRI of T1-, T2-weighted, and STIR images were quantified by CT and confirmed as an intravertebral gas collection. Lumbar CT images also confirmed no mineralized callus formation either in or out of the IVC, indicating the nonunion of IVC.

Controversy on the Pathophysiological Hypothesis of KD

The sequences of KD events is an initial trauma with negative X-ray, after a symptom-free period for several months, followed by persistent back pain with vertebral collapse. However, no imaging examination following the initial traumatic event can confirm the first step in the KD sequence was either vertebra avascular necrosis or OVF. As Laredo and Matzaroglou suggested,^{8,17} spinal MRI examination following initial trauma is vital for identifying the first step in the KD sequence. In the presented case, no signals related to OVF were detected on all MRI sequences following the trauma, excluding the controversy of OVF as the first step of KD sequence. The theory of post-traumatic avascular necrosis can better explain the sequences of KD events: osteonecrosis by itself is asymptomatic, and back pain appears only at the time of delayed vertebral collapse from chronic and durable avascular necrosis and resorption.

Treatment and Prognosis

IVC is a sign of intravertebral pseudarthrosis formation and stability, which is the main reason for intractable back and even neurologic dysfunction.¹⁸ Conservative therapy for patients with KD had little effect, and a percutaneous kyphoplasty is a widely accepted option for most orthopaedic surgeons to immediately ameliorate back pain by injecting bone cement into the IVC to eliminate intravertebral

instability,¹⁹ as described in our study. Both back pain and quality of life improved by the end of follow-up. Considering the pathological characteristics of this clinical entity, achieving fracture union may be a better way to eliminate back pain. Recently, 1-34PTH has been reported for KD treatment by boosting fracture union and preventing vertebral collapse.^{9,20} They found that 1-34PTH treatment boosted the union of IVC and prevented vertebral collapse by increasing vertebral strength.

Strengths and Limitations

The strength of the present study was that it was the first to exclude the theory of vertebral “microcracking,” indicating that avascular necrosis could better explain the sequences of KD events. The limitation of the study was that KD was not diagnosed until back pain reappeared. A bone scan, which is a sensitive and effective method for the diagnosis of early ischemic necrosis, was not performed following a traumatic event, which is another limitation in the present study.

Conclusions

We confirmed that the OVF was not the first step in the KD sequence and the IVC - KD - could from an initial MRI-negative spine trauma. Special attention should be paid to the early detection of vertebral avascular necrosis, especially in patients with negative spinal X-rays or MRI findings.

Acknowledgments

This work was supported by the National Natural Science Foundation of China (81871124 to Yuan Xue).

Conflicts of Interest

All the authors declare no conflict of interest.

Authorship Contribution Statement

Yuan Xue and Feng Chang designed the study. Pengguo Gou, Wanli Jing, and Jiaming Zhou drafted the manuscript. Rui Wang and Zun Wang performed the imaging analyses. Feng Chang and Yuan Xue contributed equally to this manuscript. Pengguo Gou, Wanli Jing, and Jiaming Zhou contributed equally to this manuscript.

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