


CASE REPORT

Consecutive Kummell's Disease Combined with Parkinson's Disease and Experienced Internal Fixation Failure: A Case Report and Literature Review

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Background: The continuous occurrence of Kummell's disease is extremely rare in clinical practice, and its treatment is difficult. The study aimed to present a rare case of consecutive Kummell's disease combined with Parkinson's disease (PD) and experienced internal fixation failure.

Case presentation: A 69-year-old female patient had a history of PD for 10 years, and was treated by posterior decompression, fixation, and fusion because of Kummell's disease of T12 with neurological damage. The patient's back pain and lower limb pain were significantly improved after surgery. Twenty-two months later, the patient was rehospitalized for Kummell's disease of L4 with neuropathic pain of left lower extremity. She received almost identical surgical procedures as T12 lesion, and the difference was no L4 vertebroplasty performed due to the fact that the L4 vertebrae collapse was not obvious, the intravertebral vacuum cleft (IVC) range was small, and the pedicle screw fixation strength was high. The pain symptoms were significantly relieved after operation. Unfortunately, there was a complication of internal fixation failure that occurred a month later, and a revision operation was carried out.

Conclusion: Osteoporosis combined with PD may lead patients to become prone to consecutive Kummell's disease, and patients are prone to experience failure of internal fixation. Bone cement filling of vertebral IVC and effective support of anterior vertebral column are very important procedures to ensure the clinical efficacy of treating Kummell's disease.

Key words: Bone cement; Osteoporosis; Parkinson's disease | Pedicle screw

Introduction

Kummell's disease, first described in 1891, is a rare and poorly documented disease. Patients had no symptoms for weeks to months after mild spinal trauma, but gradually became symptomatic, including experiencing back pain,

spinal canal stenosis, neurological deficits, spinal instability, progressive kyphosis, and even diminished quality of life.¹⁻³ Currently, the pathogenic mechanism of Kummell's disease is still not fully understood and no uniform standard surgical strategy exists. Conservative treatment is usually ineffective

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Grant sources: This work was supported by the Key Research Plan in Shaanxi Province of China (No. 2020GXLH-Y-003). Funding covered the cost of collecting the follow-up data and publication fee.

Disclosure: The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article. Received 25 August 2021; accepted 18 February 2022

and may delay the disease leading to neurological damage or severe kyphosis.^{4,5} Because of the reliable curative effect, surgical treatment has been highly praised by many scholars.

Traditionally, the vast majority of patients with back pain as the main symptom can often be treated with minimally invasive vertebroplasty (VP) or kyphoplasty (KP). It is the most important analgesic mechanism to restore physiological stability and eliminate motion at the intravertebral vacuum cleft (IVC) by the injection of bone cement for VP and KP.^{3,6,7} For patients with neurological impairment, it is often necessary to use open decompression, fixation and fusion, or even osteotomy.⁸⁻¹⁰ Because VP or KP alone cannot alleviate the neurological symptoms, and patients with neurological symptoms are often in the late stage of Kummell's disease, with serious collapse of the vertebral body and loss of spinal stability only using bone cement cannot achieve the surgical goal, and may have

complications such as fracture of adjacent vertebral body, leakage, and displacement of cement.¹¹⁻¹³

The continuous occurrence of Kummell's disease is extremely rare in clinical practice, and its treatment is difficult. If the failure of internal fixation occurs, the treatment will be trickier. In this study, we present the case of a patient with consecutive Kummell's disease accompanied by neurological impairment who suffered from failure of internal fixation after the second operation, and review the literature to better understand the clinicopathological features and the risk factors of internal fixation failure.

Case Presentation

A 69-year-old female patient presented to the clinic in January 2016 with low back pain and limited activity for 2 years, increased pain and weakness of both legs for



Fig. 1 Radiographs before the first operation. The patient experienced Kummell's disease in T12 vertebrae. (A, B): lateral and anteroposterior X-ray shows anterior column collapse; (C, D, E): coronal, sagittal, and transverse CT shows the IVC located below the upper endplate; (F, G): T1 sequence, T2 sequence, and transverse MRI shows the posterior tissue of vertebral body protruded into the spinal canal, resulting in corresponding segment spinal canal stenosis and spinal cord compression.

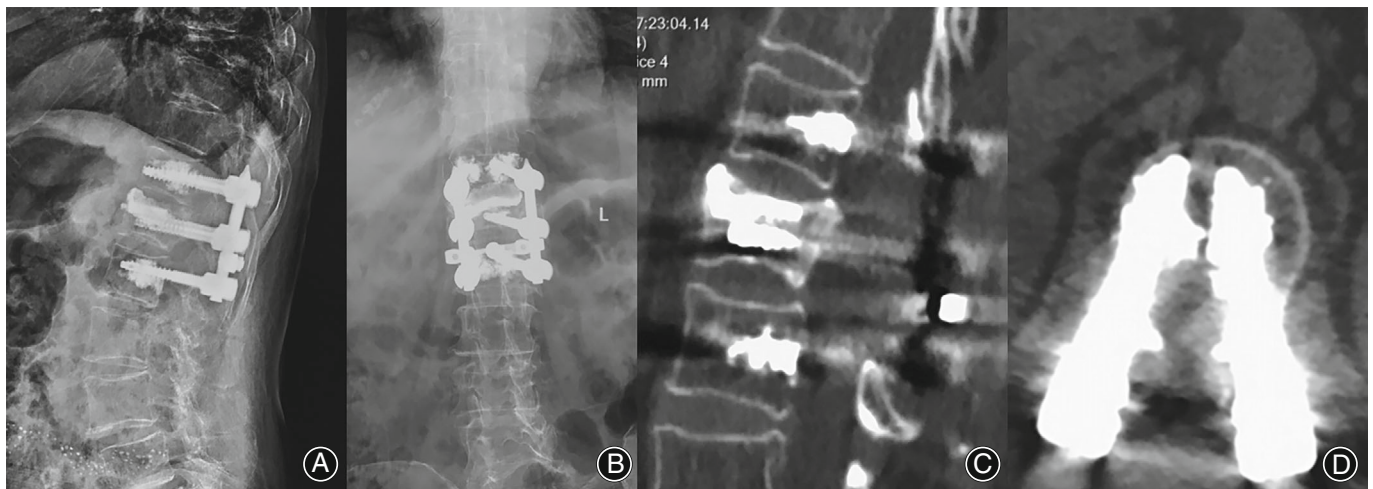


Fig. 2 Radiographs after the first operation. (A, B): lateral and anteroposterior X-ray shows the T12 vertebral body height recovered well; (C, D): sagittal and transverse CT shows the IVC was fully filled with bone cement, spinal cord decompression was complete, and dural sac compression was relieved.

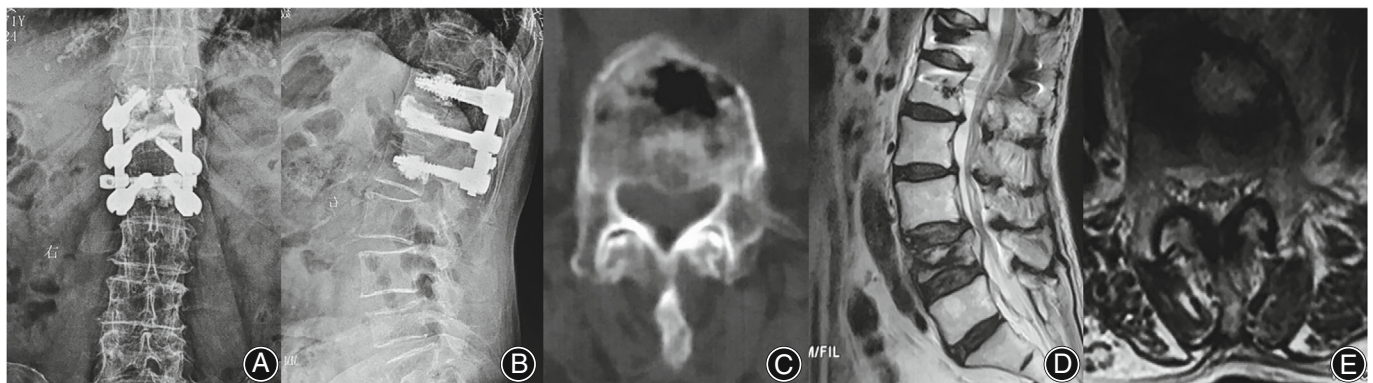


Fig. 3 Radiographs before the second operation. The patient experienced a new Kummell's disease in T4 vertebrae. (A, B): anteroposterior and lateral X-ray shows anterior column collapse mildly; (C): transverse CT shows the IVC located below the upper endplate; (D, E): T2 sequence and transverse MRI shows corresponding segment spinal canal stenosis.

20 days. The patient had a history of Parkinson's disease (PD) for 10 years and outmoded cerebral infarction for 3 years, but the patient did not have diabetes, hypertension, heart disease, or other internal diseases.

On physical examination, the patient was found to have positive pressure and percussion pain in the spinous process area of thoracolumbar section, hypoesthesia in bilateral inguinal area and front thigh, normal superficial sensation in perineum, muscle strength of bilateral iliopsoas III, muscle strength of other muscles IV, positive traction test of bilateral femoral nerve, negative straight leg elevation test, normal tendon reflex, and no pathological reflex. The X-ray films of the thoracolumbar spine showed T12 vertebral collapse, a line of transparent band was seen close to the upper endplate, and the lower lumbar spine was unstable. Computed tomography (CT) showed that the intravertebral vacuum cleft (IVC) was located under the upper endplate of T12

vertebral body, and distributed with the anterior column of the whole vertebral body in cross section. Magnetic resonance imaging (MRI) showed that T12 vertebral collapse with IVC, the posterior tissue of vertebral body protruded into the spinal canal, resulting in corresponding segment spinal canal stenosis and spinal cord compression (Fig. 1). The blood routine test, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), and tumor series test were all normal. The bone density T score measured by double X-ray absorptiometry was $-4.7\text{g}/\text{cm}^3$. The visual analog scale (VAS) score of low back pain was 9 and the visual analog scale (VAS) score of lower limb pain was 7.

After receiving informed consent from the patient, the patient was treated by surgery. The patient was placed in prone position after general anesthesia, and the T12 laminectomy was performed by posterior open surgery to complete decompression. Pedicle screws were implanted on

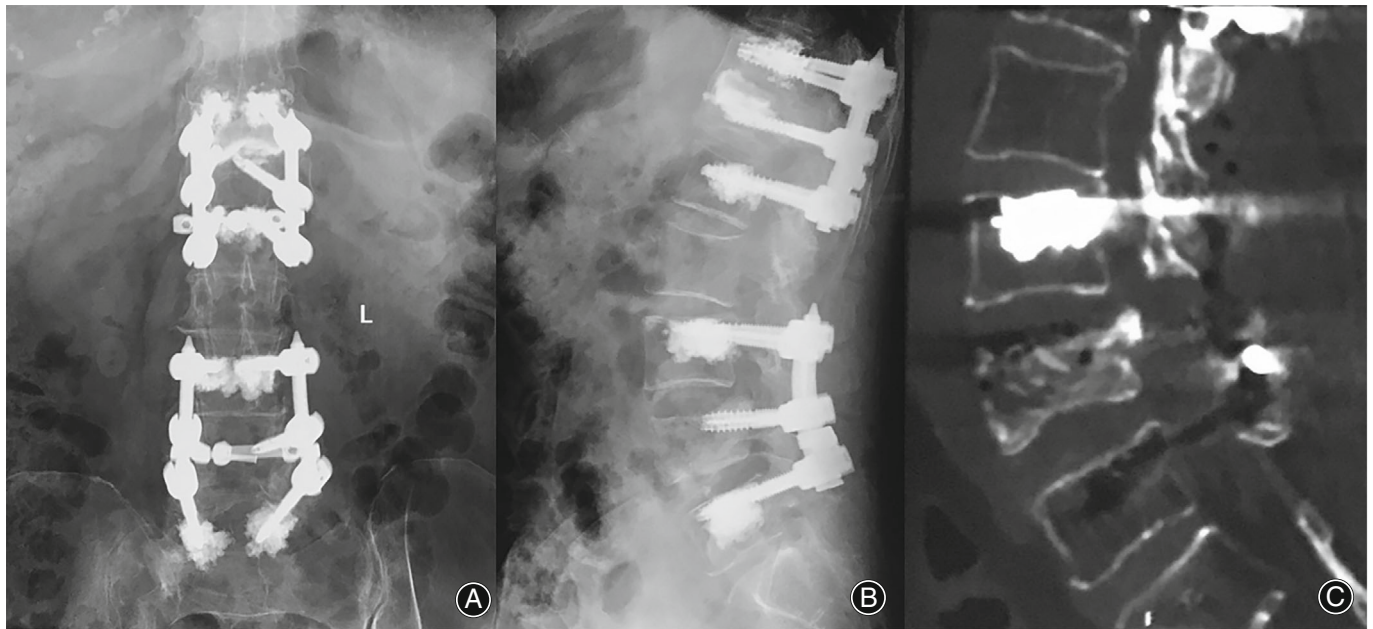


Fig. 4 Radiographs after the second operation. (A, B): anteroposterior and lateral X-ray shows the L4 vertebral body height recovered well; (C): sagittal CT shows the position of internal fixation was in good condition, and the nerve compression was relieved.

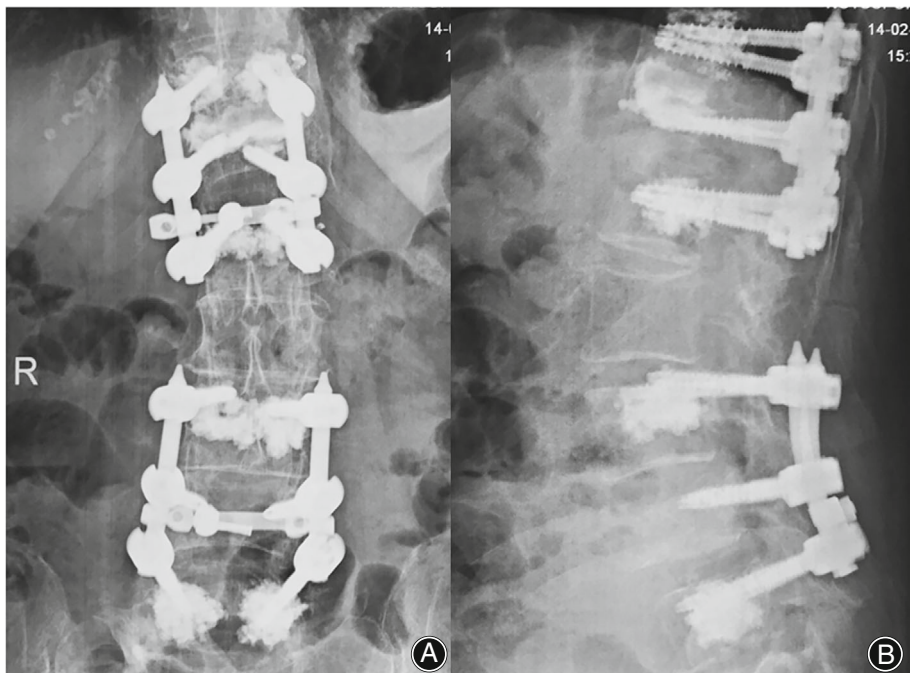


Fig. 5 Radiographs 1 month after the second operation. (A, B): anteroposterior and lateral X-ray shows the right screw of L3 vertebral body loosened and pulled out.

both sides of T11, T12, and L1 and augmented with bone cement. After installing titanium rods to restore T12 vertebral body height, the bone fragments that burst into the spinal canal were incorporated into the vertebral body. IVC was fully filled with bone cement, the transverse connection was installed, and the fusion of posterolateral bone graft was

performed (Fig. 2). One week after the operation, the VAS score of low back pain was 3, and the VAS score of lower limb pain was 1. During 1-year follow-up, the internal fixation position of the patient was good, the VAS score of low back pain was 2, the pain of both lower limbs disappeared, and the muscle strength recovered to normal.

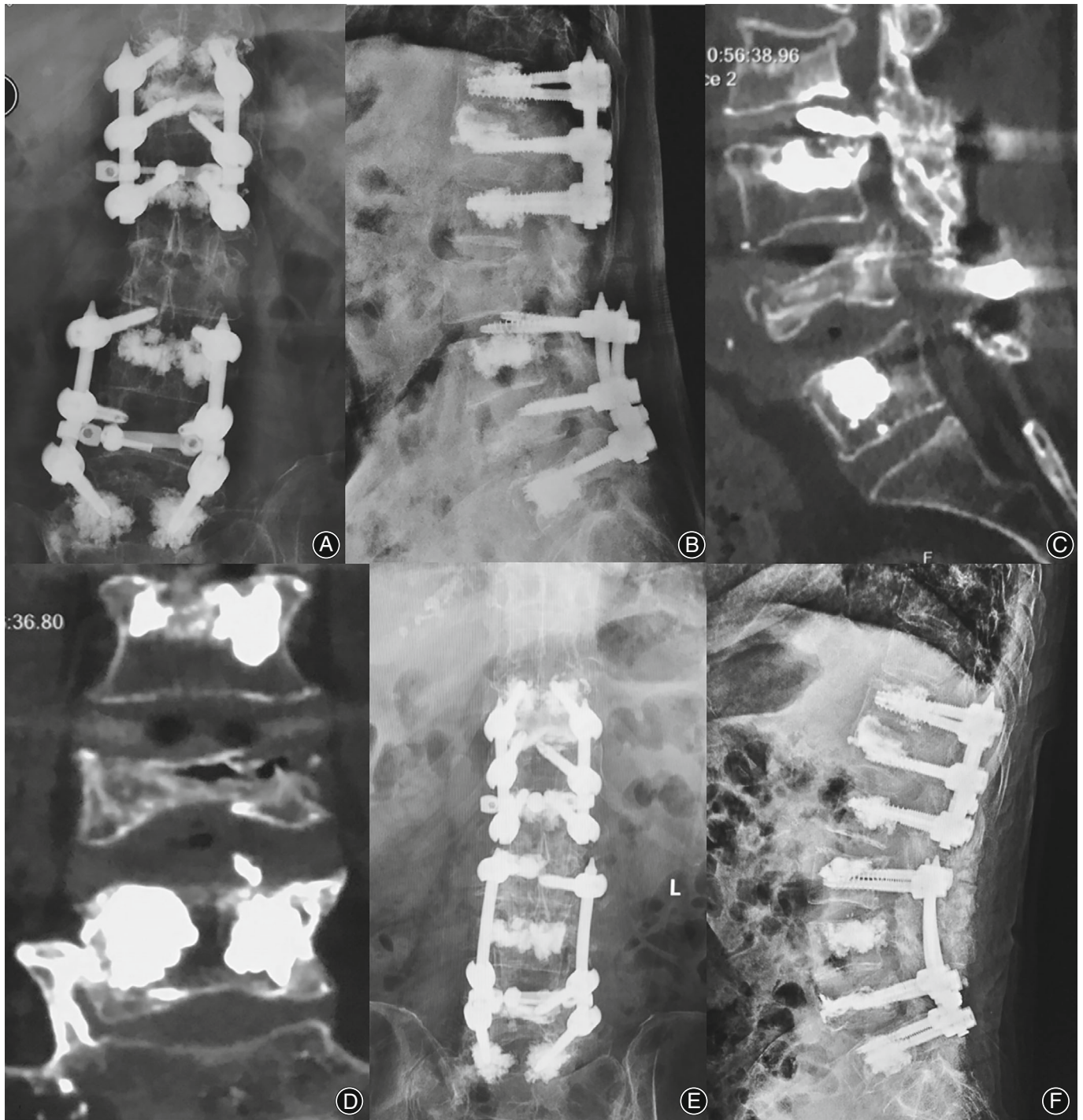


Fig. 6 Radiographs before and after the revision operation. (A, B): anteroposterior and lateral X-ray shows both screws of L3 vertebrae had loosened and pulled out, located in L2/3 disc space, the L4 vertebrae height was significantly lost; (C, D): sagittal and coronal CT shows screws and that the IVC still existed; (E, F): anteroposterior and lateral X-ray after revision surgery shows the L3 screws were removed, and the L4 vertebral IVC was filled with bone cement.

Twenty-two months after the first operation, the patient presented to the clinic in November 2017 with low back pain and left lower extremity pain for 1 month. No

significant pain relief was found after conservative treatment such as oral nonsteroidal analgesics and physical therapy.

TABLE 1 The relationship between osteoporosis and PD

Study name, year	Test subject	Results and Conclusions
Invernizzi <i>et al.</i> 2009 ¹⁹	-	Osteoporosis is a very common finding in patients with PD, affecting up to 91% of women and 61% of men.
Sleeman <i>et al.</i> 2016 ²⁴	326 patients of PD and 261 controls	The fracture rate in PD is over three times greater than controls.
Lee <i>et al.</i> 2018 ²⁵	3370 patients of PD and 16,850 controls	The risk of spinal fractures was significantly increased in PD patients. Osteoporotic vertebral compression fracture was developed in 12.5% of PD patients and in 7.4% of controls.

On physical examination, the pressure percussion pain in the spinous process area of L4 was positive, the superficial sensation in the inner side of the left leg was decreased, the muscle strength of the left quadriceps femoris and tibialis anterior muscle was IV–, the left straight leg elevation test was 50° positive, the right side was negative, the traction test of the bilateral femoral nerve was negative, and the tendon reflex was normal. The X-ray films of lumbar spine showed that the position of thoracolumbar internal fixation was good, the anterior edge of T11/12 vertebrae had formed bridging fusion, and the upper endplate of L4 vertebral body had slightly collapsed. MRI showed that the IVC in L4 vertebrae with edema signal, L4 vertebral body collapsed slightly, and the posterior upper edge of L4 vertebral body protruded into the spinal canal, resulting in spinal canal stenosis and nerve compression. CT cross section showed that there was IVC in the anterior edge of vertebral body (Fig. 3). Similarly, the blood routine test, ESR, CRP, and tumor series test were all normal. The bone density T score was -4.5 g/cm^3 . The patients failed to actively and regularly carry out anti-osteoporosis treatment during the time interval of 22 months. Because the patient needs to take a variety of drugs for internal diseases, she often refuses treatment in order to reduce the types of drugs and avoid adverse reactions. The VAS score of low back pain was 8 and the VAS score of lower limb pain was 7.

Because of the good efficacy of the first operation, we applied the successful experience to the treatment of L4 Kummell's disease, including posterior decompression, short segment fixation, bone cement augmented, and bone graft fusion (Fig. 4). The difference was no L4 vertebroplasty performed due to the fact that the L4 vertebrae collapse was not serious, the IVC range was small, and the pedicle screw fixation strength was high. The pain symptoms were significantly

relieved after operation, the VAS score of low back pain and lower limb pain were improved to 3 and 1, respectively. However, at the reexamination at 1 month after the operation, the right pedicle screw of L3 vertebrae was loose, and the screw was cut to the head side, breaking through the upper endplate and located in the L2/3 disc space (Fig. 5). It is suggested that the patient should undergo revision surgery, but she refused and adopted conservative treatment such as brace protection, bed rest, anti-osteoporosis, and so on. After 3 months of conservative treatment, the patient's back pain was intolerable, with a VAS score of 9, and she agreed to undergo revision surgery. X-ray examination showed that both screws of L3 vertebrae had been pulled out of the vertebrae, located in L2/3 disc space, and L4 vertebrae height was significantly lost. CT also showed that the IVC still existed in L4 vertebrae. In view of the destruction of the L2/3 intervertebral disc, we extended the fixed segment to L2 during the revision operation, and pedicle screws were augmented with polymethylmethacrylate (PMMA). L3 screws were removed, and the L4 vertebral IVC was filled with bone cement and supplemented the posterolateral bone graft fusion of L1–L3 (Fig. 6). After the revision surgery, the patient's pain was relieved, and in the 2-year follow-up period, the back pain did not recur, no new Kummell's disease was found, and the revision operation was considered to be effective.

Discussion

Kummell's disease is defined as delayed post-traumatic vertebral osteonecrosis, which often occurs in an osteoporotic spine.¹⁴ The continuous occurrence of Kummell's disease with neurological impairment is extremely rare. So far, no studies have been reported in the literature.

Although Kummell's disease can also be seen in young patients, the vast majority of Kummell's disease occurs in patients with osteoporosis and is positively related to the severity of osteoporosis.^{4,15} For this patient's, whose condition was combined with severe osteoporosis, the T value of bone density was less than -3.5 g/cm^3 at two times of onset, which was -4.7 g/cm^3 and -4.5 g/cm^3 , respectively. There was no active and regular anti-osteoporosis treatment in the interval, and the bone density was not significantly improved.

An important feature of this patient is that she had PD for 10 years. PD is a degenerative neurological disease associated with loss of self-sufficiency and is characterized by its cardinal features of tremors, rigidity, bradykinesia, and postural instability.¹⁶ Osteoporosis is a "hidden nonmotor face" of PD and a cause of considerable morbidity in the older general population, patients with PD usually have poor bone quality.^{17,18} Invernizzi *et al.*¹⁹ suggested that osteoporosis is very common in patients with PD, affecting up to 91% of women and 61% of men. Compared with healthy people, the normal activities of PD patients were significantly reduced. With the aggravation of PD disease, the activities of patients were further reduced, the muscle strength was significantly reduced, and the bone mass was seriously lost.²⁰ Vitamin D is not only important in human bone metabolism, but also closely related to PD, and PD patients are often accompanied

by Vitamin D deficiency to further aggravate osteoporosis.²¹ Lastly, hyperhomocysteinaemia, an independent risk factor for osteoporosis, is common concentrated in PD.²² PD not only increases the risk of spinal fracture by aggravating osteoporosis, but also is an important independent risk factor for spinal fracture.^{23–25} Sleeman *et al.*²⁴ analyzed 326 patients of PD and 261 controls, they found the fracture rate in PD is over three times greater than controls. For osteoporotic vertebral compression fracture, which is closely related to Kummell's disease, Lee *et al.*²⁵ performed a nationwide population-based study of 3370 patients of PD and 16,850 controls. The results showed that the risk of spinal fractures was significantly increased in PD patients. Osteoporotic vertebral compression fracture was developed in 12.5% of PD patients and in 7.4% of controls. Some studies on the relationship between osteoporosis and PD are shown in Table 1.

In addition, the patient had lower lumbar instability and vertebral rotation during their first incidence of Kummell's disease. There was degree I lumbar spondylolisthesis both L3/4 and L4/5. L4 vertebral body is located in the center of instability. When the above-mentioned risk factors of osteoporosis occur, slight osteoporotic vertebral compression fractures can occur easily and gradually develop to Kummell's disease.

Kummell's disease in patients with PD can lead to neural compromise due to delayed vertebral body collapse, which requires surgical treatment.²⁶ Because both T12 and L4 Kummell's diseases existed with stenosis of spinal canal, compression of dural sac, and symptoms of nerve damage, it was necessary to open decompression and relieve the nerve symptoms. For patients with Kummell's disease combined with neurological deficits, the choice of anterior or posterior approach has been controversial. Liu *et al.*⁸ reviewed 268 cases of Kummell's disease with neurological deficit, of which 37.7% received anterior surgery and the other received posterior surgery. There was no significant difference in clinical efficacy between the two approaches in terms of pain relief, neurological function improvement, and kyphosis correction. However, anterior approach surgery was associated with a higher failure rate of internal fixation, 21.6% vs posterior 14.3%. In addition, considering the patient's age, combined with various internal medical diseases, short segment fixation with cement augmentation can achieve the same clinical results as long segment fixation.¹⁰ We naturally applied the successful treatment experience of T12 Kummell's disease to L4 vertebral lesion with almost identical clinical characteristics, but the patient experienced failure of internal fixation.

Through a joint discussion with experts from multiple medical centers, we all agreed that the possible reasons for this first two operation: (a) no effective bone cement filling for IVC in L4 vertebrae; although the two procedures were basically the same, the only difference was that in the treatment of L4 Kummell's disease, the operator thought that the height loss of L4 vertebral body was slight,

and hoped that IVC can heal itself under the strong fixation with pedicle screws and effective posterolateral bone graft fusion. However, the results of this case showed that it is not feasible for treating Kummell's disease without valid filling of IVC and without effective support of anterior column of diseased vertebral body. The patient's IVC of L4 was not filled with bone cement and there was no effective support in the anterior column of the vertebral body. In the stage of posterolateral bone graft not yet fused, IVC could be further expanded, the upper endplate of L4 vertebral body collapsed, the anterior column height of L4 was further lost, and the L3 vertebrae moved downward to maintain L3/4 intervertebral space and stability, while the L3 pedicle screws fixation could not move downward, so the screw moved upward out of the upper endplate of L3, located in the L2/3 disc space. During the revision operation, the IVC of L4 vertebrae was filled with PMMA. Although the fixed segment leaped to L2, which was theoretically inferior to L3–5 fixation in strength. However, there were no complications related to implants in the 2-year follow-up, which indirectly proved that the filling of IVC and the effective support of anterior column were the most important parts in the treatment of Kummell's disease; (b) the first two operations fused the T11–L1 and L3–L5 segments of the patients, respectively, and this may be one of the important reasons for the L3 screws pullout 1 month postoperatively. So, in terms of load-sharing, considering the long-segment fixation from T11 to L5 in the second operation (the new L2–L5 fixation to be connected to the preexisting T11–L1 fixation) may have better performance; (c) the pedicle screws of L3 vertebrae were shorter; although the adoption of particle screw authentication with PMMA can effectively increase the fixation strength,²⁷ the screw length is positively correlated with the fixed strength. In this case, the L3 screws only reached the middle of the vertebral body, and the fixation strength was much smaller than that of the screws placed at the anterior edge of the vertebral body, which also increased the risk of screws pullout. The right screw was shorter than the left screw, and the right screw was the first to be pulled out, which indirectly confirmed the importance of the screw length in the anti-pullout effect; (d) in the patient with severe osteoporosis, it has been confirmed that bone density is critical to the pullout strength of pedicle screw.²⁸ Due to various factors, severe osteoporosis has not been improved and the screws were easy to pullout; (e) PD is an independent risk factor for failure of internal fixation after spinal fusion; Watanabe *et al.*²⁹ reviewed 26 cases of osteoporotic thoracolumbar vertebral fractures with PD and 296 matched controls from the data of 27 universities. The PD group showed higher rates of complications. According to the study by Kimura *et al.*³⁰ the rate of implant failure was high as 33.3% in the lumbar fusion patients with PD; (e) the placement of transverse connection between L4/5 pedicle screws and not between L3/4 pedicle screws was a risk factor.

In conclusion, the treatment of patients with Kummell's disease combined with PD is extremely difficult. Osteoporosis, PD, and many other factors may lead patients to become prone to consecutive Kummell's disease, and patients are prone to experience failure of internal fixation. Bone cement filling of vertebral IVC and effective support of anterior vertebral column are very important procedures to ensure the clinical efficacy of treating Kummell's disease.

Acknowledgements

We are thankful for the support of the nursing staff from the Department of Spine Surgery, Xi'an Jiaotong University College of Medicine, Honghui Hospital.

Authors' Contributions

BW, DH, and WG conceived the original ideas of this manuscript. BW, HY, and HS participated in the surgical and medical treatment. YZ and WY executed the follow-up examination and materials collection. YZ, GH, and BW analyzed the examination results, analyzed the data, prepared the Figures and the manuscript. All authors have read and approved the manuscript.

Consent for Publication

The authors have obtained the patient's written informed consent of his personal clinical data and all images in this study for print and electronic publication.

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