Analysis of strategy and efficacy clinical treatments of Kümmell disease: a cohort study

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Background: Most clinical reports on the surgical treatment of kümmell disease lack consistency between classification and treatment protocol. In this study, we investigate the most appropriate and effective clinical treatment strategies according to the characteristics of different types of Kümmell disease.

Methods: A retrospective analysis was performed of 48 patients with Kümmell disease treated in Tongde Hospital of Zhejiang Province, Hangzhou, Zhejiang, China for which complete follow-up data were available. All cases were classified into six types: type I, vertebral body height loss less than 20% and no intervertebral disc degeneration from adjacent segments; type II, vertebral body height loss more than 20% and accompanied by degeneration or mild instability of intervertebral discs at adjacent segments; type III, posterior vertebral cortical rupture and dural sac compression, and some accompanied by spinal cord nerve injury. Type III includes type IIIA (recoverable stable type), type IIIB (recoverable unstable type), type IIIC (spinal stenosis type), and type IIID (kyphosis type). Methods of surgery: patients of types I, II, and IIIA were treated with percutaneous vertebroplasty (PVP) or percutaneous kyphoplasty (PKP), type IIIB were treated with posterior fixation and fusion, type IIIC were treated with posterior decompression and fixation fusion, and type IIID were treated with posterior osteotomy, orthopedic fixation, and fusion. All patients were followed up for 10–44 months (mean, 20.5±4.5 months). The preoperative and postoperative visual analog scale (VAS) scores, Owestry disability index (ODI) scores, secondary height loss and kyphosis, and neurological improvement were followed up and statistically analyzed.

Results: The VAS and ODI scores of all cases were improved compared with those pre-surgery (P<0.05). A total of 8 cases showed loss of vertebral height or secondary kyphosis. The American Spinal Injury Association (ASIA) grades of patients with neural impairment were all improved at the last follow-up.

Conclusions: According to the characteristics of different types of Kümmell disease, appropriate clinical treatment strategies can achieve satisfactory curative effects and reduce the occurrence of complications. This study is only a retrospective study, lacks a control group, and the sample size is small. Therefore, it has limitations and does not provide guidance.

Keywords: Kümmell disease; classification; fixation; injured vertebral height; neural function

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Introduction

Throughout the world, 1.4 million patients experience osteoporotic vertebral fractures (OVF) each year. Acute or chronic low back pain, reduced activity, obvious spinal deformity, and other complications greatly reduce the quality of life of these patients, and even cause serious kyphosis deformity or death due to long-term bed rest. In the early stage of OVF, there are no neural symptoms. However, about 30% of patients experience OVF collapse, 13% display fracture nonunion, and 3% show collapse with delayed nerve injury, which is called Kümmell disease. Kümmell disease was first proposed by the German surgeon, Herman Kümmell, in 1891 (1). The disease refers to patients with a history of mild spinal trauma whom after months to years of no obvious symptoms, show progressive, painful, and angular kyphosis. The main clinical manifestations are significantly increased pain, especially when turning over to get up, standing and walking, and vertebral collapse and progressive kyphosis. Li et al. (2) divided the disease into 3 stages according to its clinical characteristics: stage I, vertebral height loss less than 20%, no adjacent intervertebral disc degeneration, low back pain without neural symptoms; stage II, vertebral height loss more than 20%, adjacent segments of intervertebral disc degeneration or fracture vertebral instability, patients mainly present with low back pain, sometimes accompanied by nerve root symptoms; and stage III, posterior vertebral cortical rupture, dural sac compression, patients present with low back pain or spinal cord nerve injury symptoms, vertebral posterior wall rupture, dural sac compression, fracture vertebral instability, and are prone to secondary nerve injury and delayed paralysis. At present, there have been few studies on the classification and treatment of Kümmell disease in China and internationally. We retrospectively collected data from 48 cases of Kümmell disease in the Tongde Hospital of Zhejiang Province, Hangzhou, Zhejiang, China from January 2014 to January 2018. According to their clinical manifestations and different imaging morphological and pathological changes, we identified the types of Kümmell disease and summarized the surgical methods employed for each type. According to the visual analog scale (VAS) score (3), Oswestry disability index (ODI) score (4), American Spinal Injury Association (ASIA) classification and relevant imaging data during the follow-up period, the postoperative efficacy was evaluated. We present the following article in accordance with the STROBE reporting checklist (available at https://atm.amegroups.com/article/view/10.21037/atm-22-3801/rc).

Methods

Patient information

Patients inclusion criteria were as follows: (I) patients were diagnosed as Kümmell disease based on the Imaging classification and clinical; (II) conservative treatment was ineffective; (III) bone density examination criteria: osteoporosis, T value ≤−2.5; (IV) underwent surgical treatment in Tongde Hospital of Zhejiang Province; (V) retrospective study; and the data of patients were complete. The exclusion criteria were as follows: (I) patients with severe systemic underlying diseases who cannot tolerate surgery; (II) the occurrence of spinal metastases: pathological fractures caused by primary and secondary tumors; and (III) incomplete or missing follow-up data of patients.

A retrospective analysis was performed of 48 patients with Kümmell disease who presented to Tongde Hospital of Zhejiang Province, Hangzhou, Zhejiang, China from January 2014 to January 2018, including 20 males and 28 females, aged 58–90 years, with an average age of 61.5±5.6 years. All cases had a history of mild trauma, with an average of 64.3±6.2 days (range, 5 to 105 days) between the time of trauma and hospitalization. All cases showed different degrees of activity-related low back pain, accompanied by lumbar activity limitation, and some cases also exhibited nerve injury symptoms such as numbness and weakness of the lower limbs, or local kyphosis. All cases underwent a L1–L4 vertebral bone density examination, and the T values were all ≤−2.5. According to the diagnostic criteria recommended by the World Health Organization (WHO), cases with a T value ≤−2.5 can be diagnosed with osteoporosis, and those with a vertebral fracture can be diagnosed with severe osteoporosis. Therefore, all patients in this study presented with severe osteoporosis.

Imaging examination

All cases were examined by X-ray, computed tomography (CT), and magnetic resonance imaging (MRI). All patients displayed an intravertebral cleft (IVC). Some patients showed space within the spinal canal, secondary spinal stenosis, and some patients showed local kyphosis.
Classification and surgical methods

Classification

According to the imaging manifestations and Li classification, as well as clinical symptoms, the cases were classified into 3 stages: stage I, 14 cases (29.2%), vertebral height loss less than 20%, without adjacent intervertebral disc degeneration, and low back pain without neural symptoms; stage II, 12 cases (25.0%), vertebral height loss more than 20%, accompanied by adjacent intervertebral disc degeneration or mild instability; stage III, the posterior cortex of the vertebral body had ruptured and the dural sac was compressed, and these patients presented with low back pain or spinal cord and nerve injury symptoms. We further classified stage III Kümmell disease according to the stability of fractured vertebral body evaluated by CT proposed by Liang et al. (5), and the status of neurological symptoms and kyphosis. Type IIIA (recoverable stable type): 6 cases (12.5%), the height of the fractured vertebrae was significantly restored, kyphosis was corrected more than 50%, secondary spinal stenosis was relieved, and the ratio of the anterior and posterior diameter of the fracture block to the anterior and posterior diameter of the vertebral body was more than 1/2. The shape of the fractured vertebrae was relatively complete. Type IIIB (recoverable unstable type): 7 cases (14.6%), CT reconstruction on the hyperextension position showed that the reduction degree of the fractured vertebral body and the effect of secondary spinal canal release were the same as those of type IIIA; however, the fracture line was diversified, and fracture blocks were present behind the vertebral body or the ratio of the anterior and posterior diameter of the posterior bone block to the anterior and posterior diameter of the vertebral body was less than 1/2. Type IIIC: 6 cases (12.5%), with neural symptoms and space occupying the spinal canal. Type IIID: 3 cases (6.3%), with thoracolumbar kyphosis of >30°.

Surgical methods

The patients of types I, II, and IIIA were treated with percutaneous vertebroplasty (PVP) or percutaneous kyphoplasty (PKP); type IIIB were treated with posterior fixation and fusion; type IIIC were treated with posterior decompression and fixation fusion; and type IIID were treated with posterior osteotomy, orthopedic fixation, and fusion (Figures 1-5). The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by Institutional Ethics Committee of Tongde Hospital (No. Tongde Ethics Approval 2022 Research 088-JY) and individual consent for this retrospective analysis was waived.

Figure 1 Type II Kümmell disease, a 76-year-old female patient with low back pain and discomfort following trauma 2 months previously. (A) An image of the lateral recumbent thoracolumbar position: T12 and L1 wedge deformation. (B) CT showing a T12 vertebral fracture with an intact posterior wall. (C) MRI showing high signals in T12 and the L1 vertebral body. (D) Effective filling of T12 and the L1 vertebral body fracture cavity using bone cement. CT, computed tomography; MRI, magnetic resonance imaging.
Figure 2 Type IIA Kümmell disease, a 70-year-old female patient with low back pain and discomfort following trauma 3 months previously. (A) An image of the lateral recumbent thoracolumbar position: T12 and L2 wedge deformation, and signs of a T12 vertebral body crack. (B) CT showing a crack in T12. (C) MRI showing high signals in the T12 vertebral body. (D) Effective bone cement filling of the T12 and L2 vertebral body after PVP. CT, computed tomography; MRI, magnetic resonance imaging; PVP, percutaneous vertebroplasty.

Figure 3 Type IIIB Kümmell disease, a 71-year-old female with recurrent lumbar pain and discomfort following trauma 4 months previously. (A) An image of the lateral recumbent thoracolumbar position showing L1 wedge deformation with kyphosis. (B) CT examination showing a cavity in the upper part of the L1 vertebral body. (C) High signal in the upper part of the L1 vertebral body. (D) An image of the lateral recumbent thoracolumbar position showing a good spinal sequence, good internal fixation, and effective bone cement filling of the L1 vertebral fissure. CT, computed tomography.
**Evaluation index of efficacy**

The VAS score was used for observe the degree of pain relief before and after surgery, the ODI score was used for evaluate the improvement of physical function pre and post operation, and ASIA classification was used for observe the improvement of neurological function after operation. During the follow-up period, changes in the anterior vertebral height and local kyphosis Cobb angle at 7 days, 1, 3, 6, and 12 months postoperatively were observed. The anterior vertebral height was expressed by the sagittal index (SI; SI = anterior vertebral height/posterior vertebral height ×100%), and the incidence of complications such as partial loss of the height of the injured vertebrae, severe pain due to repeated collapse of the injured vertebra, secondary nerve damage were observed.

**Statistical analysis**

The software SPSS 18.0 (IBM Corp., Armonk, NY, USA) was used for statistical analysis. The measurement data were expressed as mean ± standard deviation, and a t-test was used for analysis. A P value <0.05 was considered statistically significant.

**Results**

All cases were followed up for 12–44 months, with an average follow-up period of 28.4 months. The VAS scores of patients in type I to IIID respectively decreased from 8.1±1.6, 8.0±1.5, 8.3±1.4, 8.0±1.2, 8.4±1.2, and 8.2±1.3 to 2.1±1.2, 2.1±1.2, 2.2±1.3, 2.3±1.4, 2.4±1.5, and 2.3±1.2 at the last follow-up. The ODI scores of patients in type I to IIID respectively decreased from 67.5±2.6, 69.5±2.8, 69.7±2.6, 66.7±2.6, 67.6±2.5, and 67.7±2.6 to 38.1±1.5, 37.1±1.3, 34.1±1.3, 35.2±1.4, 33.1±2.1, and 34.3±1.3 at the last follow-up, which were significantly improved compared with preoperative (P<0.05, Tables 1,2). Among 32 patients with stage I, II, and III stable types, 3 cases showed partial loss of the height of the injured vertebrae, severe pain due to followed collapse of the injured vertebra, secondary nerve damage were observed.

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**Figure 4** Type IIIC Kümmell disease, an 81-year-old female with a history of mild trauma 4 months previously. The symptoms of low back pain recurred 1 month ago, accompanied by numbness and weakness of the right lower limb and intermittent claudication. (A) A lumbar lateral X-ray film showing a serious loss of L2 vertebral height. (B) A sagittal CT reconstruction showing the L2 vertebral fracture block protruding backward and secondary spinal canal stenosis. (C) Sagittal MRI T2 images showing L2 level spinal canal stenosis and dual sac compression on the same level. (D) A postoperative X-ray showing that the patient underwent posterior lumbar open reduction and decompression, and bone cement enhanced internal fixation combined with PVP of the injured vertebra, restoring the height of the L12 vertebral body and resulting in no obvious bone block protrusion at the posterior edge of the vertebral body. CT, computed tomography; MRI, magnetic resonance injury; PVP, percutaneous vertebroplasty.
12th month after surgery, respectively, without obvious clinical symptoms. Their MRIs showed no abnormal signals in the injured vertebra, so no treatment was administered. The SI of the third patient decreased from 89.5% on day 7 after surgery to 65% on the 12th month after surgery. They exhibited local kyphosis and a local Cobb angle of 20.6° due to the repeated collapse of the injured vertebrae, resulting in severe pain. Posterior pedicle screw fixation and screw path strengthening orthopedic bone graft fusion internal fixation were performed, and the pain symptoms and kyphosis deformity were significantly improved, without height loss of the injured vertebra after 24 months of follow-up. During the follow-up period, 2 cases of type IIIB showed partial loss of the anterior height of the injured vertebrae, the SIs of whom decreased from 85% and 83% on day 7 after surgery to 78% and 75% on the 12th month after surgery.

Table 1 Comparison of VAS score between pre-operation and final follow-up

| Stage | Pre-operation | Final follow-up |
|-------|---------------|-----------------|
| I (n=14) | 8.1±1.6 | 2.1±1.2* |
| II (n=12) | 8.0±1.5 | 2.1±1.2* |
| IIIA (n=6) | 8.3±1.4 | 2.2±1.3* |
| IIIB (n=7) | 8.0±1.2 | 2.3±1.4* |
| IIIC (n=6) | 8.4±1.2 | 2.4±1.5* |
| IIID (n=3) | 8.2±1.3 | 2.3±1.2* |

*, compared with pre-operation, P<0.05. VAS, visual analog scale.

Table 2 Comparison of ODI score between pre-operation and final follow-up

| Stage | Pre-operation | Final follow-up |
|-------|---------------|-----------------|
| I (n=14) | 67.5±2.6 | 38.1±1.5* |
| II (n=12) | 69.5±2.8 | 37.1±1.3* |
| IIIA (n=6) | 69.7±2.6 | 34.1±1.3* |
| IIIB (n=7) | 66.7±2.6 | 35.2±1.4* |
| IIIC (n=6) | 67.6±2.5 | 33.1±2.1* |
| IIID (n=3) | 67.7±2.6 | 34.3±1.3* |

*, compared with pre-operation, P<0.05. ODI, Oswestry Disability Index.
No abnormal signal was found on MRI, so no treatment was given, and the follow-up was continued. A solitary case of type IIIC showed a re-loss of injured vertebral height during the follow-up, SI decreased from 86.5% on day 7 after surgery to 66.9% on the 12th month after surgery, accompanied by local kyphosis, a local Cobb angle of 25.2°, and mild back pain after exertion, which was relieved after drug treatment and functional exercise without reoperation. At the last follow-up, the ASIA grade of 6 cases with neural symptoms was improved from preoperative grade C to grade D in 1 case, and from preoperative grade D to grade E in 5 cases. During the follow-up period of 3 cases with type IIID, 1 case showed loss of the injured vertebral height to varying degrees. Their SI decreased from 86.6% on day 7 after surgery to 68.5% on the 12th month after surgery. The patient was asymptomatic, so no treatment was administered.

In conclusion, the VAS and ODI scores of all cases were significantly improved post-surgery compared with pre-surgery, and these differences were statistically significant. During the follow-up, 8 cases (16.7%, 8/48) showed loss of the injured vertebrae or local kyphosis, and 1 case underwent a second operation. At the last follow-up, the ASIA classification of patients with neural injury symptoms was improved by at least 1 grade (Table 3).

### Discussion

The pathophysiological mechanism of Kümmell disease is not clear. According to previous studies, Kümmell disease is the cumulative effect of many factors, including osteoporosis, vertebral avascular necrosis, and biomechanical changes after a fracture (6,7). Anatomically, the anterior middle third of the vertebral body is with abundant blood supply. Fractures in this area may damage the intramedullary arterioles and lead to fracture nonunion (8,9). From a biomechanical point of view, the cancellous bone of osteonecrosis following trauma bears more Von Mises equivalent stress than the normal vertebrae, and the difference in cavity volume and position may lead to more serious abnormal stress distribution (10). As a result of the occult onset, clinical features and imaging findings are important for diagnosis of the disease. At the time of treatment, patients often have a minor history of trauma, or even no clear history of trauma. The early manifestation of disease is back pain, usually for several days or weeks prior to presentation. After oral medication or self-relief, the pain symptoms appear again after several months or years, or manifest as gradually worsening kyphosis deformity. Some patients have neural and spinal cord compression, and the phenomenon of vacuum fissure in the vertebral body is considered the imaging evidence of vertebral body avascular osteonecrosis (11-14). In terms of clinical classification, Kümmell disease is mainly divided into 3 types according to the classification of Li et al. (2) and Liang et al. (5): type I, vertebral body height less than 20% and no intervertebral disc degeneration at adjacent segments; type II, vertebral body height loss more than 20% and accompanied by degeneration or mild instability of intervertebral discs at adjacent segments; type III, posterior vertebral cortical rupture and dural sac compression, and some accompanied by spinal cord nerve injury. According to CT images reconstructed from hyperextension, the type III cases were divided into type IIIA (recoverable stable type), type IIIB (recoverable unstable type), type IIIC (spinal stenosis type), and type IIID (kyphosis type). Type IIIA: the correction of fracture vertebral collapse is greater than or equal to 50%, vertebral posterior margin bone block can be reduced, secondary spinal stenosis can be relieved, and posterior vertebral fracture block anterior-posterior diameter is greater than or equal to 1/2 of the anterior-posterior diameter of the vertebral body. Type IIIB: the correction of vertebral collapse is less than 50%, or there is no obvious reduction of vertebral posterior margin bone. In most cases, Kümmell disease does not resolve naturally. Traditional conservative treatments such as bed rest and brace wearing are often ineffective, subsequently leading to chronic back pain or disability. Tripatide, a bone formation promoting

| ASIA grade at preoperative | N=6 | ASIA grade at final follow-up |
|---------------------------|-----|------------------------------|
|                           |     | A   | B   | C   | D   | E   |
| C                         | 1   | 0   | 0   | 0   | 1   | 0   |
| D                         | 5   | 0   | 0   | 0   | 0   | 5   |

ASIA, American Spinal Injury Association.
drug, is considered beneficial to patients undergoing conservative treatment, but it can take a long time to be effective (15). Surgical treatment is more effective at quickly relieving pain, correcting kyphosis deformity, and reducing the complications resulting from long-term bed rest, and is therefore widely employed. Surgical treatment mainly includes PVP, PKP, and open anterior and posterior surgery (16-22). However, it remains unclear as to which type of surgery is most suitable for each type of Kümmell disease (23,24).

Based on the classification of Kümmell disease by Li et al. (2) and Liang et al. (5), we further refined the original classification by adding type IIIC (spinal canal stenosis), spinal canal occupation accompanied by neurological symptoms, in consideration of whether patients had clinical symptoms of nerve injury, spinal canal occupying on imaging, kyphosis, and so on. Type IIID (kyphosis) thoracolumbar kyphosis deformity angle greater than 30°. Among them, type I, type II, type IIIB belong to stable type, and type IIIB belongs to unstable type. We recommend selecting different surgical treatment schemes according to different types to relieve clinical symptoms and neurospinal cord compression.

The main feature apparent when imaging patients with stable (I, II, IIIA) Kümmell disease (i.e., classic Kümmell disease) is “bone nonunion”. An IVC is evident on imaging, containing either gas or liquid. The fracture ends appear hardened, and the formation of pseudojoints is evident. The pain experienced by patients is mainly related to the movement of pseudojoints in the vertebral body (25). The purpose of surgery is to eliminate pseudojoint activity, thereby reducing pain. Bone cement filling of the IVC can stabilize the vertebral body and eliminate the micro-motion of a fracture. Therefore, PVP can immediately relieve the pain. During the follow-up period, it was found that the height of the strengthened vertebral body was reduced to different degrees among the 3 cases who received this treatment. A patient experienced severe vertebral collapse and developed local kyphosis. In response to the recurrence of intractable pain, pedicle screw fixation and screw channel enhanced orthopedic bone grafting and internal fixation were adopted. On reviewing the clinical data of these 3 cases, we detected a serious level of osteoporosis, with an insufficient amount of cement having been injected into the injured vertebrae. Furthermore, it appeared that the regular anti-osteoporosis treatment recommended after surgery had not been carried out in strict accordance with the doctor’s advice, which may be the reason for the loss of vertebral height. The unstable type (IIIB) of Kümmell disease is characterized by the obvious “mouth opening phenomenon” of injured vertebra on a dynamic position film, which is common in the thoracolumbar segment. Pain is related to instability between segments and pseudojoint activity. The purpose of surgery is to fix the unstable segments. Therefore, posterior fixation and fusion can eliminate segmental instability, stabilize the spine, and eliminate pain. In 2 cases with this type of disease, partial loss of the injured vertebral height was detected during follow-up. No abnormal signal was found in the injured vertebra on MRI, and no treatment was administered. Kümmell disease of the spinal canal stenosis type (IIIC) is accompanied by different degrees of nerve injury symptoms, in addition to local pain symptoms. It is mainly caused by the backward displacement of free bone fragments compressing the nerve. The main purpose of surgery is to relieve the nerve compression and the back pain in the patient. Therefore, the surgical method is decompression and fixation fusion (26,27). Lee et al. reported that 10 patients with Kümmell disease and neural symptoms underwent posterior decompression, fixation, and fusion. The neural function of all patients had improved by at least 1 ASIA level at the last follow-up compared with pre-surgery (28). At the last follow-up, the ASIA grade was improved from grade C in 1 case and grade D in 5 cases to grade D in 1 case and grade E in 5 cases. Neural function was improved by at least 1 level, which further supported the effectiveness of this surgical method. Kyphosis Kümmell disease (type IIID) is characterized by a severe wedge-shaped change in 1 vertebral body or a continuous wedge-shaped change in 2 or more vertebrae, malunion of the fracture, and a deformity angle of local kyphosis of ≥30°. Lumbodorsal pain is mainly caused by muscle tension in the lumbar back resulting from kyphosis. The main purpose of surgery is to correct kyphosis and relieve tension in the lumbar and dorsal muscles. Theoretically, surgical intervention may also improve kyphosis and relieve local pain symptoms. The results of this study showed that the VAS score and the local kyphosis angle were significantly improved compared with those pre-surgery, which confirmed the safety and effectiveness of posterior osteotomy.

In conclusion, establishing a corresponding surgical plan according to the characteristics of different types of Kümmell disease, combined with the specific conditions of patients with thoracolumbar instability, local kyphosis and nerve injury can achieve satisfactory clinical treatment effects. However, the onset of Kümmell disease is a complex pathological process, patients’ age, degree of osteoporosis,
mental state, and compliance with anti-osteoporosis treatment during and after treatment should be taken into consideration in the treatment process, so as to conduct comprehensive evaluation and provide personalized treatment.

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**Footnote**

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**Ethical Statement**: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by Institutional Ethics Committee of Tongde Hospital (No. Tongde Ethics Approval 2022 Research 088-JY) and individual consent for this retrospective analysis was waived.

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