

Review Article



# Kummell's Disease is Becoming Increasingly Important in an Aging Society: A Review

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**Conflict of Interest**




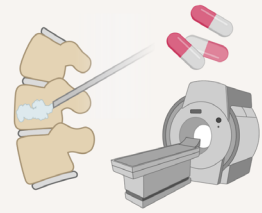
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## ABSTRACT

Kummell's disease (KD) is referred to as delayed posttraumatic vertebral collapse, avascular necrosis of vertebrae, or ischemic vertebral collapse. KD is no longer rare in an aging society. It is mainly caused by minor trauma, and nonunion occurs secondary to avascular necrosis at the vertebral body fracture site, which can lead to vertebral kyphosis or intravertebral instability. Clinical symptoms of KD range from no symptoms to severe paralysis due to nerve injury. KD is considered a complication of osteoporotic vertebral compression fractures, and conservative treatment, including osteoporosis treatment, is important. Timely interventions such as vertebral augmentation or surgery, with active regular follow-up are necessary before the onset of neurological deficits due to osteonecrotic collapse in patients with suspected KD. In this study, we summarize the pathogenesis, diagnosis, and treatment of KD, which is showing increasing prevalence in an aging society. We have presented a literature review and discussed clinical guidelines and therapeutic strategies to reduce the morbidity and mortality associated with KD.

**Keywords:** Osteonecrosis; Osteoporosis; Vertebral body; Conservative treatment; Surgical procedures, operative

**GRAPHICAL ABSTRACT**

<p><b>Kummell's Disease is Becoming Increasingly Important in an Aging Society: A Review</b></p> 	
Subject	Method
 <p><b>Kummell's disease (KD) is called</b></p> <ul style="list-style-type: none"> <li>- Delayed posttraumatic vertebral collapse</li> <li>- Avascular necrosis of vertebrae</li> <li>- Ischemic vertebral collapse</li> </ul> <p>Osteoporosis is the most representative risk factor for KD.</p>	 <p><b>Clinical presentation</b></p> <ul style="list-style-type: none"> <li>- Asymptomatic periods are present initially after minor trauma</li> <li>- Pain gradually become more severe</li> <li>- Vertebral body collapse or neurological damage occurs in the final stage</li> </ul> <p><b>Diagnosis</b></p> <ul style="list-style-type: none"> <li>- X-ray: deterioration of angular kyphosis or compression on serial follow-up</li> <li>- CT: the margin or shape of intravertebral cleft (IVC)</li> <li>- MRI: IVC / prevertebral soft tissue changes</li> </ul>
Result	
 <p><b>Conservative treatment</b></p> <ul style="list-style-type: none"> <li>- Bed rest, brace, medication, osteoporosis management</li> <li>- Osteoanabolic agents at least 3 months</li> </ul> <p><b>Vertebral augmentation</b></p> <ul style="list-style-type: none"> <li>- Amount of cement injection should be adjusted according to IVC</li> <li>- Need for continuous radiologic follow-up</li> </ul> <p><b>Surgery</b></p> <ul style="list-style-type: none"> <li>- Posterior approach is safer in elderly multi-segmental cases</li> <li>- Cement reinforcement is effective for screw loosening</li> </ul>	
<p><b>Conclusion</b></p> <p>In an aging society, KD is no longer a rare disease. In patients with suspected KD, timely interventions such as vertebral augmentation or surgery are necessary before neurological deficits due to osteonecrotic collapse occur through active regular follow-up.</p>	

**INTRODUCTION**

Kummell's disease (KD) was first reported by Hermann Kummell in 1895 and is called delayed posttraumatic vertebral collapse, avascular necrosis of vertebrae, or ischemic vertebral collapse.<sup>30)</sup> It is mainly caused by minor trauma, and nonunion occurs due to avascular necrosis at the vertebral body fracture site, which can lead to vertebral kyphosis or intravertebral instability.<sup>23)</sup> Most patients can be controlled with conservative treatment alone, but vertebral augmentation or surgery is often required.<sup>20)</sup>

Osteoporosis is the most representative of several risk factors for KD, and it has been reported that nonunion occurs in about 7%–37% of patients with osteoporotic vertebral compression fracture (OVCF).<sup>20,34)</sup> The osteoporosis gradually increases with age, and therefore its prevalence is increasing as we enter an aging society.<sup>23)</sup> Active management for osteoporosis is important to reduce the incidence and exacerbation of KD.<sup>36)</sup>

This study aims to summarize the pathogenesis, diagnosis, and treatment of KD, which is increasing in an aging society, through the previous studies, and to examine the role of a clinician to reduce the morbidity or mortality caused by it.

## PATHOGENESIS

Although many theories have been proposed to explain KD, the pathogenesis is not yet clearly understood.<sup>22)</sup> Since intravertebral vacuum cleft (IVC), considered as avascular osteonecrosis, is common in radiologic findings of KD, many authors have cited avascular necrosis of the vertebral body as the primary pathogenesis of KD.<sup>16,25,37)</sup>

It is known that osteoporosis is highly related to the occurrence of KD because IVC usually occurs in middle-aged or older OVCF patients.<sup>36)</sup> The incidence of IVC showed a negative correlation with BMD, and a decreasing of osteoblasts in osteoporotic patients is considered one of the risk factors that increase vertebral ischemia.<sup>16,36)</sup>

Long-term glucocorticoid therapy, diabetes, alcoholism, pancreatitis, radiation therapy, vein hardening, trauma, infection, and tumor are known as risk factors inducing avascular necrosis of the vertebral body, and among them, glucocorticoid has been reported as a crucial induction factor for IVC and nonunion of vertebral bodies.<sup>8,14)</sup>

KD is more common in women than in men.<sup>1)</sup> In addition, changes of bone biomechanics have been reported as a cause of KD because it is frequently observed at the thoracolumbar junction.<sup>28)</sup> It is described that fractured vertebra is in a compression state with vertebral height decreasing and kyphosis.<sup>33)</sup> And the kyphosis shifts the center of gravity of weighted spine forward, changes the biomechanics environment of the fractured vertebra and leads to more severe fracture.<sup>14,33)</sup>

### Clinical presentation

Since KD is caused by minor trauma, in the early asymptomatic period, if the clinician does not suspect the presence of minor trauma, appropriate tests may not be performed.<sup>26)</sup> Therefore, it is important to have a suspicion that asymptomatic trauma may exist at the first examination. Clinical symptoms range from asymptomatic to severe paralysis due to nerve damage.<sup>23)</sup>

Among the studies on clinical stages of KD, the studies by Benedek and Nicholas<sup>3)</sup> and Steel<sup>30)</sup> are representative. Asymptomatic periods are present initially after minor trauma, then the pain gradually become more severe, and finally vertebral body collapse or neurological damage occurs in the final stage.

If OVCF is present at the thoracolumbar junction in an elderly female patient, regardless of the presence or absence of minor trauma, regular follow-up is recommended considering the possibility of KD.

### Radiologic findings

KD was thought to be a rare disease when it was first reported, but as we enter an aging society and imaging techniques such as computed tomography (CT) and magnetic resonance imaging (MRI) develop, reports about KD are increasing rapidly.<sup>14,38)</sup>

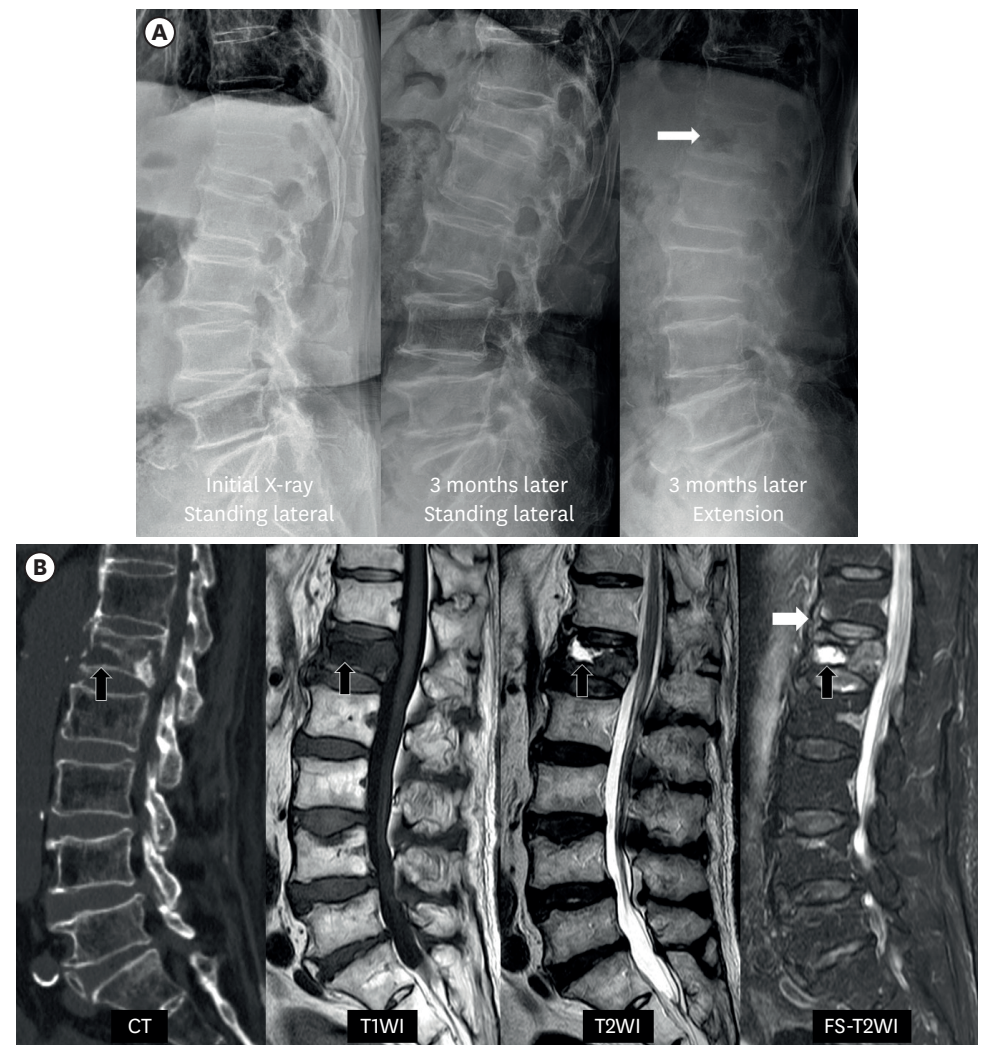
Although the diagnosis of KD is still controversial, IVC reported in 1987 using plain radiography is the most well-known radiological finding.<sup>25)</sup> However, since IVC could also be observed by other causes, such as infection, tumor, or intraosseous disc prolapse, IVC is considered as an eventual finding that strongly suggests the presence of osteonecrosis rather than a pathognomic finding of KD.<sup>14,23)</sup>

Therefore, since the diagnosis of KD through IVC alone has limitations, radiologic features for improved diagnosis of KD have been reported recently, and the related contents are summarized in **TABLE 1** and **FIGURE 1**.

**TABLE 1.** Radiologic findings for diagnosis of KD

Equipment	Method	Finding
X-ray	Serial follow-up, Dynamic imaging	<ul style="list-style-type: none"> <li>· Presence of IVC</li> <li>· Progress of vertebral collapse</li> <li>· Intravertebral instability</li> </ul>
CT	Dynamic imaging	<ul style="list-style-type: none"> <li>· Flatter margin and linear shape of IVC are more suggestive of KD</li> </ul>
MRI	T1WI, T2WI, Fat-suppressed T2WI	<ul style="list-style-type: none"> <li>· Low SI in T1WI/high SI in T2WI in acute phase</li> <li>· T1WI and T2WI varies according to IVC and diagnosis time</li> <li>· Prevertebral soft tissue changes related to IVC</li> </ul>

KD: Kummell's disease, IVC: intravertebral vacuum cleft, T1WI: T1 weight image, T2WI: T2 weight image.



**FIGURE 1.** Radiologic examples about KD.

(A) There is an acute fracture in L1 vertebral body on initial X-ray. In 3 months later, progressive kyphosis of L1 body showed in standing lateral image, and intravertebral cleft (IVC, indicated by white arrow) revealed by extension X-ray. (B) CT shows a clear image about flatter margin of the IVC (indicated by black arrow). T1WI, T2WI, and FS-T2WI show cystic IVC (indicated by black arrow), and there is a prevertebral soft tissue change (indicated by white arrow) related to IVC.

KD: Kummell's disease, IVC: intravertebral vacuum cleft, CT: computed tomography, T1WI: T1 weight image, T2WI: T2 weight image, FS-T2WI: fat-suppressed T2 weight image.

### Plain radiographs

Diagnosis of KD through plain radiographs is possible through serial follow-up imaging. Serial imaging can show the deterioration of angular kyphosis or compression well, but it has the disadvantage that early diagnosis of KD is difficult.<sup>23)</sup>

Plain radiographs show the IVC observed as a transverse, linear, or semilunar radiopaque,<sup>25)</sup> and it is easy to confirm instability related to IVC using flexion/extension dynamic imaging.<sup>6)</sup>

### CT

Since CT provides a clearer image of the bone structure than X-ray, the margin or shape of the IVC can be evaluated.<sup>5,33)</sup> It is reported that the margin of IVC is flatter in KD than in OVCF, and in the case of IVC shape, it is reported that linear type is more common in KD and irregular type is more commonly observed in case of simple OVCF.<sup>5)</sup>

CT is also capable of taking dynamic imaging and has been reported to be very useful for evaluating the instability of KD.<sup>23)</sup>

### MRI

MRI is the most important equipment for diagnosing KD, and the T1 weight image (T1WI) and fat-suppressed T2 weight image (T2WI) are particularly useful.<sup>23,38)</sup> In general, fractured vertebrae in the acute phase show low signal intensity (SI) in T1WI and high SI in T2WI, and change to iso SI as bone marrow edema decreases.<sup>2)</sup> In the case of osteonecrotic vertebral collapse, the SI of T1WI and T2WI varies depending on the contents of the IVC and the diagnosis time, gas present in the IVC is rendered as a loss of signal and can be confused with microhemorrhage or fracture lines.<sup>15)</sup> Therefore, diagnosing KD using only the presence of IVC may result in a false positive or false negative diagnosis.<sup>38)</sup>

Yu et al.<sup>38)</sup> reported IVC accompanied by prevertebral soft tissue changes observed on MRI to improve the diagnosis of KD. Prevertebral soft tissue changes in the fractured vertebral body are related to the mechanism of IVC development and may be a progenitor of IVC.

Even with the current advanced imaging tests, accurate early diagnosis of KD is still difficult in many cases. Therefore, it is considered that more careful attention of clinicians is important to reduce the morbidity and mortality of patients with KD.

### Conservative treatment

The most important treatment for KD without neurological deficit or instability is conservative treatment, including bed rest, wearing a brace, and medication.<sup>23)</sup>

According to the report by Venmans et al.,<sup>34)</sup> 60% of OVCF patients had acute pain relief within 3 months, and 40% of patients had pain that persisted for 1 year. Therefore, additional treatment was recommended if the visual analogue scale (VAS) score of 4 or higher persisted even after 3 months or more.<sup>34)</sup>

It is very important to ensure that patients with OVCF or KD are evaluated for osteoporosis and continue to receive osteoporosis treatment, including calcium, vitamin D, and osteoporosis medications.<sup>23)</sup>

Bisphosphonate is a widely used antiresorptive medication, but there are various reports that it is ineffective for fractures, slows healing, or even accelerates bone union.<sup>10,17,29)</sup> It was also reported to be associated with the occurrence of IVC in OVCF patients.<sup>13)</sup>

Osteoanabolic agents that promote bone formation are effective in reducing pain in OVCF patients.<sup>40)</sup> Compared to bisphosphonate in KD patients, it was not only effective in preventing vertebral collapse, but also showed recovery of intravertebral stability through improvement of IVC.<sup>11,12)</sup>

The prevalence of osteoporosis in Koreans during 12 years of follow-up has been reported to increase from 48.4% to 66.1%, and patients who received antiosteoporosis medication were reported to be not as high as about 41% in men and about 59% in women.<sup>7,19)</sup>

In an aging society, the surgeon's role is not only to perform surgery safely and well, but also to reduce additional outbreaks or worsening of the KD through active osteoporosis management. In this study, it is recommended to use osteoanabolic agents for OVCF or KD patients if there are no side effects or contraindications. And there is no established standard for the period of osteoanabolic agents, but considering the published literature, it is recommended to use it for at least 3 months.<sup>31)</sup> **TABLE 2** briefly summarizes the overall contents of the treatment.

### Vertebral augmentation (vertebroplasty [VP] or kyphoplasty [KP])

Both VP and KP are effective and safe procedures for persistent pain in patients with OVCF or KD.<sup>9)</sup> In many literatures on VP and KP, both procedures have demonstrated a similar improvement in clinical symptoms, and it has been reported that KP is slightly more effective than VP in correcting kyphosis, but the difference is not significantly large.<sup>13,35)</sup>

However, since the polymethyl methacrylate (PMMA) used in both procedures is not a suitable scaffold for bone union to proceed in the fractured vertebral body, it exists as a space-occupying mass without mechanical interlocking with the periphery and may cause additional vertebral body compression or displacement of PMMA inducing neurological deficit symptoms.<sup>20,21,23)</sup>

According to Sun et al.,<sup>32)</sup> they reported that injection of 4–6 mL of bone cement in OVCF patients reduced pain very quickly in patients with mild to moderate fracture. However, as the amount of cement injection increases, the possibility of cement leakage also increases, and the amount of cement must be adjusted according to the presence of IVC, so it is difficult to generalize the amount of cement injection.

**TABLE 2.** Treatment for KD

Category	Indication	Type	Recommendation
Conservative treatment	Asymptomatic–acute pain	<ul style="list-style-type: none"> <li>• Bed rest</li> <li>• Brace</li> <li>• Medication</li> </ul>	<ul style="list-style-type: none"> <li>• Osteoporosis evaluation and treatment consistently</li> <li>• Osteoanabolic agents at least 3 months</li> </ul>
Vertebral augmentation	Persistent pain ( $\geq$ VAS 4) after 3 months	<ul style="list-style-type: none"> <li>• Vertebroplasty</li> <li>• Kyphoplasty</li> </ul>	<ul style="list-style-type: none"> <li>• Amount of cement injection should be adjusted according to IVC</li> <li>• Need for continuous radiologic follow-up</li> </ul>
Surgery	Neurologic deficit, Uncontrolled pain, Kyphosis progression, Intravertebral instability, Vertebral collapse, Cement displacement	<ul style="list-style-type: none"> <li>• Anterior approach</li> <li>• Posterior approach</li> </ul>	<ul style="list-style-type: none"> <li>• Posterior approach is safer in elderly multi-segmental cases</li> <li>• Cement reinforcement is effective for screw loosening</li> </ul>

KD: Kummell's disease, VAS: visual analogue scale, IVC: intravertebral vacuum cleft.

Due to the increase in elderly patients with many underlying diseases, VP and KP, which are relatively less invasive and safe, are being performed more often, but care should be taken for cement leakage, dislocation of bone fragments, and posterior wall displacement that may occur during the procedure.<sup>27)</sup> In addition, attention should be paid to adjacent vertebral fracture or kyphosis progression that may additionally occur through continuous follow-up after the procedure.<sup>4)</sup>

### Surgery

Surgical treatment is required in cases with uncontrolled pain, neurological deficits, progress in kyphosis, intravertebral instability, and progressive vertebral body collapse or cement displacement after VP or KP.<sup>4,21)</sup>

Surgical methods can be largely divided into anterior approach and posterior approach. Both anterior and posterior approaches show improvement of pain, neurological dysfunction, and radiologic outcome, but posterior approach is more advantageous in patients with serious comorbidities or who require multilevel corpectomy.<sup>23,24)</sup> The incidence of instrument failure such as screw loosening and screw fracture was lower in the posterior approach (14.3%) than in the anterior approach (21.6%).<sup>24)</sup>

In the posterior approach, long-segment fixation is more advantageous than short-segment fixation in terms of kyphosis correction and stability, but short-segment fixation may be a more appropriate treatment when considering the patient's age or comorbidity.<sup>39)</sup> In patients with severe osteoporosis, screw loosening can be effectively prevented by using bone cement reinforcement.<sup>18)</sup>

## CONCLUSION

In an aging society, KD is no longer a rare disease. KD is considered as a complication of OVCF, and conservative treatment, including osteoporosis treatment, is very important. In patients with suspected KD, timely interventions such as vertebral augmentation or surgery are necessary before neurological deficits due to osteonecrotic collapse occur through active regular follow-up. Although there are many elderly patients with a high possibility of multiple complications, if clinicians continue their research, efforts, and sincerity to find safer and more effective surgical methods, the treatment results for KD are expected to improve even more.

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