Value of Forceps Biopsy and Kyphoplasty in Kümmell’s Disease

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abstract

The diagnosis of Kümmell’s disease mainly depends on clinical and radiologic findings. However, these are not the gold standards for diagnosing this disease; bone biopsy is required.

The authors performed modified bone biopsy and cement-filling techniques during kyphoplasty and investigated the feasibility and efficacy of kyphoplasty for the treatment of Kümmell’s disease. This study included 28 patients (9 men and 19 women; average age, 71.9 years) with Kümmell’s disease. All patients underwent the modified biopsy procedure and kyphoplasty with the modified cement-filling technique. Treatment efficacy was evaluated using visual analog scale pain scores, Oswestry Disability Index scores, vertebral height, and Cobb angles pre- and postoperatively and at final follow-up. All patients tolerated the procedure well and had immediate back pain relief after kyphoplasty. Biopsy examination revealed necrotic bone in 24 patients and sparse cancellous bone in 2; it was unsuccessful in 2 patients. No severe complication occurred in any patient. Two patients had cement leakage but no clinical symptoms. All efficacy measures were significantly better at the postoperative assessments than the preoperative assessments ($P$<.05) but were similar at the postoperative and final follow-up assessments.

Kyphoplasty is a relatively effective and safe method for treating Kümmell’s disease when modified techniques are performed to prevent cement leakage, and forceps biopsy can be used in the differential diagnosis of this condition.

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Figure: Photograph showing intraoperative biopsy specimens of bony tissue (A). Pathological examination of the biopsy specimens showing necrotic bone and reactive fibrosis (hematoxylin-eosin stain, original magnification $\times 100$) (B).
In the early 1890s, German surgeon Hermann Kümmell reported a clinical scenario in which individuals sustained a trivial spinal trauma, were essentially asymptomatic for weeks to months, and then developed symptomatic, progressive, angular kyphosis.\(^1\) Shortly thereafter, the term Kümmell’s disease was coined to refer to avascular necrosis of the vertebral body. In the middle of the 20th century, many physicians thought that Kümmell’s disease represented post-traumatic vertebral fractures that were initially asymptomatic but then progressed.\(^2\) Although some thought that this diagnosis should be restricted to individuals whose radiographs were unremarkable around the time of the trauma but later developed vertebral body collapse, it was recognized that many individuals would not undergo initial imaging because they had only sustained a mild trauma and were essentially asymptomatic.\(^1\)\(^,\)\(^3\) More recently, with the development of modern medical imaging and spinal surgery, multiple synonymous terms have been used to describe this pathology sequentially, including posttraumatic vertebral osteonecrosis; vertebral pseudarthrosis; intervertebral vacuum, cleft, or gas; delayed vertebral collapse; and nonunion of a vertebral compression fracture.\(^4\)\(^-\)\(^9\)

Kümmell’s disease has rarely been reported in the literature and can easily be misdiagnosed as a pathological fracture. Currently, the diagnosis of Kümmell’s disease mainly depends on clinical symptoms and imaging characteristics. Intraoperative biopsy can provide a direct pathological basis and is important in the differential diagnosis. However, little literature is available on the use of a minimally invasive biopsy technique in Kümmell’s disease.\(^10\) Needle biopsy, which can only be used in patients with fresh vertebral compression fractures suspected to be pathological fractures, has been recommended, but the amount of tissue obtained using biopsy needles is inadequate.\(^11\)\(^-\)\(^14\) To improve the cumulative accuracy and adequacy of the biopsy, the current authors used biopsy forceps instead of needles to obtain tissue samples.

Vertebral augmentation, by means of vertebroplasty or kyphoplasty, is a safe and effective method for treating Kümmell’s disease.\(^4\)\(^,\)\(^15\)\(^-\)\(^16\) Kyphoplasty is more effective than vertebroplasty in reducing kyphosis and restoring vertebral body height. The risk of cement extravasation is theoretically reduced because the cement can be injected under a lower pressure during kyphoplasty. However, the peripheral walls of vertebral bodies with Kümmell’s disease are not always intact, which could result in a higher incidence of bone cement leakage during vertebral augmentation. To minimize the risk of cement leakage, the authors used kyphoplasty with a modified cement-filling technique to treat patients with Kümmell’s disease.

Although some authors have reported that Kümmell’s disease with balloon kyphoplasty,\(^15\) no information is available on the use of the intraoperative forceps biopsy technique and kyphoplasty with a modified cement-filling technique in patients with Kümmell’s disease. Therefore, the authors performed a modified biopsy technique for the differential diagnosis and kyphoplasty for the treatment of Kümmell’s disease.

**Materials and Methods**

A protocol detailing the procedure was approved by the authors’ ethics committee, and written informed consent was obtained from all patients. Between January 2005 and January 2011, nine hundred seventy-five patients diagnosed with osteoporotic vertebral compression fractures underwent balloon kyphoplasty at the authors’ institution. Of these patients, 28 (9 men and 19 women) with Kümmell’s disease were selected for this study. Average patient age was 71.9 years (range, 63-91 years), and average time between pain onset and surgery was 13.5 months (range, 8-39 months). On examination, no patient had neurological deficits. The distribution of painful vertebrae was as follows: T11 in 2 patients, T12 in 10, L1 in 12, and L2 in 4. Preoperative bone density tests showed varying degrees of osteoporosis, with the average bone density being 0.642 g/cm².

The criteria for diagnosing Kümmell’s disease were as follows: (1) at least an 8-month history of pain at the fracture site; (2) low back pain that disappeared or diminished on bed rest and was aggravated by turning in bed, sitting, or walking and was unresponsive to conservative treatment; and (3) intravertebral cleft visible in a vertebral body fracture showing nonunion with hardened cleft edges and widening of the fracture line on routine radiographs or computed tomography scans (Figure 1). On magnetic resonance imaging (MRI), intravertebral cleft as a result of nonunion has a hypointense signal on T1-weighted MRIs and a hyper- or hypointense teardrop-shaped signal on T2-weighted MRIs (depending on whether the cleft contained liquid or gas) and a well-demarcated, hyperintense signal on short inversion time inversion recovery MRIs; occasionally a linear area of hyperintensity can be seen in the hypointensity on T2-weighted MRIs. This hyperintensity with a surrounding band of hypointensity on T2-weighted MRIs is called a double-line sign and corresponds with the intravertebral cleft (Figure 2).\(^17\)\(^,\)\(^5\) Histologic examination of the vertebral body reveals necrotic bone and reactive fibrosis due to ischemia of the vertebral body (Figure 3).

**Surgical Technique**

Kyphoplasty was performed with the patients under local anesthesia. Detailed technical descriptions of the procedure have been published previously.\(^14\)\(^,\)\(^18\) Briefly, bilateral transpedicular access to the fractured vertebral body was obtained by inserting guidewires. The openings were progressively enlarged using successively larger cannulas. A 4-mm biopsy forceps (Karl Storz Inc, Knittlingen,
Germany) was hand twisted through the previously inserted 4.3-mm working cannula into the bone. A bone specimen was retrieved from the intravertebral cleft in the painful vertebra using biopsy forceps. Kyphon balloon tamps (Kyphon, Inc, Sunnyvale, California) were then inserted into the compromised vertebral body. The balloons were inflated to elevate the endplates, reduce the fracture, and create a cavity. Then, they were deflated and withdrawn, and bone cement was placed in the cavity of the vertebral body. In all patients, MENDEC bone cement (Tecres S.P.A., Sommacampagna, Verona, Italy) was used, which comprises polymethylmethacrylate bone cement and 30.0% w/w barium sulfate for optimal opacification. The bone cement setting time allows 1 minute for mixing, 1 for filling the cement inserter, 5 for holding, 10 for working, and 6 for hardening (at a temperature of 23°C in the operating room). For patients with damage to the anterior or lateral wall, or both, of the vertebral body, a small amount of middle-stage bone cement in the dough phase was used to fill the defect, the bone cement inserter was withdrawn by approximately 2 to 3 mm, and 40 to 50 seconds passed before injecting more late-stage bone cement in the paste phase. For patients with damage to the posterior wall of the vertebral body, continuous fluoroscopic monitoring was used throughout the bone cement-filling process. The filling process was stopped if the bone cement distended the entire intravertebral cleft or reached the posterior fourth of the vertebral body.

The volume of cement injected was equal to or less than the inflation volume of the balloon tamps. The cement-filling pipes were then withdrawn slightly but kept in the pedicles until the cement hardened. The pipes were then twisted to break off any cement connected to the pedicles and to prevent the cement from being left in the pedicles when removing these devices from the cannulas. Routine removal of the working canal was performed after

Figure 1: A 69-year-old woman had Kümmell’s disease of the L1 vertebra. Preoperative coronal computed tomographic reconstruction showing intravertebral cleft of the L1 vertebral body with superior and lateral wall defects (A). Preoperative sagittal computed tomographic reconstruction showing anterior wall defects (B).

Figure 2: Preoperative T1-weighted magnetic resonance image showing a hypointense signal of the L1 vertebral body (A). Preoperative T2-weighted magnetic resonance image of the same area demonstrating teardrop-shaped hyperintensity area (arrow) and adjacent hypointensity (arrowhead); this is termed “double line sign” and corresponds with an intravertebral cleft (B). Preoperative short inversion time inversion recovery sequence showing a well-defined, teardrop-shaped marked hyperintensity area in the L1 vertebral body (C).

Figure 3: Photograph showing intraoperative biopsy specimens of bony tissue (A). Pathological examination of the biopsy specimens showing necrotic bone and reactive fibrosis (hematoxylin-eosin stain, original magnification ×100) (B).
inserting the bone cement. This was followed by wound closure.

Postoperatively, the biopsy samples underwent pathological examination. A complete review of these biopsies was performed by 2 experienced pathologists from different institutions. Patients were allowed to walk 24 hours postoperatively and could perform advised out-of-bed activity and gradual resumption of activities of daily living 48 hours postoperatively (Figure 4).

The anterior and middle vertebral heights were measured pre- and postoperatively and at final follow-up to determine the percentage of vertebral height restoration. The local kyphotic angles (Cobb angles) pre- and postoperatively and at final follow-up were measured using the endplates of the fractured vertebrae. When the fractured vertebrae were deformed, the adjacent-level endplates were used to obtain the measurements. Other assessments included pain measurements on a visual analog scale and the Oswestry Disability Index scores were significantly better compared with the preoperative values (\(P < .05\); Table). No significant differences existed between the immediate postoperative and final follow-up assessments in any evaluated efficacy measure (Table).

### RESULTS

All patients tolerated kyphoplasty well. The vertebral body was bilaterally punctured in all patients. Average operative time was 35.4 minutes (range, 25.55 minutes), and average volume of injected bone cement was 4.8 mL (range, 3.0–6.5 mL). Cement leakage occurred in 2 patients: 1 intradiskal leak and 1 paravertebral region leak through the lateral wall. Neither patient had any clinical symptoms. No patient sustained a subsequent fracture after balloon kyphoplasty.

Intraoperative biopsy was attempted in all patients, but samples could not be obtained in 2 patients. Of the 26 patients in whom biopsy was successful, 24 had necrotic bone (light white bone samples) and 2 had sparse cancellous bone (Figure 3).

Patients were followed for a mean of 27.6 months (range, 9–38 months). Mean postoperative anterior and middle vertebral body heights, Cobb angles, and visual analog scale and Oswestry Disability Index scores were significantly better compared with the preoperative values (\(P < .05\); Table). No significant differences existed between the immediate postoperative and final follow-up assessments in any evaluated efficacy measure (Table).

### DISCUSSION

Kümmell’s disease is a delayed disorder characterized by vertebral pseudarthrosis, vertebral compression fracture nonunion, posttraumatic vertebral osteonecrosis, and vertebral body collapse. Gas or liquid accumulates in the vertebral pseudarthrosis in the vertebral body and characteristically appears as an intravertebral cleft sign or intravertebral vacuum phenomenon on computed tomography scans or plain radiographs. This finding can indicate ischemic vertebral collapse but can also be indistinguishable from Kümmell’s disease. Moreover, an intravertebral vacuum phenomenon may solely reflect gas migrating from an adjacent intervertebral disk into the dis-

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### Table

<table>
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<tr>
<th>Variable</th>
<th>Preop</th>
<th>Postop*</th>
<th>Final Follow-upb</th>
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<td>Vertebral body height, %c</td>
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Abbreviations: deg, degrees; ODI, Oswestry Disability Index; Postop, postoperative; Preop, preoperative; VAS, visual analog scale.

*Compared with preoperatively (\(P < .05\)).

*No significant difference existed compared with the postoperative assessments (\(P > .05\)).

Vertebral body height (%)=(height of the fractured vertebral body/(upper vertebral body height–lower vertebral body height)/2).
eased vertebrae through the collapse of the vertebral endplate, not vertebral osteonecrosis.\textsuperscript{22} The intravertebral cleft sign or intravertebral vacuum phenomenon has been referred to as the Kümmell’s sign, and some patients with this sign have been reported to have Kümmell’s disease, although another cause was evident.\textsuperscript{5,21,23} The diagnosis of Kümmell’s disease mainly depends on the clinical symptoms and imaging characteristics. However, imaging characteristics are not the gold standard for the diagnosis of this disease; pathological examination is required.

Intraoperative percutaneous bone biopsy can provide a direct pathological basis and is valuable for differential diagnosis. However, percutaneous spinal biopsy is not always 100\% reliable, and accuracy rates vary widely (range, 16\%-92\%), probably due to the cumulative accuracy and adequacy of samples.\textsuperscript{24} Although needle biopsy has been recommended in the literature, the amount of tissue obtained using biopsy needles is inadequate.\textsuperscript{11-14} Gaitanis et al\textsuperscript{13} performed biopsies in 15 patients but only obtained an adequate tissue specimen in 10 (67\%) patients during kyphoplasty. Muijs et al\textsuperscript{11} performed vertebroplasty to treat 78 patients with fresh osteoporotic vertebral compression fractures. They successfully performed conventional intraoperative biopsy in 71 (91\%) patients, of whom 3 (3.8\%) had malignant tumors that were undiagnosed preoperatively. They recommended using 14-gauge (greater than 2.1 mm in diameter) biopsy needles to improve the biopsy success rate.\textsuperscript{11} However, a meta-analysis of several percutaneous spine biopsy studies showed that overall adequacy and accuracy of percutaneous spine biopsy was not significantly different when using routine or larger inner-diameter needles.\textsuperscript{24}

To improve the cumulative accuracy and adequacy of the biopsy, the current authors used minimally invasive biopsy forceps in all patients instead of biopsy needles to obtain tissue samples and reduce the failure rate. The authors retrieved osteoid tissue from the vertebral fractures in 26 of 28 patients. Light white bone-like tissue that was pathologically identified as necrotic bone was found in 24 of the 26 samples obtained. Similar results were reported by Hasegawa et al.\textsuperscript{25} In the 2 patients in whom biopsy failed, only blood clots were retrieved from the working channels during intraoperative biopsy. In such cases, surgeons should ensure that there is resistance to the biopsy forceps in the range of the vertebrae, and the entire procedure should be performed under continuous fluoroscopic guidance.

To the authors’ knowledge, the biopsy success rate in patients with Kümmell’s disease has not yet been reported in the literature. In the current study, the success rate was 92.9\%, which was higher than the biopsy rate reported by Muijs et al\textsuperscript{11} in patients with fresh fractures. Theoretically, biopsy may be more difficult to perform in patients with Kümmell’s disease than in patients with fresh vertebral compression fractures because of the small intravertebral cleft that is difficult to target in those special cases. Therefore, the authors recommend using biopsy forceps instead of biopsy needles. However, to obtain a better comparison of biopsy forceps and needles, a prospective study comparing both procedures for the treatment of Kümmell’s disease should be performed.

Kümmell’s disease is characterized by pseudarthrosis, which is associated with painful movements during changes in posture or respiration.\textsuperscript{21} Conservative treatment is often unsuccessful in this condition,\textsuperscript{25} and surgical intervention is imperative. Percutaneous vertebroplasty and kyphoplasty are being increasingly used to treat such diseases.\textsuperscript{4,15,16} Yoon et al\textsuperscript{26} reported 2 patients for whom a failed vertebroplasty (ie, back pain was not relieved due to not enough cement in the intravertebral cleft) was salvaged with kyphoplasty. The first choice of treatment for the 2 failed patients not relieved due to not enough cement in the intravertebral cleft was vertebroplasty at another institution. Cement leakage occurred intraoperatively, which compelled the surgeon to stop the injection and resulted in partial or absent filling of the intravertebral cleft. Thus, the vertebral body was not effectively stabilized, and the back pain was not relieved. Kyphoplasty was performed by Yoon et al\textsuperscript{26} for salvage of the failed vertebroplasty, which allowed for the injection of sufficient bone cement to stabilize the vertebral body, and the pain was relieved postoperatively. Garfin et al\textsuperscript{27} also reported that vertebroplasty probably led to inadequate initial fixation of a mobile nonunion that resulted in clinical failure. In the current study, the use of sufficient bone cement can fill the intravertebral cleft, anchor nonunion fractures, and restore stability to the spine. All of the current patients had a significant reduction in pain and improvement in kyphotic angle postoperatively.

Because the intravertebral cleft and peripheral cortical defect in Kümmell’s disease may increase the risk of bone cement extravasation during kyphoplasty, careful and accurate preparation of the working channels and modified bone cement-filling techniques are required to reduce complications. Fresh vertebral compression fractures are different from Kümmell’s disease. In the latter, most of the endplate has usually collapsed or ruptured. Therefore, bilateral transcuderal access to the fractured vertebral body was obtained by inserting guidewires at the 3-o’clock position of the right pedicle and the 9-o’clock position of the left pedicle. The needle is then advanced into the vertebra in a plane parallel to the vertebral endplate. This needle puncture method does not usually injure the endplate. Therefore, the chances of cement leakage into the vertebral cleft through a disrupted endplate are minimized. The peripheral walls of vertebral bodies with Kümmell’s disease are not always intact. Therefore, the current authors cautiously injected bone cement using a modified cement-filling technique that
they developed. For patients with anterior wall defects, Yang et al.\(^8\) suggested initially using small amounts of middle- or late-stage bone cement to cover anterior wall defects, thus helping to prevent cement extravasation. After the cement had hardened, early-stage bone cement was applied to allow the filling to diffuse evenly.\(^9\) However, the surgical technique was not described concretely. In the current study, the authors quantized the modified technique, and it can be easily mastered. Cement leakage occurred in 2 patients: 1 intradiskal and 1 paravertebral region leak through the lateral wall. Neither patient had any clinical symptoms.

**Conclusion**

Performing intraoperative forceps biopsy instead of needle biopsy can improve the cumulative accuracy and adequacy of the biopsy and can be used for the differential diagnosis in patients with Kummell’s disease. Kyphoplasty is a relatively safe and effective method for treating this condition when using modified techniques to prevent cement leakage.

**References**