OLGU SUNUMU / CASE REPORT

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Osteonecrosis of A Vertebral Body: Kummell's Disease Case Report and Review of The Literature

Vertebra Korpusunun Osteonekrozu: Kummel Hastalığı Vaka Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT

Post-traumatic osteonecrosis of the vertebra was first described in 1895 by the German surgeon Hermann Kummell. This disease characterized by the development of kyphotic deformity in the spine was identified prior to the emergence of radiography. The most widely accepted hypothetical pathophysiology is avascular osteonecrosis. Affected vertebrae is usually seen in the thoracolumbar region extending from T8 to L2. This disease is seen rare in the lumbosacral region. The primary aims of surgical treatment of Kummell's disease are neural decompression, stabilization and normal sagital balance of the spinal column. It can be treated by anterior reconstriction, posterior stabilisation, kifoplasty ve vertebroplasty. In this report, We review the clinical features and the diagnostic tools of Kummell's disease and also treatment options accompanied by a case report with kummel disease in L5 spine.

Key Words: Osteonecrosis, Kummell's disease, lumbosacral region

ÖZET

Vertebranın post travmatik osteonekrozu ilk olarak 1895 yılında alman cerrah Herman Kummell tarafından tanımlanmıştır. Omurganın kifotik deformitesi ile karakterize bu hastalık radyografi ortaya çıkmadan önce tanımlanmıştır. Avasküler nekroz en yaygın kabul edilen patofizyolojik hipotezdir. Vertebraya etkisi genellikle T8'den L2 ye kadar torakolomber bölgede gözlenir. Bu hastalık nadiren lumbosakral bölgede yer alır. Kummell hastalığının cerrahi tedavisindeki ana amaç nöral dekompresyon omurganın normal sagital dengesi ve stabilizasyonudur. Anterior rekonstriksiyon, posterior stabilizasyon, kifoplasti ve vertebroplasti ile tedavi edilebilir. Bu makalede Kummell hastalığının tanı araçlarını klinik özelliklerini ve aynı zamanda tedavi seçeneklerini L5 vertebrada kummel hastalığı olan bir vaka eşliğinde gözden geçireceğiz.

Anahtar Kelimeler: Osteonekroz, Kummell hastalığı, lumbosakral bölge

Introduction

Post-traumatic osteonecrosis of the vertebral body was first described in 1895 by the German surgeon Hermann Kummell (1). Kummell's disease, which was identified before radiographic examination was developed, is observed mainly women in middle and advanced age. The frequency of developing Kummell's disease is increasing as the population ages. The pathophysiology of Kummell's disease is not clear (1,2). It is considered that osteoprotic patients are secondary to ischemic necrosis in the vertebral corpus and vascular injury prevents bone healing, resulting in necrosis of the bone. The treatment strategy has not yet been fully determined. The patient's clinical and radiological findings are evaluated and decision is made accordingly. This rare disease, which is frequently observed in the thoracolumbar sector, is rarely seen in the lumbosacral junction (3). In this study, we will discuss our patient with L5 vertebra fracture due to kummel disease to whom we applied anterior transperitoneal L5 corpectomy and posterior vertebroplasty together with with short segment fixation, accompanied by the literature.

Case Report

A 78-year-old female patient was admitted to our clinic with a complaint of low back pain which had started about 1 year previously, recovered partially after 1 month and started again 7 months prior to her visit, increasing over time. It was

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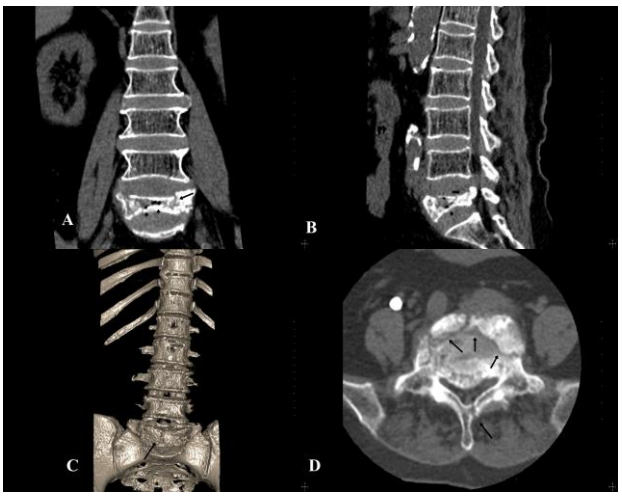

Fig. 1. A. Fracture line in the coronal plane of the reconstruction of CT sections (arrow) and intra-vertebral air density (arrowhead). B. Intra-vertebral air density in the sagittal plane of the reconstruction of CT sections (arrowhead) and vacuum phenomenon in the L4-5 intervertebral disc. C. Fracture line in the 3D CT image (arrow). D. Multiple fracture line in the vertebral body and the lamina in the axial CT section (arrow)

learned that she had complaints of leg weakness, numbness and inability to walk for the last 4 months in addition to the pain. It was learned from the medical history of the patient that she had received treatment for osteoporosis for nearly 20 years, developed a scapula fracture after a minor trauma 5 years previously and had been diagnosed with diabetes mellitus 35 years before.

Neurological examination revealed the following observations: both ankles exhibited dorsiflexion of 3–4/5, there was a decrease in bilateral Achilles and L4-L5 hypoesthesia was evident in both legs. The pain which was described in the waist and hip was scored at 8 on the visual analogue scale (VAS). Osvestri disability index (ODI) was 85%. Significant loss of height in the corpus of the L5 vertebra and fracture lines of the vertebral body and posterior lamina were observed on lumbar computed tomography (Figure 1). It was observed that narrowing in the spinal canal had occurred due to a bone fragment, and simultaneously, diffuse bulging of the disc in the posterior direction. Hypo-intense areas adjacent to the fracture line in the L5 vertebrae in T1-weighted sequences and a liquid appearance with a significantly higher signal in T2-weighted sequences were present in magnetic resonance imaging (Figure 2). Positron emission tomography examination performed using fluorine-18 fluorodeoxyglucose (18F-FDG) for the elimination of possible malignancy; 18F-FDG uptake was observed in a heterogeneous manner in the fracture line of the left lamina of the L5

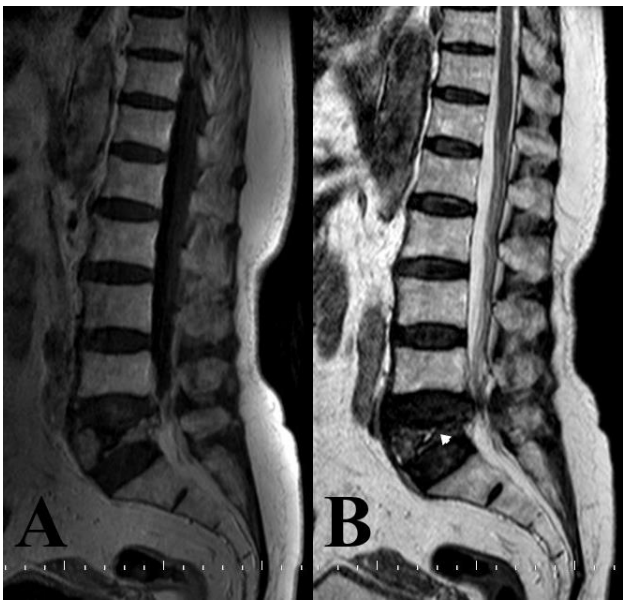

Fig. 2. A. Vertebral collapse and fracture line in the T1-weighted sagittal MR section. B. Fracture line and fluid signal in the T2-weighted sagittal MR section (arrowhead)

vertebral corpus (Figure 3). the patient due to the pain of patient increased that the pathology of malignant was not determined on the examinations

L5 trans-peritoneal anterior corpectomy and implementation of a titanium cage and L4-S1 posterior stabilisation with iliac screws were applied (Figure 4). Materials taken from the patient's L5 vertebral corpus were examined microscopically, and necrotic bone tissue was observed. The patient was mobilised using a lumbosacral corset on the first day postoperatively. Outpatient follow-up review revealed that the VAS reduced to 3/10, ODI reduced to 35% and there was no additional neurological deficits 6th month after the surgery.

Discussion

Many terms have been used to describe this disease, such as delayed post-traumatic vertebral osteonecrosis, intervertebral pseudoarthrosis, aseptic necrosis of the vertebra, intra-vertebral vacuum cleft, delayed vertebral collapse and non-union in compression fracture. Kummell's disease is becoming more frequent with the ageing of the population (1,2).

Although there are numerous theories to explain the pathophysiology of Kummell's disease, a consensus has not been achieved (4,5). The disease is thought to be caused by a deterioration of the structure supplying the vertebral body. Osteoporotic bone may lead to the development

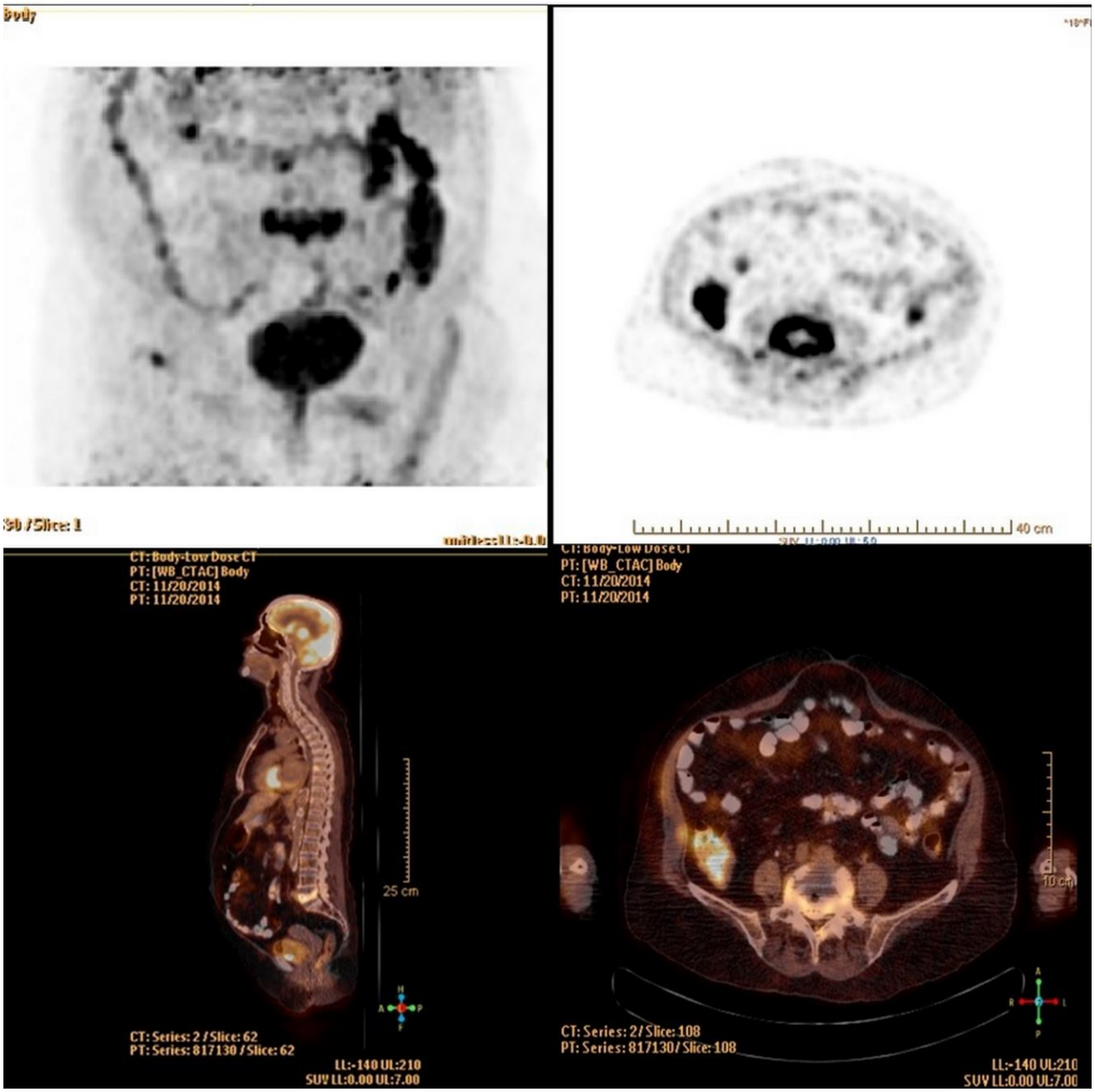

Fig. 3. Increased $_{18}$F-FDG uptake in the L5 vertebra in PET and PET-CT images

of Kummell's disease by causing the deterioration of medullary arterioles due to the presence of micro-fractures. Chronic steroid use is closely related with Kummell's disease because this can cause fat accumulation in the vertebral body and compression of the intramedullary artery. Diabetes, hypothyroidism, rheumatic diseases, pancreatitis, sickle cell anaemia, Gaucher's disease long-term bed rest are the initiators or contributing factors (5). In our case, the patient has been also diabetic and osteoporotic.

This disease, which was identified prior to the emergence of radiography, comprises four stages (1). In the first stage (stage 1: theoretical phase), the low back pain is the most important symptom; this can emerge after a more trauma or even an event that patients cannot remember. Pain is typically experienced in the lower thoracic and upper lumbar regions. Neurological symptoms are not seen in the first stage. Then, an asymptomatic period extending from months to years is observed. The second stage (instability phase) is characterised by recurrent attacks of back pain which may extend from weeks to months after the initial event. Attacks of pain emerge due to kyphotic deformity in the thoracolumbar region due to the collapse of the vertebral body. The third stage (fixed deformity phase) is characterised by the development of kyphotic deformity in the spine (2,6). In our case, there was also a symptom-

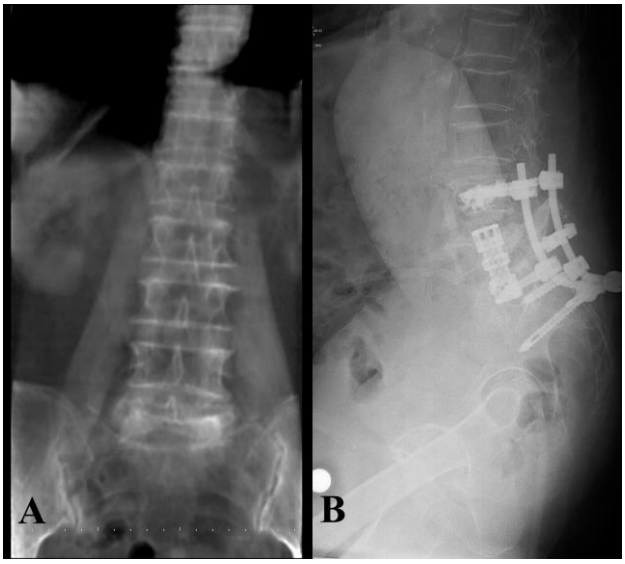

Fig. 4. A. Collapse and intra-vertebral radiolucency in the L5 vertebra in anterior–posterior lumbar spine X-ray film. B. Image of the posterior fusion material and cage in the lateral lumbar region obtained postoperatively

free period of about 4 months after a period of low back pain lasting 1 month. Following this, neurological symptoms have developed in the lower extremity in addition to the recurring and increasing back pain in the last four months.

Traumatic vertebral compression fractures are thought to occur in most, but not all cases. Post-traumatic vertebral fractures are initially asymptomatic and radiologically negative. However, the vertebral body collapse develops at the end. The affected vertebra is usually seen in the thoracolumbar region extending from T8 to L2, and is frequently the T12 vertebra (7). In our case, the L5 vertebra was affected, which is a finding that we do not encounter frequently.

The ideal diagnosis of Kummell's disease is determine using recurrent plain radiographs. While no evidence of a fracture is observed in the initial films, compression fracture, kyphotic deformity and an intervertebral air image can be monitored in advanced stages depending on the position and timing of the tests (8,9). The image called an intervertebral vacuum cleft was first described by Maldague et al (4). Initially, the accumulation of gas inside the vertebral corpus on CT and plain radiography was defined as pathognomonic for Kummell's disease (6). However, it was determined in subsequent studies that this image could be observed in some other conditions such as malignancy, intraosseous disc prolapse, infection, osteoporosis, chronic administration of steroids, radiotherapy, pancreatitis, cirrhosis, arteriosclerosis and alcohol abuse (8). Radiolucency in the central part of the L5 vertebral corpus was observed in preoperative X-ray radiography in our case, and we observed the air density in the corpus on sagittal and coronal multiplanar reconstruction images of the CT images of this area (Figure 1).

Kummell's disease usually exhibits a signal increase in T1-weighted MR images and a signal decrease in T2-weighted images. It is separated from osteoporotic vertebral fractures in that a signal change is not observed with MRI in those cases (6). Intervertebral bone marrow edema and reactive fibrosis and fluid accumulation similar to transudate have been identified. Fluid collection is observed as a well-defined hypo-intense area on T1-weighted MR images and a hyper-intense area on T2-weighted MR images. This result is referred to as the fluid sign (7). In the MRI examination in our case, a hypo-intense intra-vertebral fluid signal on T1-weighted sequences and a hyper-intense intra-vertebral fluid signal on T2-weighted sequences were present in the neighbourhood of the fracture line.

A three-dimensional (3D) image is obtained with PET. Although an increase in activity can be observed with this technique with high sensitivity, this is not specific to the lesion in practice (5). On PET images in our case, 18F-FDG uptake was observed as heterogeneous in the vertebral corpus; the circumference was hyper-metabolic and the central part was hypo-metabolic (SUVmax: 3.2) and on the fracture line of the left lamina (SUVmax: 2.9).

No consensus has been reached related with the best treatment options for Kummell disease (3). Conservative treatment methods are recommended in the initial phase of the disease for most patients, including bed rest, lumbar traction, corsets, analgesics and osteoporotic drug applications (9-10).

Surgical treatment is recommended for patients with pain which does alleviate even with conservative treatment for 3 months, with radiculopathy or progressive neurological deficits or with significant kyphotic deformity. The aim of surgery is to bring the vertebra into a normal sagittal balance and generate neural decompression. Combined anterior–posterior, posterior-only and anterior-only methods are applied in the surgical treatment of Kummell's disease (3,6,9). Generally, patients are exposed to instrument failure after operation due to osteoporosis. Therefore, the choice of surgical treatment should be decided by evaluating the bone structure of the patients and taking the

proximity to the thoracolumbar or lumbosacral transition region into consideration. Balloon kyphoplasty and vertebraplasty are used successfully in this disease for appropriate patients without neurological deficits (9). Posterior and anterior stabilization methods are applied for patients with neurological deficits (3). The correction of kyphosis can help the patient to return to active life through the placement of a titanium cage or bone graft after anterior or posterior corpectomy (9,10). Liu et al. (10) compare anterior intervention and posterior osteotomies in their study and state that anterior interventions should be frequently applied together with posterior intervention. In our case, the patient first underwent anterior trans-peritoneal L5 corpectomy and a titanium cage was placed. In the second session, the patient was stabilised with bilateral posterior L4-S1 iliac screws.

The increase in human life expectancy, and as a result, osteoporosis suggests that Kummell's disease will be encountered much more frequently in the advanced stages. This disease should be considered in patients whose low back pain emerges following an asymptomatic period and the development of kyphotic deformity. It is important to consider neurosurgery because the disease is not frequently encountered and can be treated successfully using surgical procedures.

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