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## Case Report

# Kienböck's Disease in a Patient with Hemiparetic Stroke: A case report

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Kienböck's disease of the wrist is a multifactorial, progressive osteonecrosis of the lunate bone. Patients with this disease often complain of dorsal wrist pain and other wrist dysfunctions. Image studies are usually required for definite diagnosis; however, careful history taking and physical examination are also necessary. Treatment varies according to the stage of the disease; although surgery is needed in most of the cases, rehabilitation after the surgery seems helpful as well.

Kienböck's disease is rarely seen in our daily practice, and even though we do recognize the disease, diagnosing it correctly in patients with comorbidities such as stroke is often difficult. We thereby present a case of a patient with hemiparetic stroke in our ward who complained of shoulder and wrist pain. The patient was diagnosed of shoulder-hand syndrome first, but he then turned out to be a victim of Kienböck's disease. This case report provides an important insight for physiatrists to have a second thought of any refractory wrist pain in a patient with stroke even though it is being treated as a more prevalent disease. Furthermore, physiatrist could further instruct the therapists to adjust their rehabilitation program accordingly and collaborate with orthopedic surgeons to provide a more thorough care for the patients. ( Tw J Phys Med Rehabil 2020; 48(2): 133 - 140 )

**Key Words:** Kienböck's disease, hemiparesis, stroke, rehabilitation, wrist pain

## INTRODUCTION

Kienböck's disease, also known as lunatomalacia, is the avascular necrosis of the lunate. It was first proposed by Dr. Robert Kienböck in 1910, who was a radiologist from Vienna.<sup>[1]</sup> The disease is more common in males aged between 20 and 40; however, it can still occur in younger and older populations. Interestingly, it's more often seen in their middle age in female patients.<sup>[2]</sup>

Patients with Kienböck's disease usually presented with pain, weakness, swelling, and decreased range of motion (ROM) in the dominant hand; however, some evidence showed that both sides are equally affected, especially in women.<sup>[3]</sup> Patients usually complain activity-related dorsal wrist pain with or without swelling, and decreased flexion and extension of the wrist are also common. Some of the patients report weakness in gripping as well. Upon examination, local tenderness at the radiocarpal joint is frequently seen.<sup>[2]</sup> Special tests associ-

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ated with the lunate such as Reagon's ballot test and Waston's test might be positive.

Conservative treatment such as splint immobilization and nonsteroidal anti-inflammatory drugs (NSAIDs) is given in stage I, whereas surgery is recommended for later stages.<sup>[4]</sup> However, some found that a majority of patients with stage I Kienböck's disease still required surgery due to ongoing degeneration.

We hereby reviewed a case of Kienböck's disease to provide important insights in diagnosis and further improve the clinical strategy in medical care. Although this disease is not frequently seen in our daily practice, it warrants for a physiatrist to identify this unique disease promptly and provides a combined care with orthopedic surgeons.

## CASE REPORT

A retired 72-year-old man with a history of hypertension, type II diabetes mellitus, gout, dyslipidemia and gastric ulcer was admitted to our ward on 2020/3/26 due to left corona radiata infarction on 2020/2/7 with right hemiparesis.

Upon admission, his Brunnstrom stage was rated as IV-V and his muscle power was 3 out of 5 for right upper and lower limbs. Standard medical care of physical and occupational therapy including balance training, muscle strengthening, functional training and activities of daily living (ADL) training were provided.

Soon after admission, the patient complained right shoulder, hand and knee pain. His right knee was treated with echo-guided intra-articular (IA) dextrose injection after osteoarthritis was confirmed by X-ray and musculoskeletal (MSK) ultrasound. As for his right shoulder and hand pain, shoulder-hand syndrome was first suspected due to the clinical presentation such as local swelling, hyperalgesia and the elbow-sparing pattern; however, right shoulder ultrasound was still performed and revealed diffuse supraspinatus tendinosis with thickened subacromial-subdeltoid bursa. Thus, both echo-guided shoulder intra-articular and bursal steroid injections were provided as treatment.

The right shoulder and knee pain became much better after the injection; nevertheless, his right hand pain persisted even with the use of celecoxib (Celebrex). The

pain was most prominent while undergoing occupational therapy, especially when using the Reha-Slide. Celebrex was once changed to clonazepam (Rivotril) under the impression of shoulder-hand syndrome, but the pain persisted.

The pain was reassessed because of the poor response of treatment. The pain mainly located at the right wrist and exaggerated with wrist extension and flexion. Local tenderness was also noted at dorsal side of wrist. Furthermore, the Reagon's and the Watson's tests were both positive. A plain film of right wrist was performed and showed no obvious fracture but moderate joint narrowing. Furthermore, although there was no obvious sclerosis of the lunate bone in the plain film, patchy radiolucency was noted (Figure 1A and 1B). Next, MSK ultrasound of right wrist was performed and showed irregular bony cortex of the lunate without hyperemia (Figure 2A, 2B, and 2C).

Therefore, with the aforementioned clinical clues, Kienböck's disease was suspected. Consequently, a right wrist MRI (Figure 3A, 3B, 3C, and 3D) was arranged and the result showed bone marrow edema of right lunate which was suggestive of tissue necrosis, while no definite lunate collapse, negative ulnar variance, rotation of scaphoid or wrist joint osteoarthritis was noted. Since the results of MRI were compatible with stage II Kienböck's disease, an orthopedic surgeon was consulted. The surgeon suggested the use of cock-up splint and arranged the elective surgery in an outpatient clinic after discharge.

## DISCUSSION

Kienböck's disease is a rare etiology contributing wrist pain. After searching the Cochrane library, Pubmed, Google scholar and Uptodate using the keywords "Kienböck's/Kienbock's disease", along with "stroke" or "infarction" or "hematoma" or "hemorrhage" or "hemiplegia/hemiplegic" or "hemiparesis/hemiparetic", this is, to our knowledge, the first case reported in patients with hemiparetic stroke. In these patients, the symptoms of Kienböck's disease might often be masked by a more prevalent disease, the shoulder-hand syndrome; therefore, making the correct diagnosis is difficult. If Kienböck's disease is not diagnosed promptly or not properly treated in these patients with stroke, wrist joint arthropathy and

severe functional impairment may inevitably develop. Alternatively, if the disease is identified in time, we could adjust the rehabilitation program for these patients and work with orthopedic surgeons to bring them a better outcome.

#### Our patient with a stage II Kienböck's disease in subacute phase of stroke

Our patient was an elderly man whose dominant

hand was diagnosed of stage II Kienböck's disease with neutral ulnar variance. Theoretically, radial wedge osteotomy, distal radius core decompression or revascularization procedure should be performed;<sup>[5]</sup> however, the condition of this patient was more complicated because of the subacute phase of stroke. Therefore, the temporary use of cock-up splint and elective surgery arranged after the stroke rehabilitation course seemed to be a reasonable alternative for him.

Table 1. Lichtman classification of Kienböck's disease

Stage	Image findings
Stage I	Normal plain film appearance; decrease signal in T1-weighted MRI, T2-weighted signal is variable
Stage II	Lunate sclerosis without collapse
Stage III	Lunate collapse
III A	Without carpal collapse
III B	With carpal collapse
Stage IV	Radiocarpal or midcarpal joint arthritic change



Figure 1. Right wrist plain film in (A) anterior-posterior view and (B) lateral view. Radiolucent patches were noted within lunate bone with minimal sclerosis. No obvious rotation of scaphoid was noted.

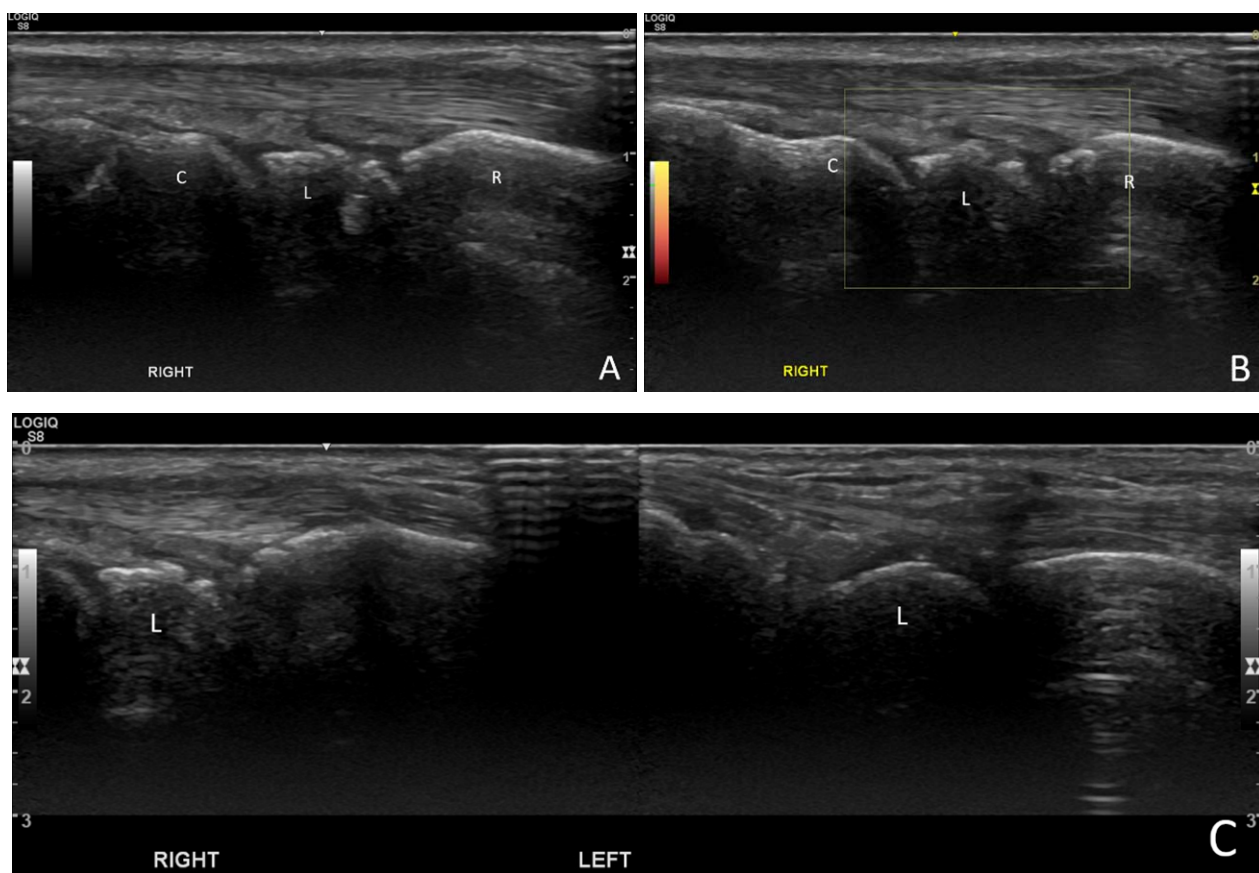
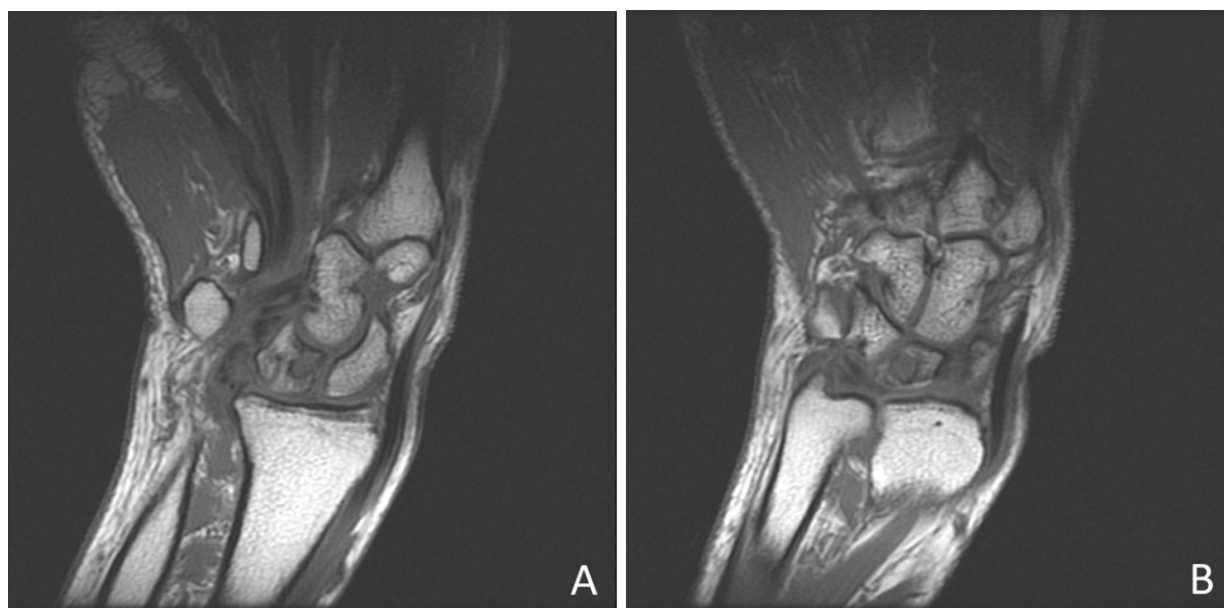


Figure 2. Ultrasound of wrist in sagittal plane. (A) Right radiocarpal joint (B) right radiocarpal joint under power Doppler (C) comparison of left and right radiocarpal joint. Note the cortex of the right side lunate is much more irregular than the left side. C, capitate; L, lunate; R, distal radius.



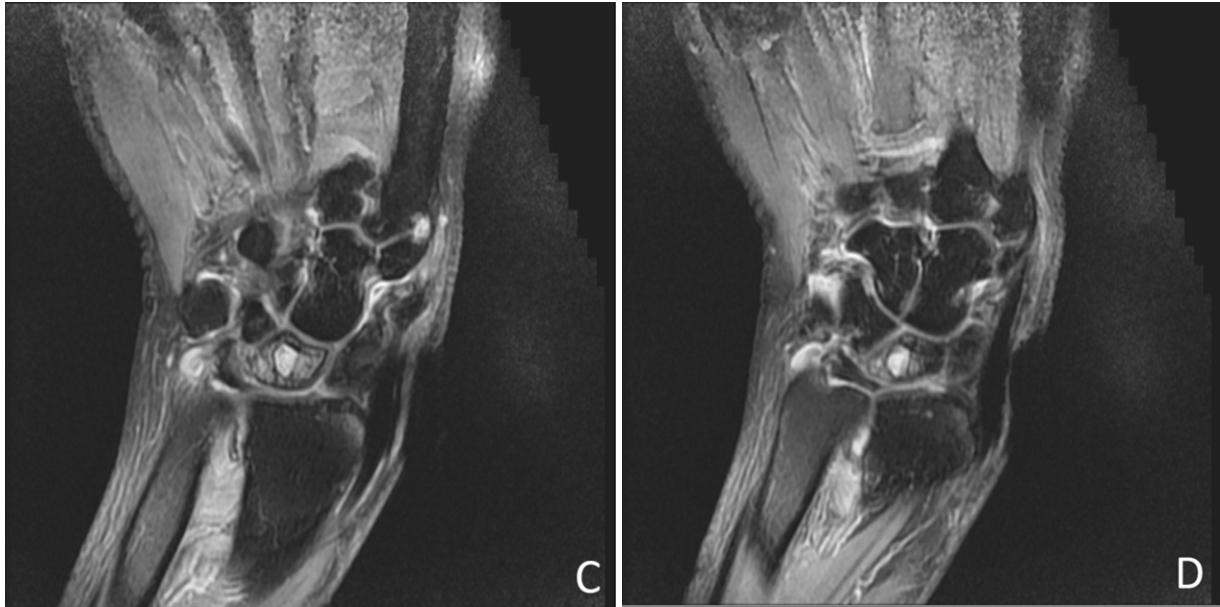


Figure 3. MRI of the right wrist. The lunate showed decreased signal in T1 weighted images (A and B) and increased signal in proton density (PD) weighted images (C and D). Also, the results of MRI showed no negative ulnar variance of this patient.

Although the actual cause of Kienböck's disease is still uncertain, several hypotheses of its etiology exist.<sup>[6]</sup> Some claimed that negative ulnar variance might lead to increased pressure on the lunate, whereas others thought that variation of blood supply plays a more important role: those having only a single vessel running through their lunate carry a greater risk of having this disease.<sup>[7]</sup> Other factors having been advocated include repetitive microfractures during the daily use of wrist and the bone geometry of the lunate.<sup>[8]</sup>

The actual etiology of Kienböck's disease in this patient is unclear and might be multifactorial, just as most of the cases.<sup>[9]</sup> Reviewing his history, we speculate the intensive rehabilitation of his affected right upper limb might be one of the aggravating factors of the disease. In occupational therapy, a majority of modalities such as the Reha-slide, climbing bar or table-top shoulder/arm wheel may increase the pressure on the lunate, whether any resistance was added or not, especially when the wrist was not fixed properly. The transfer training and sit-to-stand training in a physical therapy program might also contribute to a certain degree of stress on the carpal bones.

The day-to-day rehabilitation of the upper limb in-

cluding the wrist surely has benefit on patients' hemiparetic status and shoulder-hand syndrome; however, the repetitive microtrauma to the wrist from the daily rehabilitation program might accelerate the progression of Kienböck's disease.<sup>[9]</sup> Frequent reassessment of the type, intensity, and density of rehabilitation program according to the response and condition of the patient is therefore critically important for a physiatrist.

#### Differentiating Kienböck's disease and other upper limb diseases in patients with stroke

Upper limb pain of the affected side is quite often seen in a patient with stroke. Possible differential diagnoses include shoulder subluxation, rotator cuff tendinopathy, secondary frozen shoulder and shoulder-hand syndrome.<sup>[10]</sup> Although wrist swelling and pain are common in shoulder-hand syndrome<sup>[11]</sup>, isolated wrist swelling/pain or residual wrist pain in properly treated shoulder-hand syndrome should stir up the suspicion of other diagnoses, such as Kienböck's disease, at all times.

Besides the history taking and physical examination, the diagnosis and staging of Kienböck's disease rely on plain film and magnetic resonance imaging (MRI), the four-stage Lichtman classification is currently being

used.<sup>[5]</sup> Stage I Kienböck's disease appears normal on plain film, whereas MRI shows positive findings. Other findings such as lunate sclerosis, collapse and osteoarthritis are found in patients with stage II, III or IV Kienböck's disease (Table 1).

Plain film is useful in screening the disease, it could tell us not only bone fractures or joint osteoarthritis but also subtle changes like those found in this case report. Even though no obvious lunate sclerosis was noted, we should be cautious about the abnormal radiolucency in the lunate or any other carpal bones. MRI usually helps identify the very early stage of Kienböck's disease, which showed decreased T1 signal and variable T2 signal of the lunate. Additionally, MSK ultrasound showed its usefulness in diagnosing this patient by detecting the irregular cortex of the lunate. MSK ultrasound was considered helpful in delineating osteoarthritis as well as osteonecrosis in the hips,<sup>[12]</sup> and in our opinion, screening osteonecrosis in the wrist is also possible. Some also claimed that bone scan may show increased uptake of the lunate but it's thought to be nonspecific.

### Treatment and rehabilitation for Kienböck's disease in patients with stroke

The wrist pain of this patient is, again, complicated. It was not only shoulder-hand syndrome but also superimposed by a Kienböck's disease. Shoulder-hand syndrome is relatively common in patients with stroke; therefore, well-trained physiatrists could handle patients with shoulder-hand syndrome smoothly using NSAIDs, IA shoulder steroid injection, suprascapular nerve block and stellate ganglion block.<sup>[13,14]</sup> Nevertheless, Kienböck's disease is a rare disease<sup>[15]</sup> in either normal population or patients with stroke warranting for surgical interventions. If left untreated, the disease will progress to a greater extend and later stage, for which a more aggressive surgery is often warranted, such as proximal row carpectomy and partial or total wrist fusion.<sup>[4]</sup> As a result, we should always try our best to detect the disease earlier and collaborate with an orthopedic surgeon as soon as possible.

Rehabilitation (e.g., kinesiotherapy, electrotherapy, thermotherapy, and therapeutic exercises) following surgery of Kienböck's disease has brought benefits to the patients in a previous study.<sup>[16]</sup> Higher satisfaction and

better outcomes regarding pain, muscular strength, and ROM were presented in the intervention group than in the standard care group in their study. Similarly, other studies revealed that low-intensity pulsed ultrasound, massage, and taping after surgery improved the outcome as well.<sup>[17,18]</sup> In addition, both patients with stroke and with cerebral palsy may present strong flexor spasticity of the wrists in conjunction with the daily use of assistive devices, which in turn caused hyper-pressure and decreased venous return.<sup>[19-21]</sup> Local injection of Botulinum toxin into the flexor tendon have been found to relieve pain and cease the progression of Kienböck's disease in patients with cerebral palsy.<sup>[19]</sup> Therefore, patients with concurrent Kienböck's disease and stroke may consider the local injection as well. While this proposition regarding effects of treatment is only tentative, the hypothesis requires further validation.

## CONCLUSION

Kienböck's disease is a relatively rare etiology of wrist pain in our daily practice; however, it may bring suffering to our patients and could eventually lead to wrist joint osteoarthritis and wrist stiffness if not treated properly. In patients with stroke, diagnosing the disease is more challenging since other prevalent diseases may mask the symptoms and signs of Kienböck's disease. This case report presented a patient with both stroke and Kienböck's disease, and it also raised the awareness of healthcare providers regarding the symptoms and signs of Kienböck's disease. If physiatrists are cautious about the treatment response of patients and meticulous on the subtle findings in physical exams and imaging studies, we could detect the disease in time and remind the therapists to adjust their rehabilitation programs to avoid further harm to the patients' wrists while still maintaining the effectiveness of stroke rehabilitation. On the other hand, physiatrists can further collaborate with orthopedic surgeons regarding postoperative rehabilitation to achieve the best outcome for the patients.

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## 半側偏癱中風病人合併月狀骨無菌性壞死：病例報告

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發生在手腕上的月狀骨無菌性壞死是一種多因素造成的月狀骨缺血性壞死。患者通常會抱怨手腕背側疼痛，並伴隨一些手腕功能上的限制。此病的確切診斷通常需要仰賴影像學檢查，但細心的病史詢問以及理學檢查仍然是不可或缺的一環。治療的部分會依據不同的疾病嚴重度分級而有所不同，大部分的此類病人需要外科手術的介入，而文獻也顯示術後的復健也有助於病人功能恢復。

月狀骨無菌性壞死在一般的病房或門診都算不常見，即便我們知道且了解這個疾病，但在一些有各種共病症的病患身上也很難診斷出來，例如中風病患。因此，我們在此報告一個住院中抱怨肩膀及手腕疼痛的腦中風半側偏癱患者。他一開始被診斷為肩手症候群，但後來也被證實同時患有月狀骨無菌性壞死。這個案例可以提醒復健科醫師，當中風病人被診斷罹患了其他盛行率較高的疾病，若治療後手腕的疼痛仍然持續，必須重新審視是否可能有其他潛在的病因存在。而復健科醫師在正確診斷後，也可以進一步引導治療師改變其治療策略，並且跟骨科醫師合作，以提供病人更好且更全面的照護。（台灣復健醫誌 2020；48(2)：133 - 140）

**關鍵詞：**月狀骨無菌性壞死(Kienböck's disease)，半側偏癱(hemiparesis)，中風(stroke)，復健(rehabilitation)，手腕疼痛(wrist pain)