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(CASE REPORT)

Long-term functional result of vascularized bone graft for kienbock disease: A case report

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Abstract

Introduction: Kienbock's disease, or lunatomalacia, was a condition of aseptic osteonecrosis of the os lunatum, which can be progressively developed with a state of lunatum collapse, followed by complete os carpal collapse. This disease has been known for a long time, but until now, the exact cause of this disease could not be explained. .

Case Presentation: We reported a 32-year-old female patient with pain in the right wrist; the pain was felt for five years before the surgery and is getting worse. On physical examination, Phalen's and Reverse Phalen's tests found pain and limited movement in the right wrist area. A collapse of the os lunatum was found after a radiological examination. The evaluation using the DASH score was an improvement in clinical conditions with the absence of pain on the right wrist after surgery using a vascularized bone graft. The clinical evaluation using Phalen's and Reverse Phalen's tests showed normal flexion and wrist extension results with no movement limitation around 0-90° flexion-extension. The wrist pronation and supination also showed normal results with pronation angles around 0-75° and supination angles around 0-85°.

Conclusion: There were clinical, functional, and radiographic improvements in younger female patients with Kienbock's Disease treated with vascularized bone graft surgery six years after surgery.

Keywords: Neglected; Kienbock disease; Vascularized bone graft; DASH score

1. Introduction

The hand is the distal part of the body of the upper limb, and the location of the hand is separated from the forearm by the wrist. The hand can be positioned at a desired height or location relative to the body with movements in the shoulders and elbows [1]. The bones in the hand are composed of bones in the wrist, the metacarpal, and the fingers.1 Bones in the wrist include scaphoid, lunatum, triquetrum, psiformis, trapezium, trapezoid, capitatum and hamatum.[2]

Complaints of pain in the wrist can be caused by Kienbock disease. Kienbock's disease, or what can be known as lunatomalacia, is a state of aseptic osteonecrosis of the lunatum bone, which can progressively develop with a state of collapse of the lunatum bone, followed by a complete collapse of the carpal bone. This disease was first described about 100 years ago, but until now, the exact cause of the disease has not been explained.[3]

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Several intrinsic and extrinsic factors have been carried out in research to find the etiology of necrosis of the lunatum bone. Kienbock's disease is a progressive disease that runs slowly and can be recognized easily with a variety of examination options. However, the effectiveness of the examination needs to be known because Kienbock's disease is very rare in number and requires a long-term examination evaluation.[4]

2. Case Report

A female patient aged 32 years with the main complaint of pain in the right wrist. Patients present with complaints of pain in the right wrist for the previous six years. Complaints have been felt since 2006; there was no previous history of falls. Initially, complaints of pain were felt not to interfere, namely when moving and pointing the thumb of the right hand, but the longer the pain complaints in the wrist became heavier, so the patient could not lift his right wrist.

Daily, patients have activities, often making cakes by stirring the dough with his right hand, and the activity has been carried out since before 2006 and continues to this day. Physical examination found that the patient complained of pain in his wrist, with pain in the back of the right wrist, accompanied by weakness and disregard for movement. At the time of physical examination, it was found that the patient complained of pain during the examination of Phalen's and Reverse Phalen's. In this examination, the flexion and extension of the wrist are performed. From the examination results, there were restricted movements of the wrist, which complained of pain, namely the wrist dextra. The clinical conditions are shown in Figure 1. Functional examination using DASH found a score of 84.

Figure 1 Clinical photograph of patient's wrist during (a) Phalen's test and (b) Reverse Phalen's test. We can also observe limited flexion and extension on the patient's wrist (red arrow). (source: internal documentation)

The preoperative radiological examination was performed using a plain radiograph initial wrist with posteroanterior and lateral projections, as seen in Figure 2.

Then, another radiological investigation is carried out using magnetic resonance imagery (MRI) for visualization of the surrounding bone and soft tissue with various cross-sections, according to Figure 2C. The result is a picture according to Kienbock disease: lunate collapse appears to be present with anteroposterior elongation lunate in the sagittal incision and accompanied by proximal migration of the os capitatum—scapholunate dissociation with abnormal hyperintensity in the scapholunate ligament and localized synovitis with radiocarpal joint arthrosis.

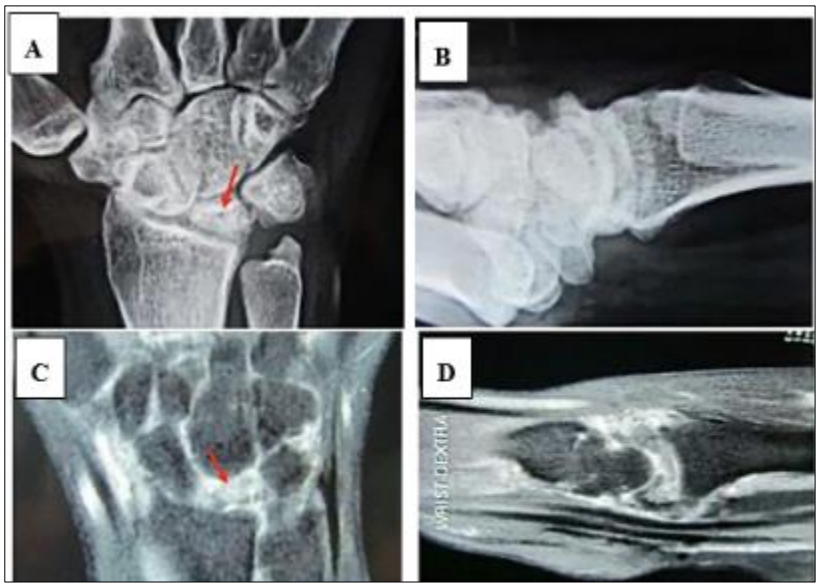

Figure 2 Plain radiograph of initial wrist dextra, (a) PA projection and (b) lateral projection. Overview of inisial wrist MRI (c) Coronal view and (d) Sagital view. Observable in the x-ray and MRI is the osteonecrosis of the lunate (red arrow). (source: internal documentation)

After undergoing a series of physical and supportive examinations, the diagnosis of the case was stated as a state of avascular lunatum called Kienbock disease. Kienbock's disease experienced stage 3 when the patient first screened the examination. At this stage, surgery is the primary choice in management. The patient underwent surgery in December 2012 in the form of a vascularized bone graft originating from the distal radius to provide blood vessels to the lunatum bone with a surgical incision carried out in a dorsal approach to the wrist. The incision begins with the incision marking in the dorsal region of the wrist, according to Figure 3.

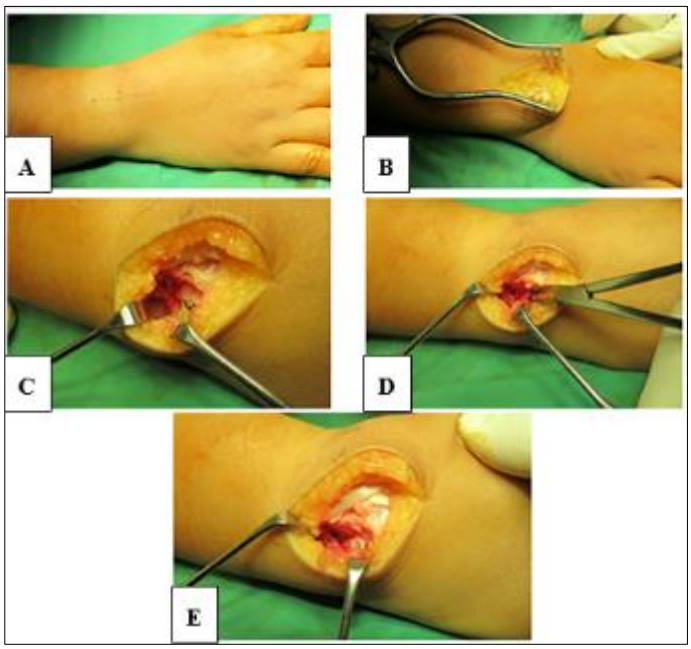

Figure 3 Stages of vascularized bone graft surgery on os lunatum. (A). Incision mark before incision is made; (B). Dorsal wrist exposure through incision mark; (C). Taking vascularized bone graft from 4.5 ECR with ligase; (D) Vascularized bone graft placed on the os lunatum with fit press; (E) Placement of vascularized bone graft on os lunatum. (source: internal documentation)

The incision on the dorsal wrist is then explored. In this case, a vascularized bone graft from the 4.5-compartment extensor artery by using a tourniquet was used to control bleeding during surgery. After the extensor artery, 4.5-

compartment are identified, and ligation is carried out proximal to the branching area. The os lunatum explored and extracted the necrotic tissue and prepared it for planting the graft on the side of the collapsed defect. In this case, external fixation was given, namely the administration of Long Arm Cast immobilization for one month for recurrence of the collapse after revascularization with vascularized bone graft.

Figure 4 A plain photo of post-operative wrist post-operative evaluation: (A) Posterior-anterior projection and (B) Lateral projection. Observable on the plain photo is the lunate bone filled with bone graft (C) Post-operative MRI 3 Tesla sagittal cuts. (source: internal documentation)

The patient evaluation was carried out in the form of a radiographic examination of the radial wrist extra with posteroanterior and lateral projections on September 20, 2018 (Figure 4). The results obtained showed a hyperdense lunatum bone. Then, the patient was evaluated by an MRI examination (Figure 4C). The results show a vascularized bone graft given to the os lunatum viable while still maintaining the carpal height.

On physical examination after surgery, Phalen's and Reverse Phalen's examination. In this examination, the flexion and extension of the wrist are performed. From the results of the examination, there was no restricted movement of the wrist dextra. DASH scores were improved to 14. Likewise, in the wrist rotation examination, there was no pain and restricted complaints from rotational movements with a pronation angle of 75^{o} and a supination angle of 85^{o} as shown in Figure 5.

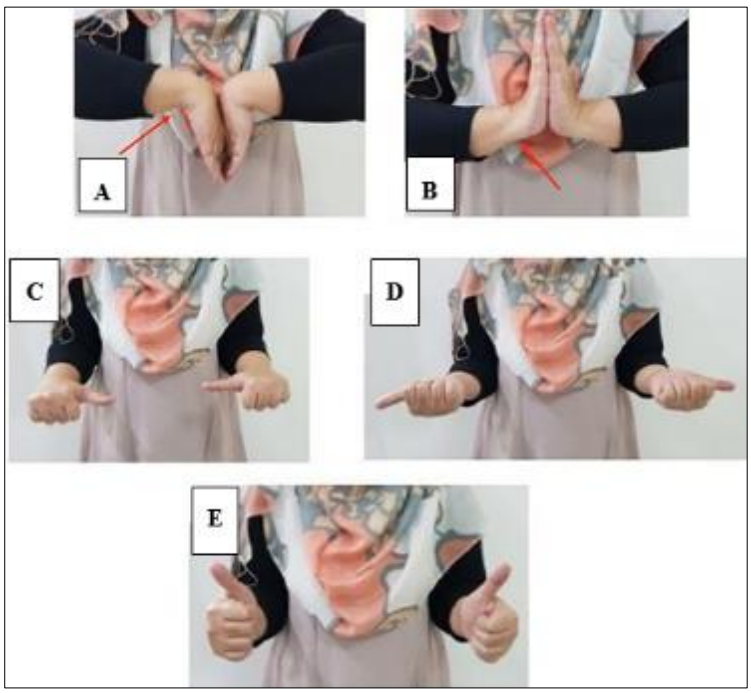

Figure 5 Post-operative examination of Phalen's test (A) and Reverse Phalen's test (B) found normal flexion and extension (red arrow); Post-operative range of motion wrist rotational examination shows no restrictions on joint motion. Observed are pronation (C), supination (D) and neutral position (E) (source : internal documentation)

Post-operative evaluation results showed clinical improvement of ROM rotation and flexion extension on the wrist showed normal results, namely pronation of 75^{o}, supination of 85^{o}, flexion and extension of 90^{o}. Besides, the improvement of pain complaints is no longer felt.

3. Discussion

Kienbock disease is an avascular necrotic condition of the lunatum bone, which is also known as osteonecrosis, lunatomalacia, and aseptic or lunatum ischemic necrosis, which can be accompanied by a progressive collapse of the quantum bone and followed by the collapse of the carpal so that it can cause wrist instability.[3,9]

Kienbock's disease was first described in 1910 by Robert Kienbock. [8] Robert Kienbock, a radiologist from Austria, explains that the process of this disease and the accompanying clinical disease are caused by impaired vascular supply of nutrients to the lunatum bone.[4] A history of trauma is often reported in patients with this disease, including minor trauma or repetitive trauma.[3]

The pathophysiological mechanism of Kienbock disease is multifactorial and cannot be explained. Anatomic factors associated with increased shear force on the lunatum bone have a role in Kienbock's disease.[4]

Hulten et al. Explain that there is a negative variant ulnar relationship in Kienbock's disease that cannot be explained.[3] The incidence is found in about 78% of cases.4 Biomechanical studies have shown that the negative variant ulnar has an essential role in the development of the collapse of the lunatum bone. Moreover, several other authors found changes in the ulnar variant associated with age, sex, wrist position, and secondary osteoarthritis of Kienbock's disease.[3]

Morphological factors also have a role in the causes of Kienbock's disease.[3] Square or rectangular geometry formation on the lunatum bone as in the classification of Antuna Zapico type I and type II, which has an angle of more than 110 degrees, which will pose a greater risk of Kienbock's disease.[4]

Many surgical procedures have been described but have not been proven to improve the natural causes of the disease. Management includes mechanical techniques to relieve pressure or burden on the lunatum to increase spontaneous revascularization, graft procedures directly on the lunatum bone, and several other aggressive options, such as carpectomy on the proximal side, joint replacement, and fusion of the carpal bone.[4] A thorough wrist evaluation is needed for proper management selection, including a biomechanical evaluation of the effects of lunatum bone collapse and the biological effects of wrist stiffness that may be related to synovitis and/or the resulting fibrosis. In addition, the presence or absence of osteoarthritis may be a management consideration between reconstructive and salvage procedures.[3]

One management that can be done is by revascularization. This management can be achieved directly by the implantation of blood vessels or indirectly through vascularized bone graft (VBG) in grades 1 and 2 and at stage 3A (the collapse of the lunatum without any rotation of the scaphoid bone).[3,7] A graft can be a free graft (bone) or a pedicle graft.[3] Vascularized bone graft, which is often used in the management of Kienbock's disease, is arterial blood vessels 4,5 ECA (Extensor Compartmental Arteries).[7]

Managing vascularized bone graft in Kienbock's disease has the advantage of improving vascular insufficiency. Donor source variations include second dorsal metacarpal artery, pedicle vascular pisiform, quadratus bone flap pronator, dorsal distal radius pedicle grafts, nonvascularized bone grafting with core revascularization, and free vascularized iliac crest bone grafting. The vascularized bone graft should be used in patients with intact cartilage tissue and no degenerative abnormalities.[11]

4. Conclusion

There were clinical, functional, and radiographic improvements in younger female patients with Kienbock's Disease treated with vascularized bone graft surgery six years after surgery. Further evaluation with a greater number of cases and a longer term of evaluation is needed to assess the efficacy and survival using this technique.

Compliance with ethical standards

Disclosure of conflict of interest

There is no conflict of interest in this study.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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