DOI: 10.7860/JCDR/2025/76055.21424

Case Report

Anatomy Section

Advanced Kienbock's Disease with Chronic Regional Pain Syndrome: A Case Report

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ABSTRACT

Kienbock's disease is a rare and painful disorder of the wrist, of unknown etiology, characterised by osteonecrosis of the lunate bone visible on wrist radiographs. It is typically seen in individuals aged 20-40 years, often in the dominant wrist, with a history of trauma. If left untreated, it can lead to fragmentation of the lunate, collapse and shortening of the carpus, and secondary arthritic changes throughout the proximal carpal area. We present a case of Kienbock's disease Lichtman stage IV with Chronic Regional Pain Syndrome (CRPS) in a 53-year-old female who presented with right wrist pain and swelling persisting for one month, alongside a loss of range of motion of about 20 degrees in palmar and dorsal flexion, following a fall on an outstretched hand one year prior. Initially, the patient was managed conservatively with splinting, physiotherapy, and pharmacotherapy (Vitamin C, Vitamin D, Diclofenac) until the CRPS subsided. Subsequently, the patient underwent total wrist arthrodesis with bone grafting, resulting in pain relief and a satisfactory pain score.

Keywords: Arthrodesis, Aseptic osteonecrosis, Avascular bone necrosis, Avascularity, Reflex sympathetic dystrophy, Causalgia, Lunate bone, Osteochondrosis, Stiff wrist

CASE REPORT

A 53-year-old female presented with right wrist pain {Numeric Rating Scale (NRS) 7} and swelling for one month. She had a history of a fall on an outstretched hand one year ago, which was managed conservatively with a slab for four weeks, though no records or radiographs were available. The patient also had type 2 diabetes mellitus, controlled by medication.

Examination revealed diffuse swelling of the right wrist with severely restricted movements, local temperature rise, and erythema without trophic changes. There was no elbow or shoulder pain.

X-rays of the right wrist showed advanced lunate collapse and sclerosis, along with degenerative changes in adjacent intercarpal joints and severe osteoporosis (see [Table/Fig-1]). The diagnosis was advanced carpal collapse of the right wrist secondary to Kienbock's disease, Lichtman stage 4, with Chronic Regional Pain Syndrome (CRPS), confirmed by radiographs and the Budapest criteria, while ruling out other diagnoses.

[Table/Fig-1]: Preoperative X-ray right wrist shows advanced collapse and sclerosis of lunate with advanced degenerative changes in adjacent intercarpal joints with severe osteoporosis.

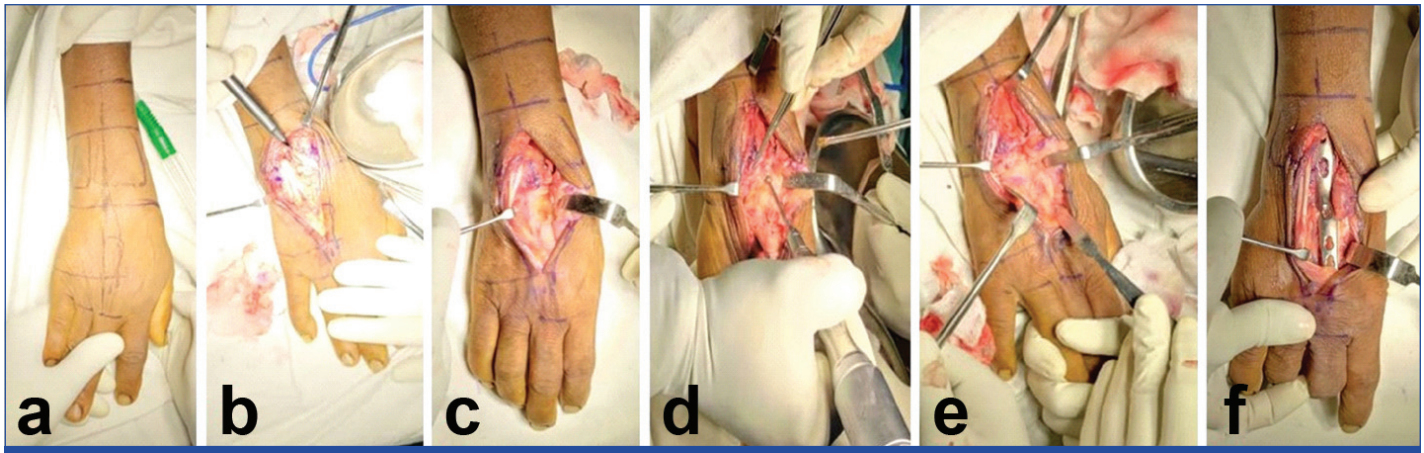
The initial protocol consisted of conservative management with splinting, physiotherapy, and pharmacotherapy (Vitamin C, Vitamin D, Diclofenac) for six weeks, until the CRPS subsided. Once the wrist showed no local temperature rise or swelling, the patient was planned for total wrist arthrodesis with bone grafting for Kienbock's disease.

The patient was positioned supine, with her hand on an arm table, and an arm tourniquet was applied. A 15 cm longitudinal dorsal incision was made at the radiocarpal joint and Lister's tubercle. The extensor retinaculum was incised between the third and fourth compartments in a Z fashion, exposing the distal radius, carpus, and long finger metacarpal. Decortication of the scaphoid, lunate, capitate, and long finger carpometacarpal joint was performed. A contoured dynamic seven-holed compression plate was fixed with six 3.5 mm cortical screws (three screws for the radius and three screws for the metacarpals). An iliac crest graft of 15 cc volume was placed over the exposed bone surface (see [Table/Fig-2]).

Postoperative pain management was initiated with a Buprenorphine patch (10 mg) and aqueous Diclofenac injections as needed, transitioning later to oral Diclofenac tablets and vitamin C supplements. Postoperatively, the patient was splinted with a below-elbow slab for six weeks, followed by daily physiotherapy for two weeks, active and passive finger movements while sparing the slab for three weeks, and finger and MCP joint mobilisation after three weeks for an additional three weeks. Non-axial weight-bearing with a load of 2 kg was allowed for three months. The patient achieved good bony union within one year (see [Table/Fig-3]).

DISCUSSION

Kienbock's disease has an incidence of 0.0066% in the general population, with a prevalence of 0.5%, which increases to 1.1% - 2% in populations exposed to strong vibrations, such as workers using jackhammers [1]. Other risk factors include industrial vibrator operators, sickle cell disease, and lupus. It is considered CRPS when the pain from Kienbock's disease becomes widespread, involves abnormal sensitivity to touch, and is accompanied by changes in skin temperature and color. In such cases, it can be classified as CRPS, particularly CRPS type I, which is not associated with direct nerve injury [1].



[Table/Fig-2]: Intraoperative pictures showing: a) dorsal wrist incision based on lister tubercle; b) incision on extensor retinaculum between 3rd and 4th compartment exposing extensor polices longus and extensor digitorum communis; c) tendons retracted and periosteum and wrist capsule elevated to expose bones; d,e) decortication was performed; f) A seven holed compression plate was fixed.

[Table/Fig-3]: Postoperative radiographs showed bony union at radiocarpal and third carpometacarpal joints at one year.

Kienbock's disease has an incidence of 0.0066% in the general population, with a prevalence of 0.5%, which increases to 1.1% - 2% in populations exposed to strong vibrations, such as workers using jackhammers [1]. Other risk factors include industrial vibrator operators, sickle cell disease, and lupus. It is considered CRPS when the pain from Kienbock's disease becomes widespread, involves abnormal sensitivity to touch, and is accompanied by changes in skin temperature and color. In such cases, it can be classified as CRPS, particularly CRPS type I, which is not associated with direct nerve injury [1].

The co-existence of CRPS, characterised by pain, swelling, and temperature changes, further complicates the clinical picture, necessitating a multifaceted approach to treatment [2,3]. Management of CRPS in this patient required a staged approach, initially focussing on symptomatic relief with splinting, limb elevation, and pharmacological agents including vitamin C and calcium supplements. The resolution of CRPS symptoms allowed for subsequent surgical intervention, highlighting the importance of addressing the inflammatory and pain components prior to definitive orthopaedic procedures [4].

In stages I and II of the disease, before the onset of sclerosis and collapse, conservative management can be attempted with casting and repeat X-rays at a later date to check for occult fractures or Avascular Necrosis (AVN) changes of the lunate. When the disease has progressed to stage IV with secondary arthritic changes throughout the wrist, proximal carpal row resection and wrist arthrodesis become the treatment of choice [5].

If left untreated, Kienbock's disease will lead to fragmentation of the lunate, collapse with shortening of the carpus, and secondary arthritic changes throughout the proximal carpal area [6].

Wrist arthrodesis was performed on this patient because she was in stage IV of Kienbock's disease with proximal carpal collapse. Furthermore, arthrodesis provides faster pain relief compared to other treatment modalities such as proximal row carpectomy or limited intercarpal fusion. Decortication of the dorsal fourth of the scaphoid, lunate, capitate, and long finger carpometacarpal joint was done to promote bony fusion of the joint [7]. The ulnar midcarpal joint and the first carpometacarpal joint were not decorticated, as no arthritic changes were noted in those areas. The patient was placed in a slab on the volar aspect extending from the metacarpal head to the proximal third of the forearm [8].

Wagner ER et al., conducted a study on long-term functional outcomes after bilateral total wrist arthrodesis, showing that out of the 13 patients who underwent unilateral total wrist arthrodesis, 12 (93%) returned for contralateral wrist arthrodesis [9]. The case study by Omor Y et al., involves a 17-year-old male with stage III Kienböck's disease, diagnosed via Magnetic Resonance Imaging (MRI) and radiography, who presented with wrist pain, limited motion, and reduced grip strength [10]. This patient declined surgical intervention, opting instead for conservative management involving immobilisation and activity restriction.

Both cases highlight the importance of early imaging in diagnosing Kienböck's disease but differ significantly in patient demographics, stages, and management approaches [10]. This case underscores the significance of early diagnosis and comprehensive management of wrist injuries to prevent progression to advanced disease stages. It also emphasises the need for individualised treatment strategies in patients with complex presentations, such as those with concurrent CRPS and systemic conditions like diabetes, which may complicate healing and recovery processes.

CONCLUSION(S)

This case report provides a thorough overview of the clinical presentation, diagnostic challenges, and therapeutic interventions required for managing advanced Kienböck's disease complicated by CRPS. It offers valuable insights into the multidisciplinary approach needed for optimal patient outcomes. The successful outcome in this case demonstrates the efficacy of wrist arthrodesis in providing long-term pain relief and functional stability in advanced Kienböck's disease, despite the inherent limitations of a fixed wrist position.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Oct 09, 2024
- Manual Googling: Apr 22, 2025
- iThenticate Software: Apr 24, 2025 (12%)

ETYMOLOGY: Author Origin

EMENDATIONS: 6

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Oct 07, 2024**
Date of Peer Review: **Jan 22, 2025**
Date of Acceptance: **Apr 26, 2025**
Date of Publishing: **Sep 01, 2025**