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## Case Report

## Concomitant Bilateral Kienböck and Legg–Calvé–Perthes Diseases

Ahmadreza Afshar, MD, \* Ali Tabrizi, MD, \* Hasan Taleb, MD, \*  
 Mohammad Javad Shariyate, MD \*

\* Department of Orthopedics, Imam Khomeini Hospital, Urmia University of Medical Sciences, Urmia, Iran



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Avascular necrosis involving more than one bone either simultaneously or sequentially is uncommon. We report a rare case of concomitant bilateral Kienböck disease and Legg–Calvé–Perthes disease. No definitive predisposing or precipitating factor could be identified. Because of persistent wrist pain, radial shortening osteotomy was performed sequentially on both wrists. At the latest follow-up, the patient's wrist pain had resolved, and he was satisfied with the clinical outcomes.

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Avascular necrosis (AVN) affecting more than one bone either simultaneously or sequentially is a rare occurrence. Multifocal osteonecrosis is defined as osteonecrosis involving three or more distinct anatomical regions, either concomitantly or consecutively. Various combinations of skeletal involvement, including the femoral head, knee, shoulder, wrist, and ankle, have been reported in the literature.<sup>1–3</sup>

The concomitant occurrence of bilateral Kienböck disease and Legg–Calvé–Perthes disease represents an exceptionally rare combination. To the best of our knowledge, only three case reports have described this association.<sup>4–6</sup> The present report adds another case of concomitant bilateral Kienböck and Legg–Calvé–Perthes diseases to the existing literature.

## Case Report

A 25-year-old man presented with a 1-year history of pain and limited range of motion in his left wrist. Plain radiographs demonstrated Lichtman stage II Kienböck disease with negative ulnar variance. Because of persistent pain, radial shortening osteotomy was offered and performed.

At the 1-year follow-up, the patient developed pain in the right wrist. Radiographs revealed Lichtman stage II Kienböck disease with negative ulnar variance on the right side as well (Fig. 1).

Radial shortening osteotomy was subsequently performed on the right wrist (Fig. 2). At the latest follow-up, pain in both wrists had resolved, and the patient reported satisfaction with the clinical outcomes.

The patient's past medical history revealed bilateral limping since childhood. According to family members, hip pain and limping began at approximately 8 years of age on the left side and subsequently at around 10 years of age on the right side. The left lower limb was approximately 3 cm shorter than the right. Pelvic radiographs demonstrated sequelae of bilateral Legg–Calvé–Perthes disease (Fig. 3). The patient denied any history of trauma to his limbs.

Although involvement of both wrists and hips was bilateral, the disease processes were not at the same stage. Given the multifocal nature of AVN, we investigated potential systemic predisposing factors. Laboratory evaluations, including coagulation profiles, hypercoagulability work-up, rheumatologic studies, and the three key tests for antiphospholipid syndrome, were all within normal limits. The patient continues to be followed to identify any potential underlying etiology and to address future orthopedic needs.

Written informed consent was obtained from the patients for patients' anonymized information to be published in this article.

## Discussion

The pathogenesis of avascular necrosis remains incompletely understood. Numerous predisposing factors have been implicated in the development of atraumatic multifocal AVN, including corticosteroid use, smoking, gout, alcoholism, infection, storage

**Corresponding author:** Ahmadreza Afshar, MD, Urmia University of Medical Sciences, Department of Orthopedics, Imam Khomeini Hospital, Modaress Street, Ershad Boulevard, Urmia, 57157 81351, Iran.

E-mail address: [afshar\\_ah@yahoo.com](mailto:afshar_ah@yahoo.com) (A. Afshar).

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**Figure 1.** Anteroposterior plain radiograph of the right and left wrists demonstrates bilateral Kienböck disease. The left side was treated with radial shortening one year before.



**Figure 2.** Anteroposterior plain radiograph of the right and left wrists demonstrates bilateral Kienböck disease treated with radial shortening osteotomy.

disorders, marrow-infiltrating diseases, malignancy, chemotherapy, coagulation disorders, hemoglobinopathies, fat embolism, liver disease, hyperlipidemia (types II and III), hemophilia, inflammatory arthritis, systemic lupus erythematosus, and pancreatitis.<sup>7–11</sup>

Only three prior case reports have described the rare coexistence of bilateral AVN of the lunate and proximal femur. Rostom et al<sup>5</sup> reported a patient with antiphospholipid syndrome who developed multifocal AVN involving both lunates and proximal femora,



**Figure 3.** An anteroposterior plain radiograph of the pelvis demonstrates sequel of bilateral Legg–Calvé–Perthes disease.

suggesting a possible association between antiphospholipid syndrome and multifocal AVN. Baltzer et al<sup>4</sup> described a child with factor V Leiden thrombophilia complicated by bilateral Legg–Calvé–Perthes disease and Kienböck disease, proposing thrombophilia as a contributing factor. Wollstein et al<sup>6</sup> reported a similar case of concomitant bilateral Kienböck and Legg–Calvé–Perthes diseases without identifying a predisposing factor.

In the present case, no precipitating or predisposing mechanism could be identified. Given the current understanding of AVN pathophysiology, a substantial proportion of cases without identifiable causes are classified as idiopathic.<sup>9,10</sup> Accumulation of additional case reports may improve understanding of the etiology, diagnosis, and management of multifocal AVN.

### Conflicts of Interest

No benefits in any form have been received or will be received related directly to this article.

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### Ethical Approval

The study was approved by the medical research and ethics committee of the Urmia University of Medical Sciences with registered Numbers: IR.UMSU.REC.1404.464

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### Informed Consent

Written informed consent was obtained from the patients for patients' anonymized information to be published in this article.

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