

Medium-term outcome of closed radial wedge osteotomy of the distal radius for Preiser disease with concomitant Kienböck disease

Two case reports and a literature review

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Abstract

Rationale: Idiopathic avascular necrosis of the scaphoid or lunate bone are known as Preiser disease and Kienböck disease, respectively. Although there are reports of avascular necrosis involving more than one carpal bone, concurrent idiopathic avascular necrosis of the scaphoid and lunate bones is rare, with only five cases reported in the English literature (including the two herein). Although the optimum treatment for Preiser disease with concomitant Kienböck disease has not been established, our cases underwent closed radial wedge osteotomy based on the evidence of satisfactory outcomes for treating Kienböck disease. We report the medium-term results of closed radial wedge osteotomy of the distal radius in two cases of Preiser disease with concomitant Kienböck disease.

Patient concerns: We presented two patients with concomitant Preiser and Kienböck diseases. Although both smoked cigarettes, neither had any other risk factors; there was no history of trauma, although both women had jobs that required relatively heavy or repetitive manual labor.

Diagnoses: Two patients were diagnosed by radiographs and magnetic resonance imaging of the wrists.

Interventions: A non-surgical strategy of splint immobilization and analgesia was not effective, and surgery was ultimately required.

Outcomes: Satisfactory medium-term results were achieved with closed radial wedge osteotomy of the distal radius in both cases. Although there was imaging evidence of progression of dorsal intercalated segmental instability deformity, neither of the patients was symptomatic and both declined salvage surgery.

Lessons: We compare our cases and treatment strategy with others reported in the literature. Our cases suggest that closed radial wedge osteotomy of the distal radius is a safe and relatively straightforward means of treating patients with this rare combination of wrist disorders, and appears to obviate the need for more extensive salvage procedures.

Abbreviations: AVN = avascular necrosis, CRWO = closed radial wedge osteotomy, DISI = the dorsal intercalated segmental instability, MMWS = The Modified Mayo Wrist Score, MRI = magnetic resonance imaging, RLA = the radio-lunate angle, SLA = The scapho-lunate angle.

Keywords: avascular necrosis, case report, closed radial wedge osteotomy, Kienböck disease, lunate, Preiser disease, scaphoid

1. Introduction

Idiopathic avascular necrosis (AVN) of the scaphoid bone is known as Preiser disease, and idiopathic AVN of the lunate bone is known as Kienböck's disease. Both are rare. Although there have been reports of AVN affecting more than 1 carpal bone,^[1]

concurrent AVN of the scaphoid and lunate bones is very rare indeed.^[2–5] Other cases of concurrent disease were reported in 2009,^[5] and we report the medium-term results of closed radial wedge osteotomy of the distal radius (CRWO)^[6] in 2 cases of Preiser disease with the concomitant Kienböck disease.

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2. Case reports

2.1. Case 1

A 50-year-old woman who worked as a post office clerk presented with spontaneous pain in her dominant right wrist. Swelling and tenderness were identified dorsal to Lister's tubercle and the anatomical snuff-box. Although she smoked cigarettes, she had no history of alcohol misuse or steroid use. There was no history of trauma, and no other joints were involved. Seven years previously, she had had surgery to the right breast for cancer; however, the possibility of metastasis was excluded on plain radiograph and magnetic resonance imaging (MRI).

On examination, pain was worse on movement, particularly with wrist extension, and function was restricted by pain. Extension of the right wrist was 35°, and flexion was 60°. Grip strength was 17 kg on the affected side and 21 kg on the

Table 1		
Modified Mayo Wrist Score^[6].		
Category	Score	Findings
Pain (25 points)	25	No pain
	20	Mild pain with vigorous activities Pain only with weather changes
	15	Moderate pain with vigorous activities
	10	Mild pain with activities of daily living
	0	Pain at rest
Satisfaction (25 points)	25	Very satisfied
	20	Moderately satisfied
	10	Not satisfied but working
	0	Not satisfied, unable to work
Range of motion (25 points) (% of normal)	25	100
	15	75–99
	10	50–74
	5	25–49
	0	0–24
Grip strength (25 points) (% of normal)	25	100
	15	75–99
	10	50–74
	5	25–49
	0	0–24
Final results (points)	Excellent 100, Good 80–89, Fair 65–79, Poor <65	

unaffected side. The Modified Mayo Wrist Score (MMWS, Table 1) was 0-0-10-15 (25 points).^[6]

Radiographs of the right wrist showed collapse of the proximal pole of the scaphoid bone without fragmentation, and sclerotic changes in the lunate bone. There was negative ulnar variance (Fig. 1A).

The scapho-lunate angle (SLA) was 53° and the radio-lunate angle (RLA) was 40° on the lateral view (Fig. 1B).

Magnetic resonance imaging of the right wrist showed diffuse reduction in signal intensity throughout the scaphoid and lunate bones on T1-weighted sequences, compatible with AVN (Fig. 2A); T2-weighted fast spin echo sequences revealed that the scaphoid was markedly flattened, with extensive collapse of the subchondral trabecular bone and high signal intensity compatible with bone edema (Fig. 2B). Taken together, the radiographic and MRI findings were diagnostic of Preiser disease, classified as Herbert and Lanzetta^[7] stage 2, Kalainov et al^[8] type 1, with concomitant Kienböck disease, classified as Lichtman et al^[9] stage 2 (Tables 2 and 3).

A nonsurgical strategy of splint immobilization and analgesia was not effective, and surgery was ultimately required. Informed consent was obtained from the patient for surgery, and for publication of the case. The operation was performed under general anesthesia and a pneumatic tourniquet was applied. The CRWO technique was performed as first described by Kojima et al.^[10] Briefly, a palmar incision was made over the radius using the Henry approach, and a 15° radial closed wedge osteotomy was performed 3 cm proximal to the tip of the radial styloid process, which was fixed with a volar plate in the distal radius. Postoperatively, a short-arm cast was applied for 6 weeks (Fig. 3A and B). Removal of the radial plate was performed after 12 months to reduce the risk of tendon attrition. At the 12-month postoperative follow-up evaluation, MRI revealed that the signal intensity of the distal pole of the scaphoid and the entire lunate matched the intensity of the remaining carpal bones on T1-weighted sequences (Fig. 4A). On T2-weighted sequences, although the proximal pole of the scaphoid remained flattened, the high intensity signal in the scaphoid was markedly decreased, indicating normalization of the blood supply of the scaphoid (Fig. 4B).



Figure 1. Preoperative radiographs in Case 1. (A) Preoperative posteroanterior radiograph showing collapse of the proximal pole of the scaphoid bone without fragmentation, and sclerotic changes in the lunate bone. (B) Preoperative lateral radiograph showing a scapho-lunate angle (SLA) and radio-lunate angle (RLA) of 53° and 40° degrees, respectively.

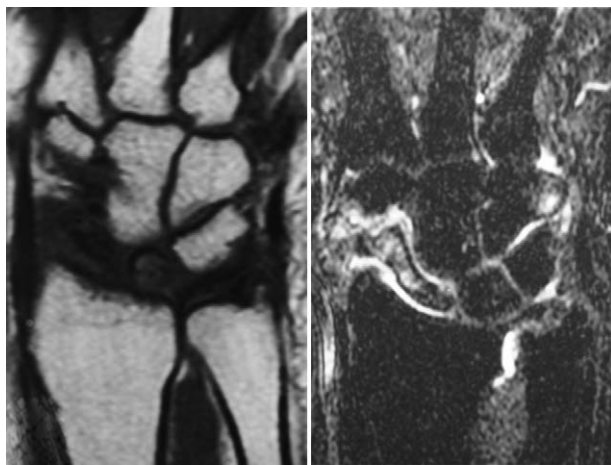


Figure 2. Preoperative radiograph and magnetic resonance images in Case 1. (A) Preoperative posteroanterior T1-weighted magnetic resonance images (MRI) of the right wrist showing diffuse decreased signal intensity throughout both the scaphoid and lunate bones, compatible with Kalainov type 1 Preiser disease and Lichtman stage 2 Kienböck disease. (B) Preoperative posteroanterior T2-weighted fast spin echo MRI showing a markedly flattened scaphoid with extensive collapse of the subchondral trabecular bone and high bone intensity, compatible with bone edema.

The patient reported no residual pain at a further follow-up evaluation after 28 months. The right wrist was nontender, and had 40° of extension, 60° of flexion, and full pronation and supination. Her grip strength was 19 kg, 90.5% of that of the left wrist (21 kg), and the MMWS score was 25-25-10-15 (75 points). Radiographs of the right wrist showed radio-scaphoid arthritis with flattening of the proximal scaphoid, but there was no evidence of exacerbation of sclerotic change in the lunate bone (Fig. 5A). The SLA and RLA were 70° and 29°, respectively (Fig. 5B). The patient was satisfied with the surgical results and had no desire to pursue salvage procedures, despite progression of the dorsal intercalated segmental instability (DISI) deformity (Table 4).

2.2. Case 2

A 59-year-old woman who worked as a health care assistant presented to our hospital with moderate pain in the dominant right wrist provoked by activities of daily living. On physical examination, there was swelling and tenderness dorsal to Lister’s tubercle and the anatomical snuff-box. Although she smoked cigarettes, she had no history of alcohol misuse, or steroid use.

Table 2
Staging classification of Preiser and Kienböck diseases according to radiographs and magnetic resonance imaging^[7-9].

Herbert and Lanzetta grading scale ^[7] for Preiser disease	
Stage 1	Normal radiograph MRI signal change abnormal sign on bone scan
Stage 2	Increased density of the proximal pole scaphoid
Stage 3	Fragmentation of the proximal pole scaphoid without pathological fracture
Stage 4	Carpal collapse with osteoarthritis
Lichtman grading scale ^[9] for Kienböck disease	
Stage 1	Normal radiograph MRI signal change abnormal sign on bone scan
Stage 2	Lunate sclerosis without collapse
Stage 3A	Lunate fragmentation and collapse without scaphoid rotation
3B	Lunate fragmentation and collapse with fixed scaphoid rotation
Stage 4	Perilunate arthritic change
Kalainov criteria ^[8] for Preiser disease (magnetic resonance imaging)	
Type 1	Complete necrosis
Type 2	Incomplete necrosis

There was no history of trauma, and no other joints were involved.

The patient had intolerable pain in the right wrist during wrist flexion and extension, severely restricting function. The patient’s right wrist had 37° of extension and 34° of flexion. Her grip strength was 8 kg on the affected side and 25 kg on the unaffected side. The MMWS score was 5-10-10-5 (30 points, Table 1).

Radiographs of the right wrist showed marked collapse of the whole scaphoid bone without fragmentation, and osteosclerotic change of the lunate bone (Fig. 6A). Assessment of the lateral radiograph revealed an SLA and RLA of 74° and 7°, respectively, which was compatible with DISI deformity (Fig. 6B).

The T1-weighted MRI sequence identified segmental low intensity of the right scaphoid and the whole right lunate, compatible with AVN (Fig. 7A); T2-star-weighted sequences revealed flattening of the proximal scaphoid with extensive collapse of the subchondral trabecular bone and high intensity at the distal pole, compatible with bone edema. T2-weighted sequences did not show abnormal intensity in the right lunate (Fig. 7B). Radiologic and MRI findings were diagnostic of Preiser disease, classified as Herbert and Lanzetta stage 3, Kalainov type 2, with concomitant Kienböck disease, Lichtman stage 2 (Tables 2 and 3).

Table 3
Presurgical status of 2 patients with concomitant Preiser and Kienböck diseases.

Case	Age (y) /sex	Affected wrist (dominance)	Work (duration)	Duration of pain, months	Pain	Presurgical stage			Presurgical radiographic measurement			Presurgical wrist function (affected/unaffected)						
						Herbert class	Kalainov class	Lichtman class	DISI	SLA, (°)	RLA (°)	Function	Ext	Flex	Pr	Sp	GP, kg	MMWS, points
1	50/F	Right	Post office clerk	11	Pain at rest	2	1	2	No	53	40	Unable to work	35/72	60/80	80/85	80/85	17/21	25
2	59/F	Right	Health care assistant (3 y)	9	Moderate pain with ADL	3	2	2	Yes	74	7	Working but severely restricted	37/64	34/62	90/90	90/90	8/25	30

ADL = activity of daily living, DISI = dorsal intercalated segmental instability, Ext = extension, F = female, Flex = flexion, GP = grip strength, Herbert class = Herbert and Lanzetta classification, Kalainov class = Kalainov classification, Lichtman class = Lichtman classification, MMWS = Modified Mayo Wrist Score, Pr = pronation, RLA = radio-lunate angle, SLA = scapho-lunate angle, Sp = supination.



Figure 3. Postoperative radiographs in Case 1. (A) Postoperative posteroanterior radiograph. Closed radial wedge osteotomy of the distal radius was performed with a volar flexed-angle locking plate. (B) Postoperative lateral radiograph showing a scapho-lunate angle (SLA) of 74° and dorsal intercalated segmental instability (DIS) deformity.

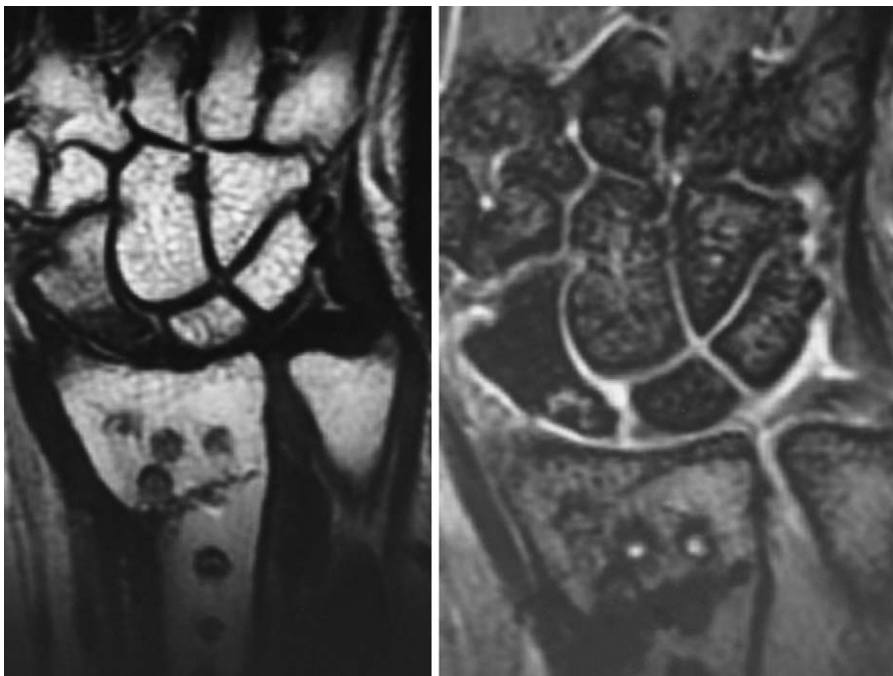


Figure 4. Postoperative magnetic resonance images in Case 1. (A) Postoperative T1-weighted magnetic resonance images (MRI) showing that the signal intensity of the distal pole of the scaphoid and the entire lunate had recovered to the same intensity as the other carpal bones. (B) Postoperative T2-star-weighted MRI showing decrease in the previous high intensity of the scaphoid despite residual flattening of the proximal pole.



Figure 5. 28-month follow-up radiographs in Case 1. (A) 28-month follow-up posteroanterior radiographs of the right wrist showing radio-scaphoid arthritis with flattening of the proximal scaphoid. (B) 28-month follow-up lateral radiograph showing a scapho-lunate angle (SLA) and radio-lunate angle (RLA) of 70° and 29°, respectively, and progression of the dorsal intercalated segmental instability (DISI) deformity.

Several weeks of rest, immobilization with a wrist splint and orthosis, and analgesia were not effective, and CRWO was required to treat pain and improve wrist motion (Fig. 8A and B). Informed consent was obtained from the patient for surgery and for publication of the case. The radial plate was removed after 12 months. The patient reported no residual pain at a follow-up evaluation 73 months after surgery. On examination, the right wrist was nontender, extension was 40°, flexion was 50°, and there was full pronation and supination (Fig. 9). Her grip strength was 23 kg on both sides, and MMWS score was 25-25-10-25 (85 points). Radiographs of the right wrist showed radioscaphoid arthritis, carpal collapse, scaphoid fragmentation, and lunate sclerosis (Fig. 10A). Assessment of the lateral radiograph revealed an SLA and RLA of 85° and 30°, respectively, compatible with DISI deformity (Fig. 10B). The patient was satisfied with the surgical results and had no desire to pursue a salvage procedure,

despite progression of the DISI deformity and osteoarthritis of the radiocarpal joint (Table 4).

3. Discussion

To the best of our knowledge, there have been only 5 cases of Preiser disease with concomitant Kienböck disease reported in the English literature, including these 2 cases (Table 5).^[2-5] Although cases of AVN involving more than 1 carpal bone have been reported, almost all have been attributed to steroid therapy for systemic illness, such as autoimmune hemolytic anemia,^[1] systemic lupus erythematosus, and renal transplantation, or have involved risk factors, such as smoking, alcohol misuse or infection.^[2] The case reported by Budoff^[2] had a 3-month history of steroid use and was a cigarette smoker; another case reported by Bhardwaj and colleagues^[4] had been taking herbal medication

Table 4
Postsurgical outcome of 2 patients treated for concomitant Preiser and Kienböck diseases.

Case	Follow-up period, months	Pain	Postsurgical stage			Postsurgical radiographic measurement			Postsurgical wrist function (affected/unaffected)					MMWS, points	
			Herbert class	Kalainov class	Lichtman class	DISI	SLA, degree	RLA, degree	Function	Ext	Flex	Pr	Sp		GP, kg
1	28	No	4	2	2	Yes	70	29	Very satisfied	40/70	60/70	85/85	80/80	19/21	75
2	73	No	4	-	2	Yes	85	30	Working without restriction	40/65	50/70	85/85	80/80	23/23	85

DISI = dorsal intercalated segmental instability, Ext = extension, F = female, Flex = flexion, GP = grip strength, Herbert class = Herbert and Lanzetta classification, Kalainov class = Kalainov classification, Lichtman class = Lichtman classification, MMWS = Modified Mayo Wrist Score, Pr = pronation, RLA = radio-lunate angle, SLA = scapho-lunate angle, Sp = supination; -, data not available.



Figure 6. Preoperative radiographs in Case 2. (A) Preoperative posteroanterior radiograph showing marked collapse of the proximal pole of the scaphoid bone without fragmentation, and osteosclerotic change in the lunate bone. (B) Preoperative lateral radiograph showing a scapho-lunate angle (SLA) and radio-lunate angle (RLA) of 74° and 7° , respectively, which were compatible with dorsal intercalated segmental instability (DISI) deformity.

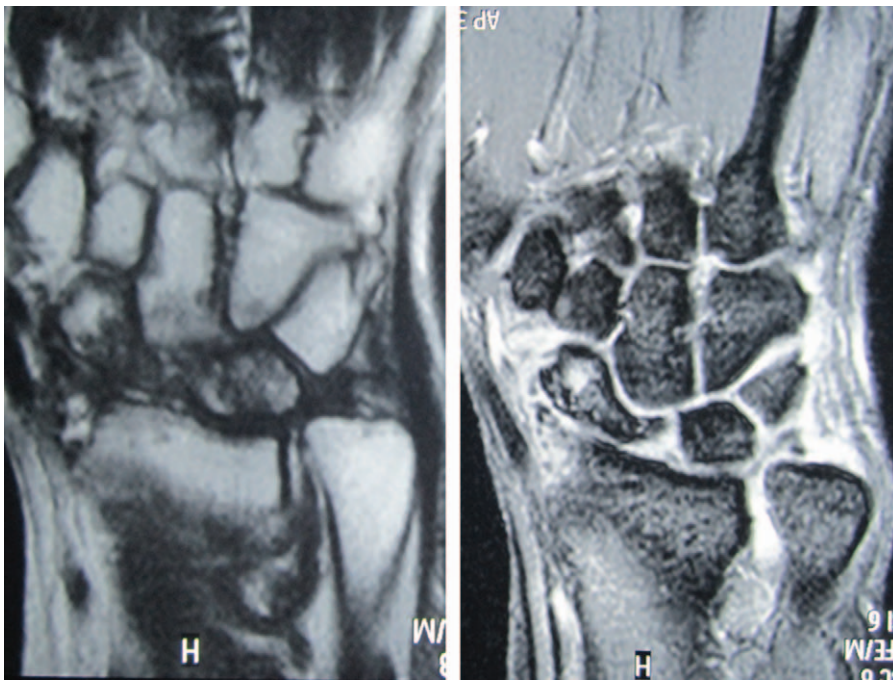


Figure 7. Preoperative magnetic resonance images in Case 2. (A) Preoperative posteroanterior T1-weighted magnetic resonance imaging (MRI) showing segmental low intensity of the proximal pole of the scaphoid and the entire lunate, compatible with AVN on T1-weighted sequences. (B) Preoperative posteroanterior T2-weighted MRI showing flattening of the proximal scaphoid and extensive collapse of the subchondral trabecular bone, and high signal intensity in the distal pole of the scaphoid, compatible with bone edema. The MRI findings were diagnostic of Preiser disease, Kalainov type 2, with concomitant Kienböck disease, Lichtman stage 2.

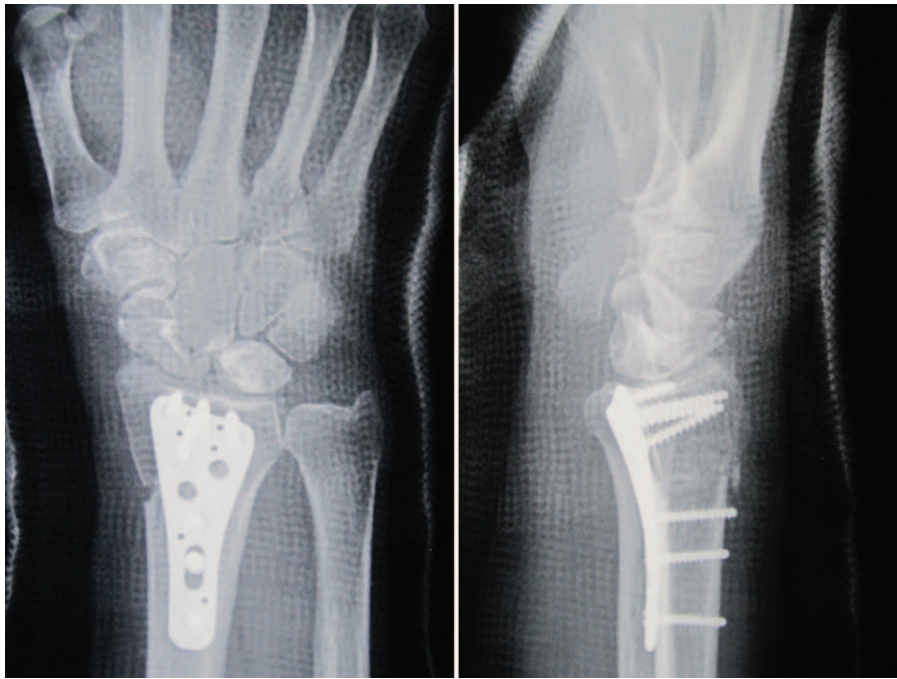


Figure 8. Postoperative radiographs in Case 2. (A) Postoperative posteroanterior radiograph. Closed radial wedge osteotomy of the distal radius was performed with a volar flexed-angle locking plate. (B) Postoperative lateral radiograph showing a scapho-lunate angle (SLA) of 89° and dorsal intercalated segmental instability (DIS) deformity.

that may have contained a steroid (Table 5). Both our cases were smokers. Only 1 of the 5 patients had no history of steroid use or smoking, and therefore is the only case in which the cause can be considered idiopathic. Although the etiology of idiopathic AVN of the scaphoid and lunate is incompletely understood, undeveloped vascular networks and mechanical stress, such as repetitive strain and overloading, could be predisposing factors.^[7–10] In our cases, since the patients both smoked and were engaged in relatively heavy or repetitive manual work, congenital undeveloped vascular networks and mechanical stress

combined with vascular contraction due to smoking were the most likely precipitating factors.

Although the optimum treatment for Preiser disease with concomitant Kienböck disease has not been established, our cases underwent CRWO based on the evidence of satisfactory outcomes for treating Kienböck disease.^[10–12] Generally, surgical treatment for early stage Kienböck disease involves a joint-leveling operation or vascularized bone grafting. Joint-leveling procedures refer to radial shortening osteotomy of those patients Kienböck disease who have negative ulnar variance, and closing



Figure 9. Follow-up evaluation in Case 2. At the 73-month follow-up evaluation, the patient had no tenderness or residual pain. The right wrist had 40° of extension, 50° of flexion, and full pronation and supination.



Figure 10. 73-month follow-up radiographs in Case 2. (A) 73-month follow-up posteroanterior radiographs of the right wrist showing radio-scaphoid arthritis with flattening of the proximal scaphoid and lunate sclerosis. (B) 73-month follow-up lateral radiograph showing a scapho-lunate angle (SLA) and radio-lunate angle (RLA) of 85° and 30°, respectively, and the persistent dorsal intercalated segmental instability (DISI) deformity.

or opening wedge osteotomy^[10–13] of those patients with neutral variance. Salvage procedures, such as proximal row carpectomy, partial wrist arthrodesis or total wrist arthrodesis, are indicated for advanced-stage Kienböck disease. We elected to undertake CRWO as a joint-leveling operation in both our cases as both were early-stage AVN of the lunate, Lichtman stage 2, and, despite their early stage, outcomes were satisfactory in both cases.

The CRWO seeks to address 2 issues in AVN: decompression of the radio-lunate and radio-scaphoid joints by enlarging the

spaces between the radius and the lunate or scaphoid bone,^[5,10–13] and inducing revascularization of the carpals following osteotomy.^[11,12] Additionally or alternatively, CRWO may decompress venous hypertension in a similar way to osteotomy in AVN of the femoral head;^[11] normalization of intraosseous pressure may result in the reduction of pain and consequently improve range of motion. The underlying mechanisms that afford the therapeutic benefits of CRWO are, however, still a matter of some debate.

Table 5

Comparison of all 5 known published reports of concomitant Preiser and Kienböck diseases.

Authors	Age (y) /sex	Affected wrist (dominance)	Medical history	Risk factors	Herbert class	Kalainov class	Lichtman class	Tx	Follow-up period, months	Residual pain	ROM, degrees Ext/Flex/ Pr/Sp	GP, kg Affected/ Unaffected	MMWS
Budoff ^[2]	50/F	Right	None	Steroid use* Smoking	3	1	3B	PRC	40	50% reduction [‡]	65/40/ 60/full	—/—	—
Park et al ^[3]	56/F	Right	None	No	1	2	3B	PWA	12	No	—/—/ —/—	—/—	—
Bhardwaj et al ^[4]	20/M	Right	None	Suspected steroid use [†]	3	1	1	PRC	12	No	45/25/ —/—	20/30	70
Our cases	50/F	Right	Breast cancer	Smoking	2	1	2	CRWO	28	No	40/60/ 80/85	19/21	75
	59/F	Right	None	Smoking	3	2	2	CRWO	73	No	40/50/ 80/85	23/23	85

CRWO = closed radial wedge osteotomy, F = female, GP = grip strength (affected/unaffected), Herbert class = Herbert and Lanzetta classification, Kalainov class = Kalainov classification, Lichtman class = Lichtman classification, M = male, MMWS = modified Mayo wrist score, PRC = proximal row carpectomy, PWA = partial wrist arthrodesis, ROM = range of motion (Ext, extension; Flex, flexion; Pr, pronation; Sp, supination), Tx = treatment; —, data not available.

* Taking steroids for 2 months.

† Taking herbal medication for 5 years.

‡ Compared with preoperative status.

The revascularization effects of CRWO in Kienböck disease have been reported by Nakamura and colleagues,^[12] who observed revascularization of the lunate on MRI. Although postoperative MRI was not obtained in our second case, postoperative follow-up MRI in Case 1 showed improvement of the intensity of the scaphoid and lunate on T1- and T2-weighted sequences, suggesting that CRWO may have a revascularization effect on these carpal bones.

Based on the outcomes in our cases, CRWO for Preiser disease with concomitant early stage Kienböck disease is a plausible temporizing option *in lieu* of a reconstructive procedure; however, the outcome may depend on the stage of Kienböck disease. Soejima and colleagues^[13] have reported that CRWO was effective even in advanced Kienböck disease, suggesting that CRWO should be effective when Preiser disease and advanced Kienböck disease coexist.

In our cases, CRWO effectively treated pain and restricted range of wrist motion. Although at follow-up we identified radiographic evidence of progressive deterioration of the affected wrists, radiographic findings do not necessarily correlate with symptoms, and AVN can be asymptomatic.^[11] In the medium-term, both our patients were satisfied with the surgical results, despite progression of the DISI deformity and osteoarthritis of the wrists. We cannot rule out that degenerative arthritis may, however, impair their wrist function in the longer term.

The main limitation of our study is that we only report the outcomes of 2 patients, but this reflects the rarity of the disease. Although we cannot compare CRWO with other operative or nonoperative treatments, our case reports offer valuable insight into the treatment of Preiser disease with concomitant Kienböck disease.

4. Conclusion

We found that medium-term outcomes after CRWO of the distal radius in 2 cases of Preiser disease with concomitant Kienböck

disease were satisfactory. Both patients were satisfied with the surgical results, had no residual pain and no desire to pursue a salvage procedure, despite progression of the DISI deformity and osteoarthritis of the wrist. Although we cannot explain why CRWO successfully treated pain and reduced range of motion, CRWO appears to be a safe and relatively straightforward surgical strategy for Preiser disease with concomitant Kienböck disease.

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