Clinical Case Report

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Preiser disease after repeated local glucocorticoid injections

A case report

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Abstract

Rationale: Preiser disease or avascular necrosis (AVN) of the scaphoid causes intolerable wrist pain and malalignment of the carpal bones. In previously reported cases, patients have had a history of steroid use for systemic illness such as autoimmune hemolytic anemia, systemic lupus erythematosus, or renal transplantation, or have had other risk factors, such as smoking, alcoholism, or infection. In particular, systemic glucocorticoid therapy has been most commonly associated with the disease. Although there are reports of AVN of the scaphoid induced by systemic glucocorticoids, no prior report has associated AVN of the carpal bones with repeated local injections of glucocorticoids.

Patient concerns: We present a case in which it was strongly suspected that AVN of the scaphoid was induced by repeated local glucocorticoid injections. The patient had no history of excessive alcohol use, smoking, or trauma, except for local repeated steroid injections.

Diagnoses: Initially, she had diagnosed with de Quervain's disease and was treated by repeated local glucocorticoid injections followed by surgery for de Quervain's disease. Five years after surgery for de Quervain's disease, the patient presented at our hospital with sudden onset of intolerable pain in her right wrist without a history of trauma. In spite of nonsurgical treatment with rest, immobilization, analgesia, and surgery, her wrist pain was not improved. After further repeated local steroid injections in her wrist, radiographs, and magnetic resonance imaging of her wrist showed the AVN of the scaphoid.

Interventions: Surgery was performed and the fragmented proximal scaphoid and the entire lunate were resected.

Outcomes: The diagnosis was confirmed according to the histopathological examination of the proximal scaphoid bone, which showed the characteristic of AVN of the scaphoid. At follow-up evaluation, radiographs of the right wrist showed no progression of osteoarthritis. The patient had no tenderness or residual pain at the wrist and had no desire to pursue additional surgery.

Lessons: We have presented a case with AVN of the scaphoid, which was strongly suspected to be associated with the repeated local steroid injections. Further studies are required to more fully elucidate the association between AVN of the scaphoid and repeated local steroid injections.

Abbreviations: AVN = avascular necrosis, GC = glucocorticoid, MMWS = The Modified Mayo Wrist Score, MRI = magnetic resonance imaging.

Keywords: aseptic avascular necrosis, glucocorticoid, Preiser disease, scaphoid, steroid, wrist

1. Introduction

Preiser disease or idiopathic avascular necrosis (AVN) of the scaphoid,[1,2] is an uncommon disorder. Previously reported cases have involved patients who received steroids for systemic illness such as autoimmune hemolytic anemia,[3] systemic lupus erythematosus,[4] or renal transplantation,[5] and those with other risk factors, such as smoking, alcoholism, or infection.[6,7] Although the etiology of idiopathic AVN of the carpal bones, including the lunate and scaphoid bones, remains unknown, undeveloped vascular networks and mechanical stress are predisposing factors.[1,2,6,8–10,11] Repetitive stress and overloading induce joint swelling and interruption of intra- and extra-osseous blood supply. In patients with undeveloped vascular networks, pathological changes can cause AVN. Glucocorticoid (GC) use is also a pivotal risk factor in the development of AVN. Although the pathogenesis of GC-induced osteonecrosis is not completely elucidated, multiple actions of GCs could cause reduced blood flow in the osseous microcirculation, leading to osteonecrosis.[9–12] To date, systemic

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Patient Concern: Informed written consent was obtained from the patient for publication of this case report and accompanying images

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GC use has been reported as the most common cause of AVN of the carpal bones.[2,3,13–17] In experimental animal studies, repeated local GC injections have been used to establish a model of AVN of the femoral head.[18–22] However, no prior clinical report has associated AVN of the femoral head or carpal bones with repeated local injection of GC. We herein report a case in which AVN of the scaphoid was strongly suspected of being induced by repeated local injection of GC.

2. Case report

A 55-year-old, right-handed woman employed as a sanitation worker presented with gradual onset of pain in the right wrist over the previous month. Her pain began after hard work at her job. She initially felt mild pain in the right wrist, but noticed a gradual increase in wrist and thumb pain. She visited her family physician who diagnosed de Quervain's disease and recommended medications and a local steroid injection in the wrist. After 3 injections of betamethasone phosphate (Rinderon), 2 mg, with 0.5 mL 1% xylocaine within 1 month, the patient presented to our hospital. She had swelling and tenderness at the radial styloid, with normal range of motion. She had no history of excessive alcohol use or steroid use for systemic illness such as systemic lupus erythematosus or organ transplantation. There was no history of trauma, and no other joints were involved. Radiographs of the patient's right wrist were within normal limits. Provocative joint test (Finkelstein's test) was positive for pain and she was again diagnosed with de Quervain's disease. Although the patient had continued to take medications for 8 months, she described the pain as a constant ache with use of her right hand. A thumb spica orthosis was applied for 2 months for pain management, but the patient's pain was ongoing and limited her activities of daily living and work. Two local injections of triamcinolone acetonide (Kenacort), 10 mg, with 0.5 mL 1% xylocaine) within 1 month decreased the patient's pain for 2 months; however, pain recurred at 2 months. Surgery for de Quervain's disease was performed. On perioperative inspection, the tendon sheath of the first compartment was markedly swollen and thickened, consistent with de Quervain's disease. The patient's wrist pain improved after surgery, but recurred 5 months after surgery. The patient took medication and had intermittent local injections of GC in a 3-year period; 2 injections of triamcinolone acetonide (10 mg), and 2 injections of betamethasone phosphate (2 mg).

Five years after surgery for de Quervain's disease, the patient presented at our hospital with sudden onset of intolerable pain in her right wrist without a history of trauma. She had residual pain in her right wrist dorsal to Lister's tubercle and the anatomical snuff box. The pain increased with motion, especially with wrist extension, and severely restricted her function. The patient's right wrist had 22° of extension and 41° of flexion (35.5% and 66.1%, respectively, of values for the left wrist). Radial and ulnar deviations were 0 and 26°, respectively. Forearm supination and pronation were within normal limits. The patient's power grip was 16.5 kg, which was 77.1% of the value for the left wrist. The patient's modified Mayo wrist score (Table 1) was 25 points (0-0-10-15).

Radiographs of the right wrist showed collapse of the proximal pole of the scaphoid bone without fragmentation (Fig. 1A). On the lateral view, the scapholunate angle was 55° and the radiolunate angle was 20° (Fig. 1B).

Magnetic resonance imaging (MRI) of the right wrist showed diffusely decreased signal intensity throughout the scaphoid on T1-weighted sequences, compatible with AVN (Fig. 2A). T2-weighted fast spin-echo sequences revealed marked flattening of the scaphoid with extensive collapse of the subchondral trabecular bone and high intensity of the scaphoid, compatible with bone edema (Fig. 2B). Radiological evaluation and MRI indicated Preiser disease, classified as Herbert and Lanzetta[2] stage 3 and Kalainov et al[24] type 1 (Table 2).

Table 1

Modified Mayo wrist score[23].

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
Result (points)	Excellent 100, Good 80–89, Fair 65–79, Poor <65	

The patient wished to decrease her severe pain and retain some motion in the wrist. Informed consent was obtained from the patient for surgery and for publication of this case. Surgery was performed under general anesthesia with application of a pneumatic tourniquet. A straight dorsal incision was made on the wrist; the extensor retinaculum was exposed with blunt dissection. A retractor was placed between the extensor pollicis longus and radial wrist extensor tendons radially and the common extensor tendons ulnarly. The dorsal wrist capsule was exposed and reflected away from the distal radius in an inverted T fashion. The condition of the articular surface was visually inspected. The proximal scaphoid was collapsed and fragmented. The radioscaphoid and radiolunate articular surfaces showed significant degeneration. The fragmented proximal scaphoid and the entire lunate were resected (Fig. 3A and B).

3. Histopathology

Histopathological examination of the proximal scaphoid bone showed an area of necrosis involving the entire subchondral zone, with necrotic debris in the intertrabecular space and empty lacunae without any viable cells (Fig. 4A and B). There was a multilayered tide mark in the boundary between the articular cartilage and the subchondral zone, and the cartilaginous tissue

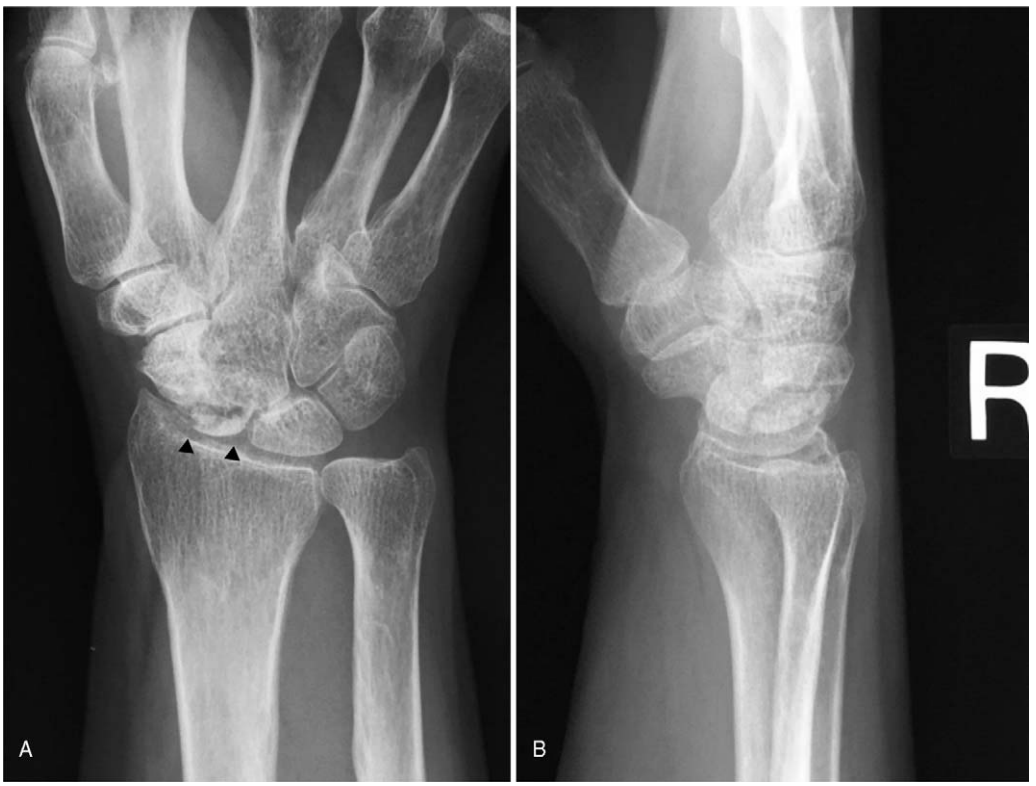

Figure 1. (A) Preoperative posteroanterior radiograph shows collapse of proximal pole of scaphoid bone without fragmentation (black arrow heads). (B) Preoperative lateral radiograph shows a scapholunate angle of 55° and a radiolunate angle of 20°.

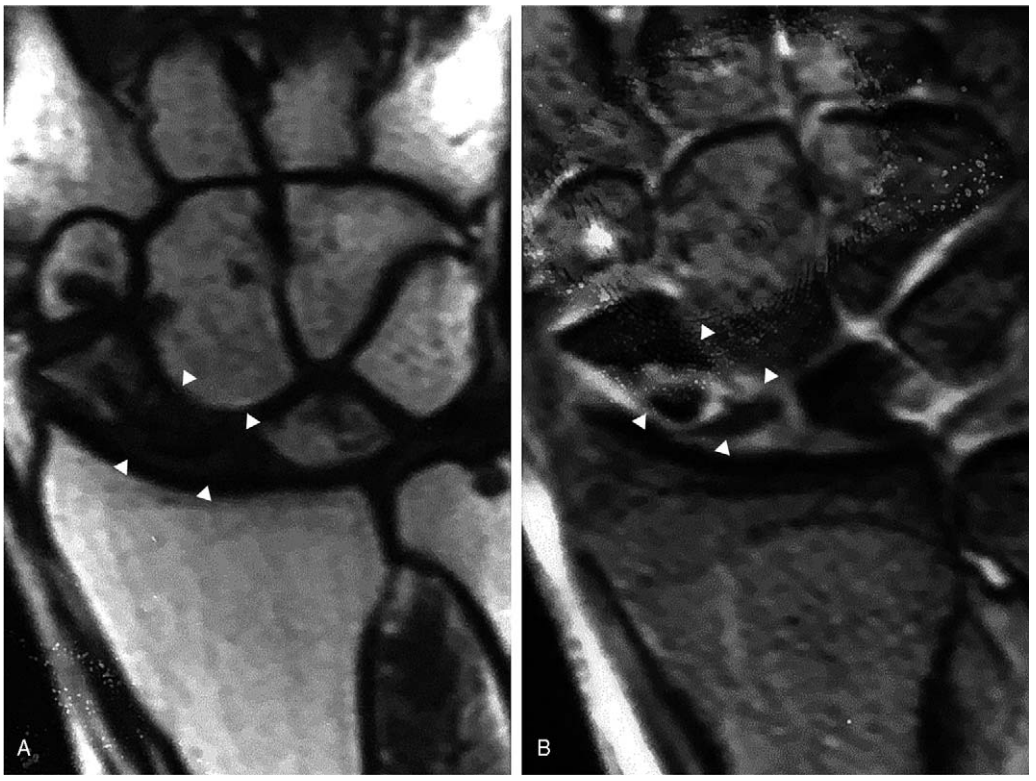

Figure 2. (A) Preoperative posteroanterior T1-weighted MRI shows diffusely decreased signal intensity throughout the scaphoid and lunate bones (white arrow heads), compatible with Kalainov Type 1 Preiser disease. (B) Preoperative posteroanterior T2-weighted fast spin-echo MRI shows marked flattening of the scaphoid with extensive collapse of the subchondral trabecular bone and high intensity of the scaphoid (white arrow heads), compatible with bone edema.

Table 2

Classification for Preiser disease according to Herbert and Lanzetta grading scale and Kalainov criteria.

Herbert and Lanzetta grading scale[2] 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
Kalainov criteria[24] for Preiser disease (magnetic resonance imaging)	
Type 1	Complete necrosis
Type 2	Incomplete necrosis

showed chondrocytic cloning, collectively suggesting a regenerative process following collapse of the proximal scaphoid (Fig. 4C and D). All these findings are characteristic of AVN of the scaphoid bone

At follow-up evaluation 4 months after her first operation for AVN of the scaphoid, the patient's range of motion was restricted because of impingement of the distal scaphoid by the radial styloid. Additional surgery was performed to resect part of the distal scaphoid and part of the radial styloid.

At follow-up evaluation 1 year, 4 months after her first surgery for AVN of the scaphoid, the patient had no tenderness or residual pain. The right wrist had 25° of extension and 26° of flexion (41.9% and 66.1%, respectively, of the values for the left wrist). Forearm pronation and supination were the same as on the contralateral side. The patient's power grip was 20.4 kg, which was 98.6% of the value for the left wrist. Her modified Mayo wrist score was 60 points (25-10-10-15). Radiographs of the right wrist showed no progression of osteoarthritis (Fig. 5A and B).

Although there was a long interruption between visits to the hospital, the patient had no tenderness or residual pain at her last follow-up evaluation, 7 years, 1 month after her initial operation for AVN of the scaphoid. The right wrist had 41° of extension and 46° of flexion (66.1% and 74.2%, respectively, of values for the left wrist). Pronation and supination were the same as for the left wrist. The patient's power grip was 16.6 kg, which was 70.3% of the value for the left wrist. The patient's modified Mayo wrist score was 70 points (25-25-10-10). Radiographs of the right wrist showed no progression of osteoarthritis (Fig. 6A and B). The patient was satisfied with the results and had no desire to pursue additional surgery.

4. Discussion

The etiology of GC-induced osteonecrosis has been investigated in animal models of AVN of the femoral head.[18–20,22,25] Studies have shown that GCs can not only induce apoptosis of osteoblasts and osteocytes,[9–12] but can also increase the number of bone marrow adipocytes and adipocyte hypertrophy.[21,26] In addition, GCs may have direct effects on endothelial cells by modulating their responses to vasoactive substances and potentiating local hypertension and vasoconstriction.[12,19] Thus, GCs can reduce the blood supply to the bone marrow and cause lipid embolism due to hyperlipidemia.[27] GCs influence multiple physiological pathways that decrease osseous blood flow and increase intraosseous pressure, potentiating osteonecrosis.[9,10,12] Although a strong association has been reported between GCs

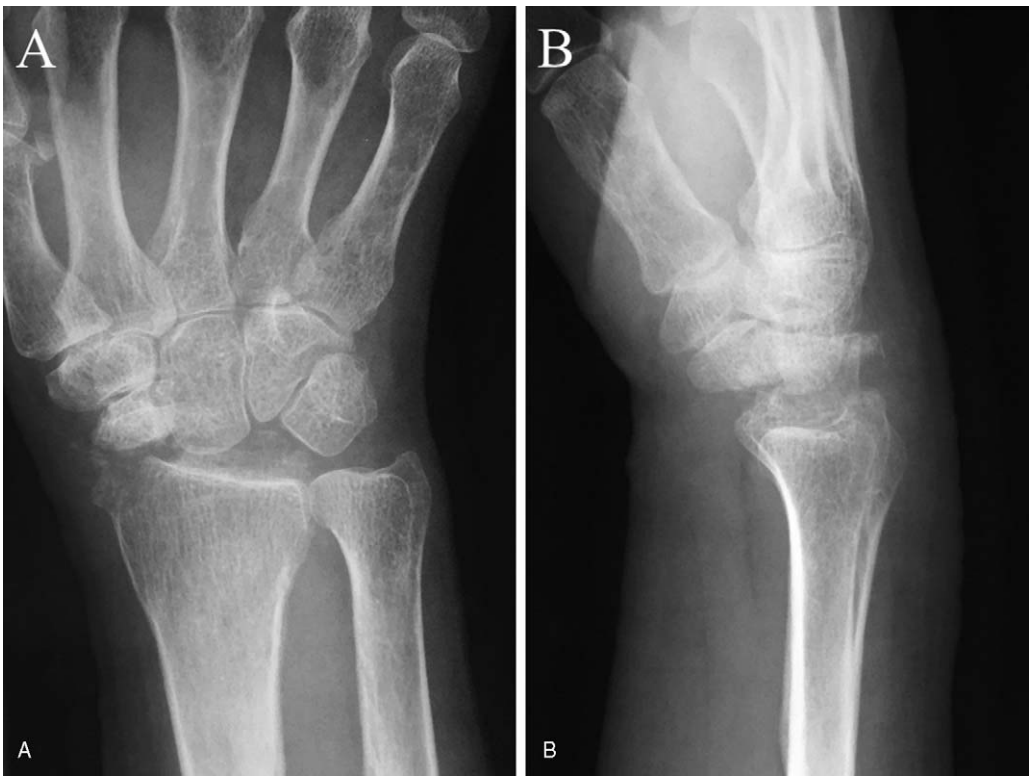

Figure 3. A. Postoperative posteroanterior (A) and lateral (B) radiographs. The fragmented proximal scaphoid and whole lunate were resected.

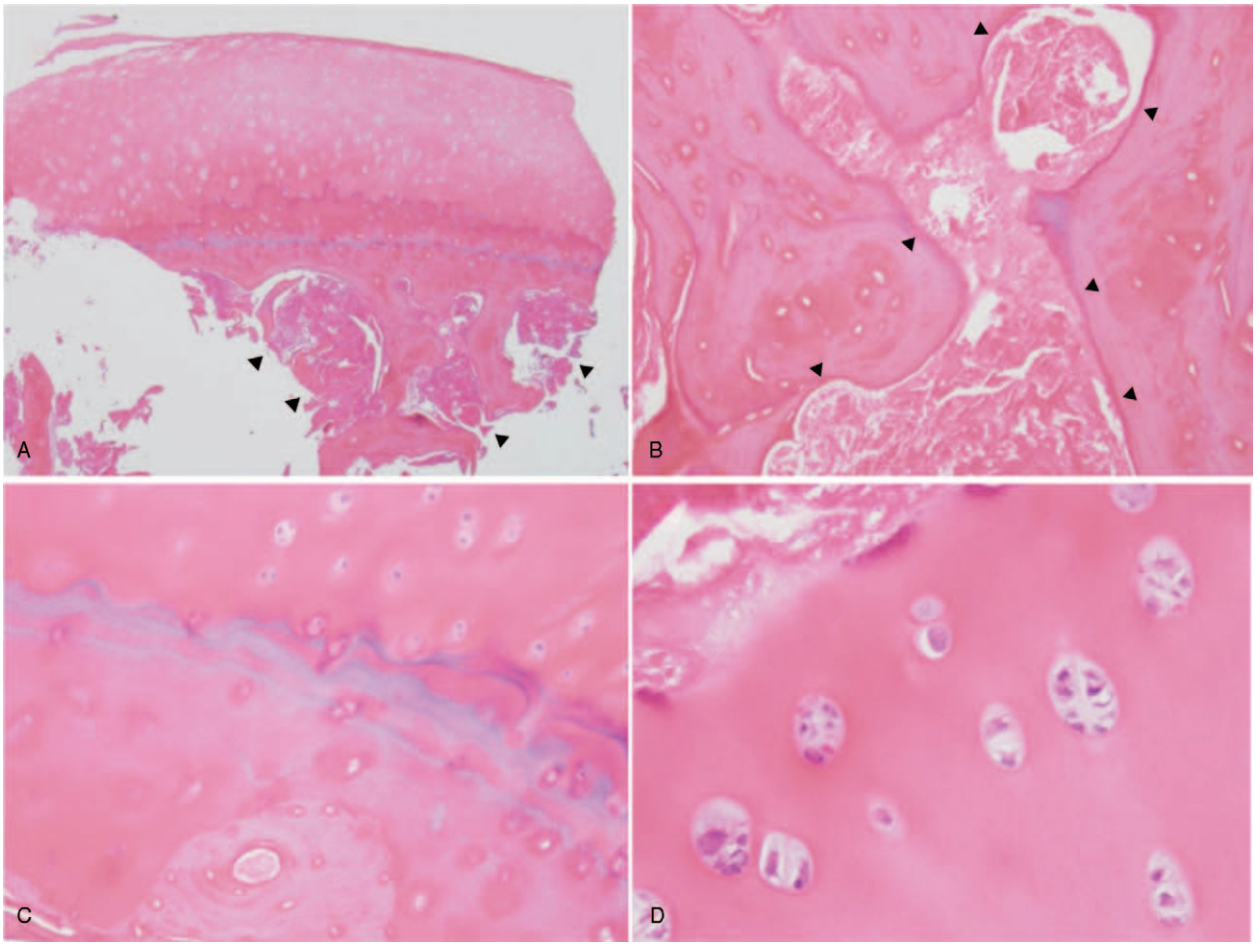

Figure 4. A fragment of the scaphoid bone shows an area of necrosis involving the entire subchondral zone (black arrow heads) (A) represented by empty lacunae and necrotic debris in the intertrabecular space (black arrow heads) (B). At higher magnification, multilayering of a tide mark (C) and chondrocytic cloning (D) are noted, suggesting a regenerative reaction. All histological findings are characteristic of avascular necrosis of the scaphoid bone. Hematoxylin and eosin stain, ×12.5 (A), ×100 (B), and ×200 (C, D).

and AVN of the carpal bones,[2,3,13–17] the underlying mechanisms remain unclear. However, GC usage is thought to induce AVN of the carpal bones by these same mechanisms.

A single or several intramuscular GC injections can induce AVN in animal models;[18–22,26] however, AVN induced by local steroid injection has not been reported in a clinical case. In fact, clinical case series and case reports have reported the use of local GC injection as non-surgical treatment for AVN of the scaphoid.[28] Although our patient had no history of taking high-dose systemic GCs, multiple local GC injections were administered over a short time period. Preoperative T1- and T2-weighted MRI of the right wrist showed insufficient blood supply and bone edema in the scaphoid.[24] The histopathological findings of the fragmented proximal scaphoid were consistent with AVN. The scaphoid has a poor blood supply. Its major blood supply arises primarily from the radial artery, and only 2 vessels enter the scaphoid.[29–31] The vessels entering the dorsal ridge of the scaphoid account for 70% to 80% of the internal vascularity of the bone; the distal segmental artery provides all of the vascular supply of the proximal pole of the scaphoid.[31] These conditions explain why the highest incidence of osteonecrosis is in the proximal third of the scaphoid. Although it is difficult to draw conclusions about human AVN from studies of animal models because of interspecies differences, repeated local GC injections might disrupt the blood supply of the scaphoid and might thus cause AVN. To date, no report has associated AVN of the scaphoid with repeated local GC injections. In this case, a relationship between the repeated local GC injections and AVN of the scaphoid was strongly suspected.

Generally, the early radiographic changes of AVN of the carpal bones are mostly undetectable, and nonoperative treatment is initially performed. However, nonsurgical treatment, including immobilization, oral analgesics, GC injections, and electrical stimulation, among others, do not provide satisfactory outcomes.[2,6,15,28] Our patient was initially treated with immobilization, oral analgesics, and repeated local GC injections, but her intolerable wrist pain persisted and radiographic progression of the condition was observed, indicating that surgery was required for AVN of the scaphoid. In our case, radiographs showed collapse of the proximal pole of the scaphoid and sclerotic changes of the lunate associated with degenerative arthritis of the radiolunate joint perioperatively. Resection of the collapsed proximal scaphoid and the entire lunate with styloidectomy restored pain-free wrist function. At final follow-up, the patient had no wrist tenderness or residual pain, though she had ongoing restriction of range of motion. She was satisfied with the results and had no desire to pursue additional surgery.

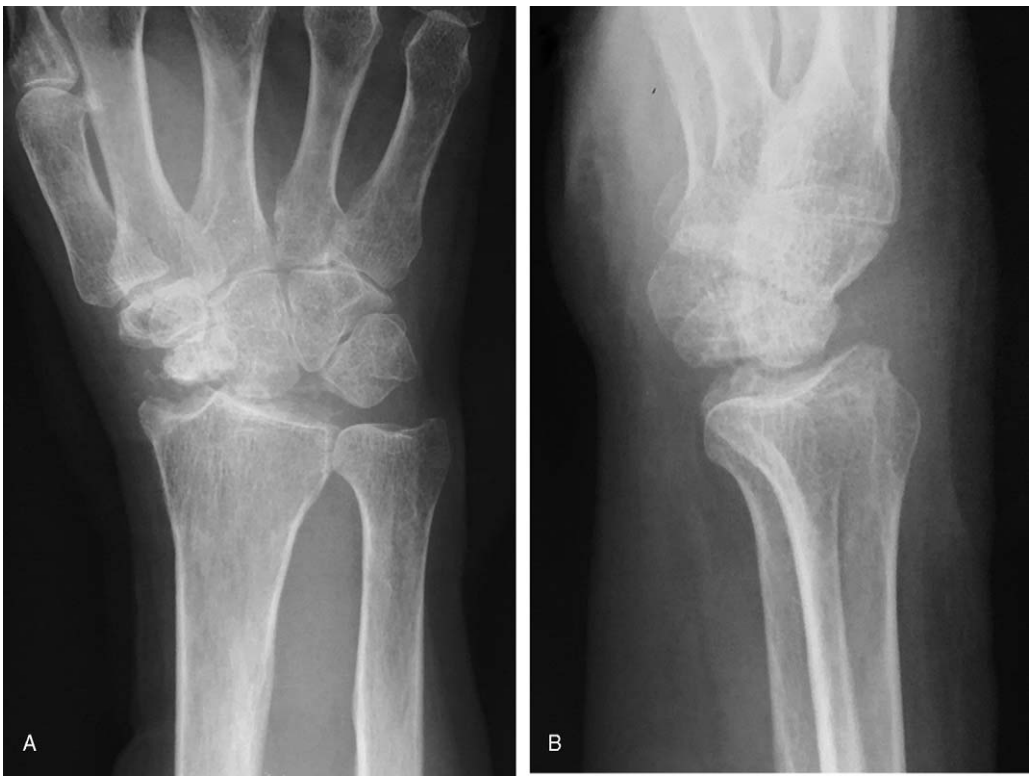

Figure 5. Posteroanterior (A) and lateral (B) radiographs of right wrist taken 1 year, 4 months after initial surgery show no progression of osteoarthritis.

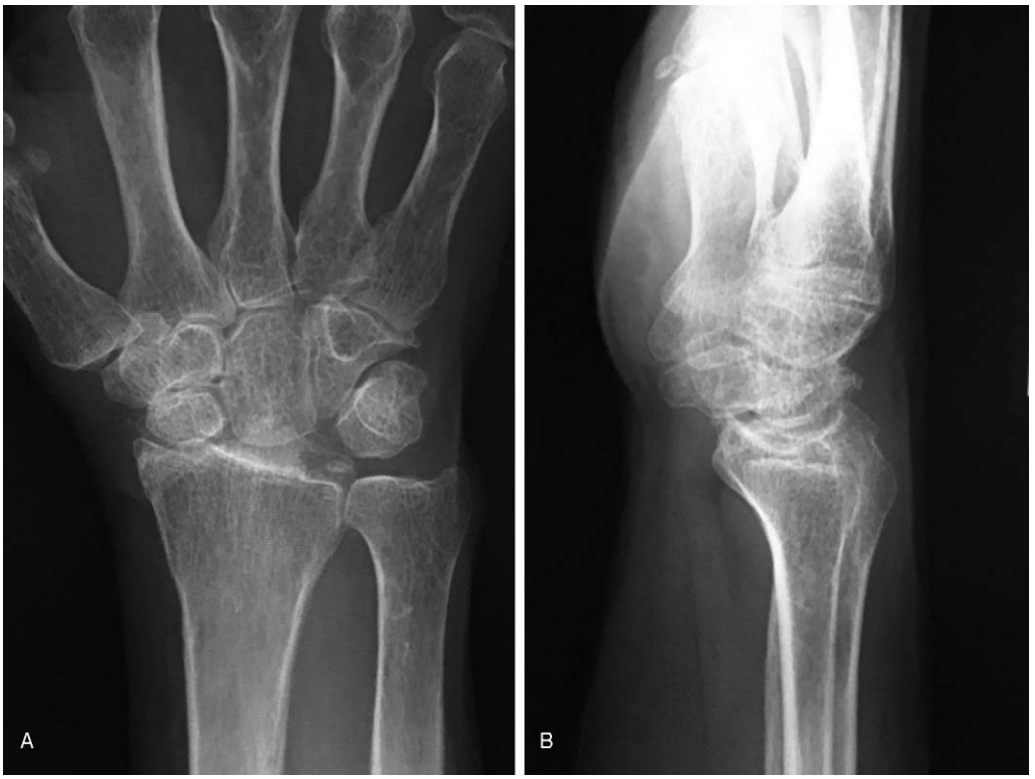

Figure 6. Posteroanterior (A) and lateral (B) radiographs taken 7 years, 1 month after surgery show no progression of osteoarthritis of the wrist. The patient was satisfied with results and had no desire to pursue additional surgery.

Although the optimal treatment has not been established because of the rarity of this condition, previously reported cases of Preiser disease have been treated with joint-leveling operations,[7,32–34] vascularized bone grafts,[16] or salvage procedures,[3,13,15,35] including proximal row carpectomy, partial or total wrist arthrodesis, and arthroplasty. In this case, we performed limited resection of the scaphoid and lunate. Histopathology of the proximal scaphoid, however, showed viable chondrocytes and chondrocytic proliferation in the articular cartilage, with findings suggesting avascular necrosis of the scaphoid. Thus, a vascularized bone graft might have been an optimal procedure in the present case.

In the present case, AVN of the scaphoid was strongly suspected to have resulted from repeated local steroid injection. Although AVN of the scaphoid induced by repeated local GC injection has not been reported in a clinical case, our report offers valuable insights for the appropriate use of local GC injections around the wrist joint. Further studies are required to determine whether repeated local injection of GC induces AVN of the carpal bones.

5. Conclusion

We have presented a case of Preiser disease after repeated local steroid injections. The patient had no history of excessive alcohol use, smoking, or trauma, except for local repeated steroid injections. AVN of the scaphoid was strongly suspected to be associated with the repeated local steroid injections. Further studies are required to more fully elucidate the association between AVN of the scaphoid and repeated local steroid injections.

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Author contributions

Data curation: Yuji Tomori, Takuya Sawaizumi.
Supervision: Takuya Sawaizumi, Shinro Takai.
Writing – original draft: Yuji Tomori.
Writing – review & editing: Norio Motoda, Ryuji Ohashi, Mitsuhiko Nanno.

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