Behind osteoporosis, Paget’s disease of bone (PDB), is the second most common bone remodeling disease.\textsuperscript{1} PDB, also known as osteitis deformans, is characterized by increased bone resorption by osteoclasts, followed by abnormal bone deposition by osteoblasts. This process results in misshapen, sclerotic bone with altered structural integrity. PDB can affect any bone in the body. The affected bone is often increased in size, making it prone to impaction, pain, and decreased range of motion. It is known to cause bone pain, from the pathologic bone remodeling.\textsuperscript{1} Generally, PDB is not associated with arthopathy, which contributes to the intrigue of this particular case. Occasionally, the enlargement of the ulnar bone can cause distal radioulnar joint (DRUJ) dysfunction and can be severe enough to warrant surgery, as was the case in this patient.
The semiconstrained linked total distal radioulnar joint arthroplasty (TDRUJA) (Aptis; Glenview, KY), popularized by Luis Scheker, allows near or close to full forearm pronation and supination, with improved grip strength for painful conditions involving the ulnar side of the wrist. This report describes our experience caring for a patient with a rare cause of ulnar-sided wrist pain due to ulnocarpal impaction from PDB involving the ulna. To date, there are no reports in the English literature regarding surgical management on ulnar-sided wrist pain related to PDB.

The purpose of this report is to describe a case of successful surgical management of ulnocarpal impaction and DRUJ arthropathy secondary to PDB with semiconstrained linked TDRUJA. Patients with PDB have pathologically enlarged bones. Thus, they are subsequently prone to impingement; in this case, radioulnar impingement. Other procedures, such as the Darrach and Sauvé-Kapandji (SK), are often utilized in the surgical management of ulnocarpal impaction and DRUJ arthropathy. However, these procedures likely would have led to pain from impingement in this particular case. In light of this, TDRUJA remains a viable option when there is a concern for radioulnar impingement-related pain.

Case Report

A 65-year-old right-hand-dominant retired male presented with chronic right ulnar-sided wrist pain of 20-year duration, which became disabling. He had severe pain with activities of daily living and fishing, his most passionate hobby. He also noticed a limited range of motion of his wrist and forearm compared with his nondominant hand. This limited range of motion had been long-standing since his teenage years. However, it never bothered him enough to seek medical attention until recently. He had no history of trauma nor a significant past medical or family history of orthopedic conditions. On physical exam, he had marked wrist asymmetry, notably from the right-sided distal ulnar prominence dorsally with limited, painful ulnar deviation of 5 degree (contralateral 30 degree).

Wrist extension, flexion, pronation, and supination all produced pain and crepitation and were restricted to 30, 30, 45, and 10 degree, respectively. His plain radiographs demonstrated severe DRUJ arthropathy and ulnocarpal impaction due to PDB of the ulna. Nonoperative and various surgical treatment options were reviewed in detail with the patient. After discussing the advantages and disadvantages of each, he elected for a TDRUJA. He was followed closely during the first 6 months when he had excellent pain relief and improved motion. He was released without restrictions to follow-up annually or as needed for pain. Recently, he was seen for 3-year follow-up, at which he stated he had zero pain, near-normal motion, and back to successful salmon fishing in Alaska.

Discussion

The novelty of this case lies in the patient’s decision to elect for surgical management, rather than medical treatment, of his PDB-induced DRUJ arthropathy. Furthermore, his decision to opt for semiconstrained linked TDRUJA over other surgical options and his excellent clinical outcome, as compared with outcomes after other options reviewed in the literature, are what makes this case unique. PDB by itself is often a painless or mildly painful disease process, consistent with this patient’s history of low-grade pain of 20 years’ duration without a diagnosis of PDB during this period. In general, bone pain related directly to PDB can be managed medically with bisphosphonates. However, PDB-related arthropathy appears
to be a more painful condition prompting surgical intervention as demonstrated by this patient’s secondary conditions of ulnocarpal impaction and DRUJ arthropathy. Although this patient’s entire ulna was affected by PDB, his history and physical exam were consistent with pain from these two secondary wrist conditions, which prompted him to elect for surgical rather than medical management.

Fig. 3 Plain radiograph of the patient’s forearm (lateral view) at presentation. Ulnar hypertrophy is appreciated from a different angle in this view.

Fig. 4 Plain radiograph which showcases successful semiconstrained DRUJ arthroplasty in this patient (AP view). AP, anteroposterior; DRUJ, distal radioulnar joint.

Fig. 5 Plain radiograph demonstrating another view of successful TDRUJA placement (lateral view). TDRUJA, total distal radioulnar joint arthroplasty.

Fig. 6 (A) Two-year follow-up showing satisfactory stress loading. (B) Two-year follow-up demonstrating satisfactory ulnar deviation. (C) Two-year follow-up demonstrating satisfactory supination. (D) Two-year follow-up demonstrating satisfactory pronation.
Our review of the literature does not provide guidelines for the management of this specific patient with ulnar wrist pathology related to PDB. There is, however, a large body of literature regarding the surgical management of his secondary conditions, particularly in the setting of primary osteoarthritis and rheumatoid arthritis. Traditionally, many surgeons have favored the Darrach or SK salvage procedures to address this type of ulnar wrist arthropathy. Some advantages of these procedures are the potential for long-term good clinical outcomes, including improved forearm range of motion without the need for arthroplasty implants, which could lead to painful loosening or may not be available, particularly in certain parts of the world. The major known disadvantage of the Darrach and SK procedures is the progression of distal ulnar impingement syndrome and instability, which causes significant pain. Dynamic distal ulnar impingement after a Darrach procedure can be demonstrated radiographically with an ulnar loading stress view. To abate this problem, some surgeons prefer primary or secondary Achilles tendon allograft interposition arthroplasty, which has demonstrated fair to good results in certain hands. Other surgeons have preferred primary or secondary use of the distal ulnar head endoprosthesis DRUJ hemiarthroplasty. These authors, with a relatively large volume of patients (47 cases from 1998 to 2008), reported a high reoperation rate of 30% during a median 56-month follow-up period. These revisions were due to stem loosening, sigmoid notch erosion, and especially dorsal or volar DRUJ instability related to the incompetence of the distal radioulnar ligaments. The semiconstrained linked TRDUJA has thus been supported by many authors to avoid the complications mentioned above related to the alternative treatments. Although short-term complications of the semiconstrained linked TRDUJA have been documented, there is a substantial body of evidence that long-term outcome and overall patient satisfaction may be better than alternative procedures. Several reports demonstrate long-term follow-up (averaging approximately 5 years) with satisfactory and improved patient subjective outcomes and most importantly, 90 to 100% implant survival.
Our patient was presented with all the above-mentioned surgical options, along with the advantages and disadvantages of each. After several preoperative visits, thorough discussion, and his online review of the literature, he elected for the semiconstrained linked TDRUJA. At the 3-year follow-up, he was satisfied with his lack of pain and improved motion, allowing him to return to salmon fishing in Alaska. He stated he would make the same decision again or refer his family for this same procedure.

Note
The work was performed at the Reno Orthopaedic Clinic.

Conflict of Interest
None declared.

References