



Arthrofibrosis after total elbow arthroplasty: a case report

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Total elbow arthroplasty (TEA) has proved successful in achieving pain control and an increased range of motion and function in a broad range of elbow pathologic processes. Known complications including nerve palsies, infection, aseptic loosening, triceps avulsion, prosthetic failure, and periprosthetic fractures are well described. To our knowledge, arthrofibrosis has not previously been described after TEA. We present a case of a painful stiff elbow in a patient after TEA refractory to multiple surgical and rehabilitative interventions. Extensive investigation failed to identify any cause other than excessive scar formation with a resultant painful restriction in range of motion.

Case presentation

A 59-year-old woman presented to our unit with a 1-year history of a painful and stiff right elbow of spontaneous onset. The course was progressive, and she had experienced no constitutional symptoms. She had a background history

of well-controlled hypertension and type 2 diabetes mellitus. On examination, the patient had a tender joint line, but the elbow was neither warm nor swollen. There were no clinical features of complex regional pain syndrome. Her arc of flexion was 75° to 85°, with −5°/15° supination/pronation. Pain was most severe with pronation and at extremities of flexion and extension. Her Mayo Elbow Performance Score at presentation was 15/100.

The radiologist's report on the radiographs described articular space narrowing and irregularity with sclerosis of the ulnohumeral joint, suggestive of degenerative changes, and osteopenia on both sides of the joint, possibly secondary to disuse (Figs. 1 and 2). The patient's clinical picture of an almost ankylosed joint and severe pain did not correlate with these mild radiologic changes, and further workup was initiated.

Blood investigations were unremarkable, with a white blood cell count of $6.05 \times 10^9/L$, an erythrocyte sedimentation rate of 7 mm/h, and a C-reactive protein level of <1.0 mg/L. Rheumatoid markers including rheumatoid factor, antinuclear antibodies, and anti-double-stranded DNA were all negative. A technetium bone scan showed moderately increased uptake around the elbow consistent with degenerative changes. Similar increased uptake was seen in multiple joints. Subsequent magnetic resonance imaging demonstrated no synovitis or abnormal enhancement to suggest an inflammatory process or neoplastic lesion (Fig. 3). The rheumatologists were consulted at a combined orthopaedic/rheumatology meeting, and they found no features of an inflammatory arthritis.

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Figure 1 Anteroposterior radiograph at presentation.



Figure 2 Lateral radiograph at presentation.

The patient was scheduled for an arthroscopic osteo-capsular release and biopsy of the elbow. Under anesthesia, the patient had full pronation and supination, and flexion was 60° to 110°, which improved to full flexion and extension after gentle manipulation. No arthroscopic release was therefore required. A biopsy was done through a

lateral approach, where a small bloody effusion with a friable hemorrhagic capsule was observed. Both the cartilage and bone were noted to be soft. Specimens of bone, cartilage, and capsule were sent for analysis. Postoperative orders were early active range of motion and indomethacin 25 mg orally three times daily.

Microscopy showed 1+ neutrophils, with no bacteria or acid-fast bacilli observed. Aerobic cultures showed no growth after 5 days, and cultures for acid-fast bacilli were negative after 49 days of incubation. On histologic examination, the synovial and bone biopsy specimens were within normal limits, with unremarkable connective tissue and a thin, flat synovial lining with no significant inflammation.

The postoperative range of motion deteriorated within 6 weeks to 60° to 100° of flexion and -30°/40° of supination/pronation despite a compliant patient and regular physiotherapy sessions. The patient continued to experience constant pain affecting her sleep and activities of daily living. Because of her significant symptoms and the finding of soft atrophic cartilage at the time of biopsy, a Discovery TEA (Biomet Orthopedics, Warsaw, IN, USA) was performed 18 months after her first presentation. The surgery was performed by an experienced shoulder and elbow surgeon (S.R.) with more than 50 TEAs performed in the 10 years prior. A Bryan-Morrey triceps reflecting approach was used. The ulnar nerve was released in situ. Full-thickness cartilage loss was noted on both the olecranon and radial head. Both components were cemented with antibiotic cement, and the triceps was repaired with 5 Ethibond through drill holes in the olecranon. Full range of motion was achieved, and the ulnar nerve was not transposed as it had normal tracking and was not tight in full flexion. Samples were again sent for microbiology and histology.

The postoperative course was uneventful, with active flexion and gravity-assisted extension commenced on day 2. The postoperative radiograph was reviewed as satisfactory by the senior consultant (S.R.). Culture specimens were again negative for aerobic organisms and acid-fast bacilli after prolonged culture. The histology report showed normal bone, cartilage, and fibrous and fatty connective tissue, with no evidence of crystal deposition, granulomatous inflammation, or malignant disease.

At 2-week follow-up, active flexion had deteriorated to 60° to 75°, with the elbow fixed in 10° of pronation. Inflammatory markers taken at 6 weeks postoperatively revealed a C-reactive protein level of 1.3 mg/L and an erythrocyte sedimentation rate of 1 mm/h. The patient was scheduled for manipulation under anesthesia, which was performed 3 months after the TEA. After manipulation, the range was 25° to 130° of flexion and 80°/45° of supination/pronation. A week of in-patient physiotherapy and night extension splints was commenced postoperatively, after which the range was 40° to 110° of flexion and 0°/20° of supination/pronation.

The range of motion continued to deteriorate and pain persisted, with nightly pain waking her from sleep. A gallium

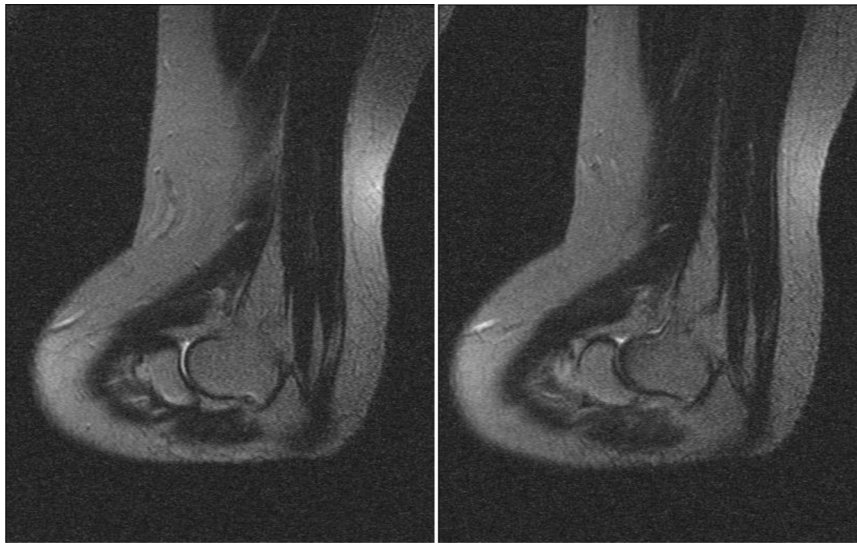


Figure 3 Sagittal T2 magnetic resonance image sequences showing no abnormal enhancement to suggest an inflammatory process, synovitis, or neoplastic lesion.

bone scan showed no evidence of sepsis. The patient was scheduled for an open release and biopsy, which was performed 1 year after the TEA. Examination under anesthesia showed flexion of 70° to 80°, with no pronation or supination. The original posterior skin incision was used and the ulnar nerve was released in situ under loupe magnification. The triceps attachment was intact. Thick fibrous tissue involving both the posterior and anterior capsule and extending between parts of the prosthesis was excised. Areas of the fibrotic capsule were observed to be up to 20 mm thick. Subperiosteal elevation of the flexors and extensors was performed and full flexion/extension was achieved. Specimens were again sent for microbiology and histology.

Microscopy showed 1+ neutrophils with no bacteria or fungi observed. Extended cultures including fungal cultures were negative. The histology report confirmed the presence of dense, vascular fibrous tissue with scanty lymphocytes and giant cells (Fig. 4). No inflammatory infiltrates, granulomas, or dysplastic or malignant cells were seen. The fibrous tissue was noted to extend into the adjacent muscle (Fig. 5). At 2-week follow-up, the range of motion was 45° to 90° of flexion and 5°/20° of supination/pronation. A manipulation under anesthesia was performed 8 weeks after the open release, and a range of motion of 25° to 120° of flexion and 45°/45° of supination/pronation was achieved. At 2 weeks after manipulation under anesthesia, the range had returned to the pre-manipulation range, despite compliance once again with aggressive mobilization protocols.

At most recent follow-up 3.5 years after the initial TEA, pain was worse, with daily analgesic requirements and interrupted sleep. Range of motion was 45° to 90° of flexion and 10°/10° of supination/pronation. Her Mayo Elbow Performance Score was 15/100. Radiographs confirmed a well-positioned prosthesis with no lucency or loosening (Figs. 6 and 7). She has subsequently been

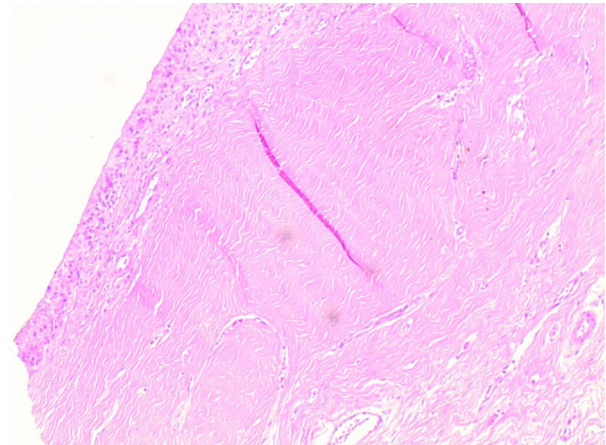


Figure 4 Histologic section demonstrating dense fibrous tissue with blood vessels and scanty lymphocytes.

referred to the knee unit for symptomatic osteoarthritis of her right knee, with a cautionary note attached regarding the outcome of her elbow arthroplasty.

Discussion

The elbow joint is prone to stiffness, particularly after trauma.⁹ Reasons for this increased susceptibility compared with other joints remain unclear, and we do not currently have a good understanding of the specific biologic, immunologic, or neurohormonal pathways implicated in this increased susceptibility.

The capsule clearly plays a dominant role as an extrinsic cause because its excision or release is required in treatment of contractures surgically.^{1,5,8} Gallay et al³ found the stiff elbow capsule to be one sixth as compliant as the normal

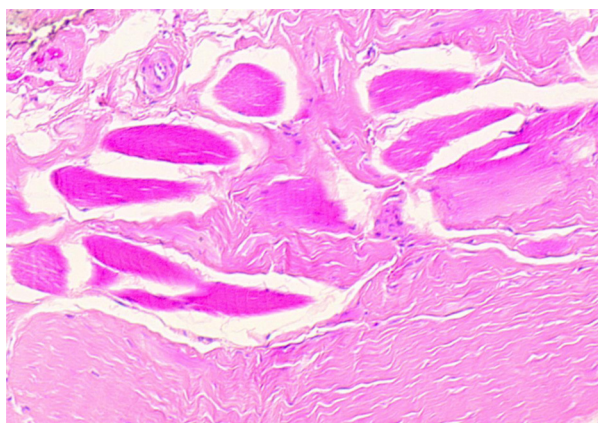


Figure 5 Histologic section demonstrating normal skeletal muscle with surrounding fibrosis.

elbow, concluding that it was altered structurally and biomechanically. They hypothesized that these changes involved hypertrophy of the capsule or a change in its collagen content. Cohen et al² demonstrated that the capsular contracture was significantly thickened (on average 7 times) compared with normal capsules. This thickening was accompanied by disorganization of collagen fiber arrangement, fibroblast and lymphocytic infiltration, and increased levels of cytokines in the capsular tissue compared with controls. Levels of type III collagen, which has been associated with normal wound healing, were present in the normal elbow capsule but consistently diminished in the contracture capsules. The cytokines analyzed in their study were the matrix metalloproteinases MMP-1, MMP-2, and MMP-3. Staining for MMP-1, MMP-2, and MMP-3 was significantly increased in the contracture group compared with the control group. An inhibitor of matrix metalloproteinases (tissue inhibitor of matrix metalloproteinase) used as a treatment modality for various malignant neoplasms has been associated with the development of frozen shoulder and suggests a key role for matrix metalloproteinases in the pathologic process of joint contractures.⁴

The patient in this report had features of both intrinsic and extrinsic stiffness. Radiologic changes, albeit mild, were present at her initial presentation to our unit. However, the initial response to manipulation under anesthesia, where she regained full range, suggested a predominantly extrinsic cause. This improvement in range, including a significant improvement while under anesthesia and before manipulation, suggests a disproportionate pain response with significant muscle spasm. We also believe that her rapid subsequent deterioration after biopsy was a result of an abnormal biologic process, possibly in response to the surgical insult, and this we believe was confirmed when a similar deterioration occurred after her TEA, with its associated extensive release.

The strengths of our case report include the thorough workup completed of the patient, which failed to identify any known cause of her condition, and the presence of



Figure 6 Anteroposterior radiograph 3.5 years after total elbow arthroplasty.

multiple histologic analyses, which confirmed after TEA that 20-mm-thick sections of dense, vascular fibrous tissue had formed within and around the prosthesis.

It is not clear whether this was a patient with an idiopathic or primary arthrofibrosis who had a TEA with subsequent



Figure 7 Lateral radiograph 3.5 years after total elbow arthroplasty.

recurrence of the arthrofibrosis or whether she developed the arthrofibrosis secondary to the TEA. Her almost ankylosed elbow at first presentation accompanied by minor radiologic changes and no identifiable cause suggests the former; however, histologic analysis at time of initial biopsy did not confirm features of arthrofibrosis, and it was only at the open release after the TEA that significant fibrosis was observed. Perhaps we encountered the patient as the disease process was unfolding, possibly accelerating its progression. Otherwise our surgical intervention must have provided the trigger for the arthrofibrosis.

Our decision to perform a TEA is open to criticism. However, in the face of the failure to respond to traditional treatment modalities for extrinsic elbow stiffness and the presence of macroscopic articular changes at the time of biopsy, we believe it was a valid option. An audit of our unit showed a mean improvement in range of motion of 36° (78°–115°) in 106 TEAs performed between 2000 and 2012 (unpublished data), and Morrey has shown it to be a successful treatment option for ankylosed joints, with a 60° mean improvement in range of motion after TEA.⁷ Unfortunately, unlike in the majority of patients treated at our unit and the patients in Morrey's series, TEA did not improve her range of motion or pain. We were ultimately left with the demoralizing outcome of a still painful and almost ankylosed elbow, after exhausting all surgical options that, in combination, had effectively removed all capsular, synovial, and cartilaginous tissue from the offending elbow joint. For unexplained reasons, surgery to address the local macroscopic disease had failed to address the driving force of her disease process, which, we can only surmise, must be on an immunologic or neurohormonal level.

The clinical course in our patient's case was consistent with a refractory elbow arthrofibrosis, a condition

previously described in another case series of 4 patients by Morrey.⁶ Patients in this series had a similar presentation, with a significant and disproportionate stiffness, moderate increase in uptake on bone scan, and early recurrence after surgical intervention. No histopathologic studies were completed in that series. The authors concluded that an awareness of this condition is required to avoid unsuccessful surgical releases. Their treatment options did not include TEA as none of the 4 patients had radiologic signs of articular cartilage loss. From our experience in this case report, we second their caution regarding surgical treatment of patients with significant and unexplained stiffness in the elbow and add that arthroplasty, with its associated soft tissue release, should be included in this group of failed surgical procedures.

Conclusion

Arthrofibrosis is a well-described complication after total knee arthroplasty, and although this poorly understood condition is known to occur in the elbow, to our knowledge it has not previously been described after TEA. In our single-patient case report, the condition proved to be refractory to multiple surgical and non-surgical interventions.

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Disclaimer

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