



ELSEVIER

ORIGINAL ARTICLE

Long-term results of total elbow arthroplasty in patients with hemophilia

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Hypothesis: It was hypothesized that the long-term survivorship and clinical outcome are reasonable, justifying total elbow arthroplasty (TEA) in patients with end-stage hemophilic arthropathy.

Methods: From 2002 to 2012, 13 primary TEAs (Coonrad-Morrey design) were implanted in 9 consecutive patients with an average age of 55 (range, 39-76) years. Type A hemophilia was diagnosed in 7 patients and type B hemophilia in 2 patients. Clinical and radiographic results of all (11 TEAs) but 1 patient were retrospectively analyzed.

Results: After a mean of 9.1 (range, 5-14) years, the mean visual analog scale score for pain, total Mayo Elbow Performance Score, and subjective elbow value were significantly improved from 5 (standard deviation, ± 3) to 2 (± 2 ; $P = .007$) points, from 64 (± 16) to 89 (± 11 ; $P = .008$) points, and from 47% ($\pm 15\%$) to 81% ($\pm 11\%$; $P < .001$), respectively. Whereas the flexion arc remained unchanged ($P = .279$), mean active pronation improved significantly ($P = .024$). Postoperative complications were recorded in 8 TEAs (62%), whereas 5 TEAs (38%) underwent partial component exchange after a mean of 7.2 (range, 3-10) years: 2 for periprosthetic infection, 2 for polyethylene wear, and 1 for humeral component loosening. Of the living patients after partial component exchange ($n = 3$), the mean final total Mayo Elbow Performance Score, flexion and rotation arc, visual analog scale score for pain, and subjective elbow value were comparable with the results of the living patients without revision surgery ($n = 8$).

Conclusions: TEA for patients with advanced hemophilic arthropathy is associated with a substantial complication and revision rate. However, even after revision without implant removal, it provides good functional and subjective long-term results.

Level of evidence: Level IV; Case Series; Treatment Study

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Investigation performed at the Balgrist University Hospital, Zürich, Switzerland.

The Cantonal Ethics Commission Zürich approved this retrospective study: KEK-ZH-Nr. 2015-0374. All patients gave their written consent for review purposes before commencement of the trial.

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In patients with hemophilia, spontaneous bleeding in the joints may lead to synovitis and cartilage damage, ending with progressive joint disease and the development of hemophilic arthropathy (HA).^{1,10} HA is debilitating, causing advanced joint destruction with contracture, and frequently involves the knee, ankle, and elbow joints.^{7,15,33} In general, adequate treatment of HA necessitates a multidisciplinary approach involving prophylaxis of the disease, management of acute intra-articular bleeds, serial imaging, and surgical intervention,⁷ when indicated. In the early stages of HA of the elbow, management includes radiosynovectomy, surgical synovectomy with or without radial head excision, and débridement.^{23,35,37} Treatment options in symptomatic end-stage HA are arthrodesis and total elbow arthroplasty (TEA).^{4,17} Nevertheless, compared with patients without bleeding disorders, the complication and revision rate after total joint replacement in the knee is higher.^{5,7,12,36,40,43} With increasing knowledge, indications for TEA have expanded,^{6,14,18,24,32} and it has also become a favorable intervention in patients with advanced HA.^{2-4,17,19,23,25,26,29,30,39} With regard to meaningful long-term outcome, however, there is still a paucity of reports of TEA in patients with HA.

It was the purpose of this study to analyze whether improvement in long-term clinical and radiologic outcome after TEA outweighs the potential complications and revision rates. It was hypothesized that the overall survivorship and clinical outcome are reasonable, justifying TEA in patients with end-stage HA.

Materials and methods

Patients

This is a retrospective cohort study analyzing 13 consecutive elbows in 9 hemophilic patients. Between March 2002 and December 2012, all elbows underwent TEA for HA; the mean age at surgery was 55 (range, 39-76) years.

The indications for TEA in all patients were the following: excessive pain; significant loss of range of motion (ROM); impaired elbow function; failed nonoperative treatment, including physiotherapy, nonsteroidal anti-inflammatory drugs, and intra-articular injection; and advanced HA confirmed on plain radiographs. Factor VIII deficiency was diagnosed in 7 patients (78%; 5 severe and 2 moderate), and 2 patients had factor IX deficiency (22%; 1 severe and 1 one moderate).⁴² Six patients had combined hepatitis B and C, and 3 had isolated hepatitis C. No patient was human immunodeficiency virus (HIV) positive. According to Arnold and Hilgartner,¹ 4 elbows (31%) were classified as stage 3 HA, 7 elbows (54%) as stage 4 HA, and 2 elbows (15%) as stage 5 HA. Before TEA, 3 elbows (23%) underwent excision of the radial head and 1 elbow (8%) underwent open synovectomy. All but 1 patient (89%) suffered from advanced polyarthropathy, with bilateral total knee replacement in all of these patients.

Surgical procedure

Surgery was performed bilaterally (consecutively) in 4 patients (44%). A posterior approach with triceps reflection was performed in all

procedures. The ulnar nerve was identified and routinely transposed subcutaneously after the implants were cemented in place. A synovectomy and contracture release were performed. In the 10 cases with the radial head in situ, an excision was carried out. The humerus and ulna were prepared, and the components were cemented in place with gentamicin-impregnated bone cement (Palacos; Heraeus Kulzer GmbH, Wehrheim, Germany). A Coonrad-Morrey prosthesis (Zimmer, Warsaw, IN, USA) was implanted in all cases. The tourniquet was deflated before closure to ensure hemostasis, and a deep drain was used for 24 hours per our unit's protocol. The wound was closed in anatomic layers, and the skin was sutured with nonabsorbable sutures.

After surgery, the elbow was placed in a plaster backslab for 2 weeks, followed by a plaster splint during the night for another 4 weeks. Postoperative rehabilitation included an active, assisted exercise regimen for 6 weeks under the direction of a physiotherapist.

Hemophilia management

The treating hematologists managed all patients during different operations and ensured sufficient factor replacement. All patients were treated with their specific factor VIII or factor IX products. The aim was a perioperative factor level of 100%, and this was usually achieved by 1 dose (50 IU of factor VIII per kilogram body weight or 80-100 IU of factor IX per kilogram body weight) before and the second dose after opening of the tourniquet calculated by the blood loss. Factor levels in the following days were reached either by continuous infusion or by injections every 4-6 hours with subsequent dose tapering. The aim was a factor level between 70% and 100% during the first 48 hours, between 50% and 70% during days 3 to 6, and between 40% and 60% until day 10. Full prophylaxis to keep trough level above 5%-10% was given during the physiotherapy period. Medical thromboprophylaxis was omitted.

Clinical and radiographic assessment

At final follow-up, an independent examiner not involved in the patient's treatment assessed the outcome of the 11 TEAs in all 8 living patients in an institutionally standardized manner. Clinical examination included measurement of active and passive ROM using a goniometer and assessment of the Mayo Elbow Performance Score (MEPS),^{20,27} visual analog scale (VAS) score for pain, subjective elbow value (SEV),³⁴ and Medical Outcomes Study 36-Item Short Form Health Survey (SF-36). The SF-36 can be summarized by 2 aggregate scores, the physical component summary and the mental component summary. Higher scores of the MEPS, SEV, and SF-36 represent better overall results. Patients rated their overall postoperative results on a scale of poor, fair, good, and excellent.

Preoperative and postoperative standardized anteroposterior and lateral radiographs were made in all patients (n = 8) and elbows (n = 11) available. Postoperative radiographs were evaluated by 2 independent raters not involved in the patient's treatment (R.S., L.E.) for the cementation status, component loosening, and bushing wear. The cement mantle was graded type 1 (adequate cementation), type 2 (marginal cementation with a 2-mm-wide radiolucent line at the bone-cement interface), and type 3 (inadequate cementation with a >2-mm-wide radiolucent line at the bone-cement interface).²⁸ Radiolucency around the components was defined as type 0 (no relevant lucency), type 1 (1-mm line involving <50% of the interface), type 2 (>1-mm line involving >50% of the interface), type 3 (>2-mm line

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