Clin Orthop Relat Res (2011) 469:1427–1435
DOI 10.1007/s11999-010-1483-5

CLINICAL RESEARCH

# Total Ankle Arthroplasty in Patients with Hereditary Hemochromatosis

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Received: 2 April 2010 / Accepted: 12 July 2010 / Published online: 28 July 2010


**Abstract**

*Background* More than half of patients with hereditary hemochromatosis (HH) have painful arthritis, often including hindfoot osteoarthritis. Total ankle arthroplasty (TAA) is increasingly recommended for patients with painful ankle osteoarthritis. However, the pain relief and function experienced by patients continues to be debated particularly as compared with ankle fusion.

*Questions/purposes* We asked whether (1) the complication rates were low; (2) the components were stable; (3) the patients achieved pain relief; and (4) the patients had satisfactory midterm function, ROM, and quality of life.

*Patients and Methods* We retrospectively reviewed all 16 prospectively followed patients (21 implants) with HH who underwent ankle arthroplasty. They had an average age of 59.5 years at the time of surgery. We obtained a visual analog scale for pain, the SF-36, and the American Orthopaedic Foot and Ankle Society (AOFAS) hindfoot score. Component stability was assessed using weightbearing radiographs. The minimum followup was 3.1 years (average, 5.3 years; range, 3.1–8.6 years).

*Results* Postoperatively, one patient had débridement of a painful cyst on the tibial side and one patient had a subfibular débridement with a lateral ligament reconstruction. The tibial and talar components were stable in all ankles. The average pain score decreased from 6.7 (range, 3–10) to 1.9 (range, 0–4). All eight categories of SF-36 score showed improvement. The hindfoot score increased from 46 (range, 22–67) to 84 (range, 74–94).

*Conclusions* Our data suggest TAA in patients with ankle osteoarthritis secondary to HH is associated with a low risk of postoperative complications and produces pain relief and good function.

*Level of Evidence* Level IV, therapeutic case series. See Guidelines for Authors for a complete description of levels of evidence.

One or more of the authors (BH) have received royalties from Integra Orthopedics (Plainsboro, NJ), which were given to the research fund at the clinic where the work was performed. One or more of the authors (AB) have received research funding from Swiss Society of Orthopaedics and Basel Foundation Orthopaedic Surgery and Biomechanics of Foot and Ankle.
Each author certifies that his or her institution approved the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

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## Introduction

HH is an inherited autosomal recessive disorder of iron metabolism in which excess iron absorption leads to tissue damage associated with a characteristic arthritis. Clinical manifestations of pathologic iron accumulation include liver disease, skin pigmentation, diabetes mellitus, cardiac enlargement, and joint osteoarthritis [1, 23]. Most commonly, the hands are affected first [14]. However, Schmid et al. [34] presented three cases that illustrated symmetric hindfoot involvement as the initial clinical manifestation of HH.

The current standard surgical treatment for patients with advanced painful arthropathy is ankle fusion [28]. This procedure reportedly relieves pain but reduces functional ability of patients with posttraumatic ankle arthritis [5] and can cause progression of degenerative changes in neighboring joints [5, 8].

TAA is becoming an increasingly recommended treatment for patients with rheumatoid or posttraumatic arthritis of the ankle [4, 10]. One report suggests patients have pain relief and high satisfaction after surgery [4]. Saltzman et al. [33] recently published initial findings from a prospective controlled trial of the Scandinavian Total Ankle Replacement System® (STAR) TAA versus ankle fusion and reported TAA led to better function and similar pain relief compared with ankles that were fused. One study reported five TAAs in four patients with HH [6] and showed all four patients had good relief of symptoms after surgery. Although TAA is becoming more commonly used, the relative benefits of TAA continue to be one of the most debated topics in foot and ankle surgery [11]. First, there is a steep learning curve associated with performing TAA [20, 27, 31, 35]. Intraoperative complications are common and include medial or lateral malleolar fractures and lacerations to the tendons (posterior tibial tendon, flexor digitorum/hallucis longus), and nerves (deep peroneal nerve, superficial peroneal nerve) [20, 27, 31, 35]. Furthermore, TAA is associated with a higher rate of major revision surgery than ankle fusion, ranging from 9% to 23% at 1 to 5 years for TAA and 5% to 11% for ankle fusion at 1 to 5 years [36]. Also, the component stability in TAA is lower than after arthroplasty of the knee or hip. The overall failure rate is approximately 10% at 5 years with a wide range in published studies (0% to 32%) [10]. The choice of fusion versus TAA remains controversial, and how TAA performs in patients with HH is not well understood.

We therefore determined (1) the intraoperative and postoperative complications, including surgical revision for any reason; (2) the prosthesis component stability; (3) whether TAA provides pain relief in the midterm; and (4) the midterm functional outcome, including ROM and quality of life.

## Patients and Methods

We retrospectively reviewed data from all 16 prospectively followed patients who had 21 TAAs for painful, disabling ankle arthritis attributable to HH between April 2001 and November 2006. The subset of patients in this study (patients with severe ankle arthropathy attributable to HH) was part of a larger prospective study group that included 655 patients who underwent TAA during the same time. There were two women (two ankles) and 14 men (19 ankles) with a mean age of 59.5 ± 10.5 years (range, 44.4–80.9 years) (Table 1). The minimum followup was 3.1 years (mean, 5.3 years; range, 3.1-8.6 years). No patients were lost to followup. No patients were recalled specifically for this study; all data were obtained from medical records and radiographs. The protocol was approved by the Ethics Committee of the University of Basel (Basel, Switzerland). All participants were informed and provided written consent before surgery and the study. The study was conducted in accordance with the Declaration of Helsinki and the Guidelines on Good Clinical Practice.

**Table 1.** Demographic data for 16 patients with HH (21 TAAs)

| Case | Gender | Age of patient* (years) | BMI* ($kg/m^2$) | Followup (years) |
|---|---|---|---|---|
| 1 | Female | 61.7 | 25.2 | 8.6 |
| 2 | Male | 64.8 | 26.5 | 7.6 |
| 3† | Male | 56.7 | 22.1 | 7.6 |
| 4 | Male | 71.6 | 34.2 | 7.0 |
| 5 | Female | 53.4 | 28.4 | 7.0 |
| 6 | Male | 51.4 | 27.6 | 6.6 |
| 7 | Male | 46.3 | 34.0 | 6.0 |
| 8 | Male | 51.5 | 29.5 | 5.9 |
| 9 | Male | 44.4 | 24.4 | 5.8 |
| 10 | Male | 80.9 | 25.6 | 5.7 |
| 11† | Male | 54.2 | 28.7 | 5.0 |
| 12 | Male | 49.9 | 29.9 | 4.9 |
| 13† | Male | 64.7 | 23.1 | 4.7 |
| 14† | Male | 74.9 | 25.5 | 4.6 |
| 15† | Male | 75.5 | 25.4 | 4.0 |
| 16† | Male | 55.9 | 27.1 | 3.3 |
| 17† | Male | 61.1 | 22.1 | 3.2 |
| 18 | Male | 66.3 | 28.7 | 3.2 |
| 19† | Male | 66.6 | 25.9 | 3.1 |
| 20‡ | Male | 49.0 | 28.3 | 3.1 |
| 21‡ | Male | 49.0 | 28.3 | 3.1 |
| Mean ± SD | | 59.5 ± 10.5 | 27.2 ± 3.2 | 5.3 ± 1.7 |

* At the time of the surgery; †bilateral TAAs; ‡simultaneous bilateral TAAs; HH = hereditary hemochromatosis; TAA = total ankle arthroplasty; BMI = body mass index.

For all patients, the diagnosis was confirmed clinically and genetically. All patients were homozygous for C282Y/C282Y (the most common mutation). Laboratory findings, involvement of other joints, and additional data regarding HH status are presented in Table 2. Antinuclear antibodies and rheumatoid factor were not detectable. Serologic tests for hepatitis B and C were negative. None of the patients had reported any major ankle trauma.

All surgery was performed by one surgeon (BH). The HINTEGRA® (Newdeal SA, Lyon, France) was used in all patients. It is a nonconstrained three-component system that provides intrinsic stability in the coronal plane (eg, against eversion-inversion) [12]. Through an anterior longitudinal incision between the extensor hallucis longus and anterior tibial tendons, anterior arthrotomy and capsulectomy were performed for ankle exposure. First, the tibial cut was made using a special tibial resection bloc (resection of 2–3 mm subchondral bone). After the tibial cut was

**Table 2.** Laboratory and other findings for 16 patients (21 TAAs) with HH

| Case | Current phlebotomy* | Serum iron level† (mg/dL) | Serum ferritin level‡ (mg/dL) | Transferrin saturation§ (%) | Other skeletal involvement | | | | | Diabetes mellitus |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | Contralateral ankle | Knee | Hip | MCP (II, III) | Shoulder | |
| 1 | Yes | 249 | 6749 | 87 | Yes | No | Yes | Yes | No | Yes |
| 2 | Yes | 267 | 2111 | 86 | Yes | No | TJA | Yes | No | Yes |
| 3‖ | Yes | 220 | 2300 | 81 | TJA | No | No | No | No | No |
| 4 | No | 199 | 5723 | 79 | Yes | No | No | Yes | No | No |
| 5 | Yes | 228 | 5328 | 91 | Yes | No | No | Yes | No | Yes |
| 6 | No | 269 | 1855 | 77 | Yes | Yes | No | Yes | No | No |
| 7 | Yes | 244 | 6219 | 83 | Yes | No | No | Yes | No | No |
| 8 | Yes | 258 | 5934 | 84 | Yes | No | No | Yes | No | No |
| 9 | Yes | 205 | 4571 | 85 | Yes | No | No | Yes | No | No |
| 10 | Yes | 218 | 4511 | 83 | Yes | Yes | No | Yes | No | No |
| 11‖ | Yes | 227 | 3200 | 79 | TJA | No | No | Yes | No | Yes |
| 12 | Yes | 221 | 4592 | 90 | Yes | No | No | Yes | No | No |
| 13‖ | Yes | 204 | 1469 | 73 | TJA | TJA | TJA | Yes | Yes | No |
| 14‖ | Yes | 198 | 1699 | 76 | TJA | No | No | Yes | No | No |
| 15‖ | Yes | 220 | 3720 | 79 | TJA | No | No | Yes | No | No |
| 16‖ | Yes | 234 | 3567 | 78 | TJA | No | No | Yes | No | Yes |
| 17‖ | Yes | 267 | 3494 | 84 | TJA | No | No | No | No | No |
| 18 | No | 288 | 5387 | 81 | Yes | TJA | No | Yes | No | No |
| 19‖ | Yes | 285 | 1703 | 80 | TJA | TJA | TJA | Yes | Yes | No |
| 20¶ | Yes | 222 | 3662 | 79 | TJA | No | TJA | Yes | No | No |
| 21¶ | Yes | 222 | 3662 | 79 | TJA | No | TJA | Yes | No | No |
| Mean ± SD | | 238 ± 28 | 3879 ± 1627 | 82 ± 5 | | | | | | |

* At the latest followup; †before treatment (normal range, 198–288 mg/dL); ‡before treatment (normal range, 12–200 mg/dL); §before treatment (normal range, 20%–40%); ‖bilateral TAAs; ¶simultaneous bilateral TAAs; HH = hereditary hemochromatosis; TAA = total ankle arthroplasty; MCP = metacarpophalangeal joint; TJA = total joint arthroplasty.

made, the medial, lateral, and then the posterior cuts of the talar surface were made and the bone slices removed. The talar component was inserted first with the aim to obtain a proper press-fit on the talar bone. The trial tibia and mobile bearing then were inserted. Before the selected final components were inserted, the alignment, stability, and joint motion achieved were checked clinically. Although primary stability of the tibial component is obtained by screw fixation and six pyramidal peaks, primary stability of the talar component is obtained by press-fit and screw fixation (Cases 1–5). Since April 2003, two pegs, instead of screws, were used for fixation of the talar component (Cases 6–21). In the case of malalignment and concomitant osteoarthritis of the adjacent joints, additional surgeries (one-stage procedures) were performed before prosthetic implantation: subtalar and talonavicular fusion (Case 6) and tarsometatarsal II fusion (Case 18). For three patients (Cases 6, 8, 20), percutaneous lengthening of the Achilles tendon was performed. In one patient (Case 19), a TKA prosthesis was implanted on the contralateral side. In one patient (Cases 20, 21), bilateral TAAs were performed as a one-stage procedure. In the other four patients, the second ankle operation was performed 12.9 ± 7.8 months (range, 4.6–20.3 months) after the first surgery. As surgery was performed with a tourniquet mounted at the ipsilateral thigh, no intraoperative bleeding was detected. The tourniquet limit of 2 hours was not exceeded. No patients required postoperative blood transfusion.

After surgery, a well-padded short-leg splint held the foot in a neutral position. After 24 hours, the drain (without suction) was removed. In all cases, the drain production was less than 150 mL. After 2 days, the dressing and splint were removed, and a short-leg walking cast was applied. Three to 4 days after surgery, when the wound condition was dry, the cast was changed to a stable walker (VACO®ped; OPED AG, Steinhausen, Switzerland). The duration of mobilization with a walker was 6 weeks when only the ankle had been replaced and 8 weeks when additional procedures such as adjacent joint fusion had been performed. Full weightbearing with a stable walker was allowed as tolerated in all patients (with and without additional surgical procedures). Active and passive motion and lymphatic drainage were performed in all patients to support the recovery of soft tissues during the first 6 weeks. A rehabilitation program was continued after walker removal for at least 4 months, including walking exercises, stretching, and strengthening of the triceps surae. Patients also received instruction in ankle motion and balance/proprioception. Postoperatively, a low (eg, hiking, swimming, biking, golfing) level of sports activities was recommended, and a normal (eg, jogging, tennis, downhill skiing) level of sports activities was allowed; however, all patients were instructed to avoid contact sports or activities that involved jumping.

The first clinical and radiographic followup was at 6 weeks to check the wound site and osteointegration and position of the implants (using weightbearing radiographs in two planes). The next clinical and radiographic followups were at 4 months, 1 year, and then annually thereafter. All patients were seen postoperatively in our outpatient clinic by two independent reviewers (AE, AB) who did not operate on any of patients. The clinical examination involved assessment of ankle alignment and ROM with the patient standing (passive ROM activity) and ankle stability with the patient sitting. The ROM was determined clinically with a goniometer along the lateral border of the leg and foot. This examination was performed preoperatively and postoperatively. Patients rated their pain on a visual analog scale (VAS) of 0 points (no pain) to 10 points (maximal pain) [13]. In addition, the AOFAS hindfoot score (an unvalidated scoring system) was calculated [17]. Each patient's sports activity level was documented preoperatively and during the latest followup using the following score: Grade 0, none; Grade 1, moderate; Grade 2, normal; Grade 3, high; and Grade 4, elite [38]. All patients also fully completed SF-36 questionnaires on quality of life preoperatively and postoperatively [39]. Finally, the patients' gait was observed clinically and then analyzed using pedobarography.

Affected ankles were evaluated preoperatively based on weightbearing radiographs in two planes. Degree of degenerative changes in tibiotalar and adjacent joints and tibiotalar angle were evaluated. In patients with obvious varus or valgus hindfoot deformity, a hindfoot alignment view as described by Saltzman and el-Khoury also was acquired [32]. Postoperative radiographic examinations were obtained using fluoroscopy to standardize the AP and lateral views of relevant components. Ankle radiographs were acquired with the patients in a weightbearing position. Two of us (AB, AE) determined alignment (coronal and sagittal alignment) and component migration in the patients' ankles digitally [12, 37], using the metric software system (ImagicAccess®; PIC Systems AG, Glattbrugg, Switzerland). Furthermore, the appearance of heterotopic ossification was registered. Loosening of the tibial component was defined as a change in position of the component's flat base by greater than 2° relative to the tibia's long axis and/or as a progressive radiolucency greater than 2 mm on the AP or lateral radiograph. We presumed 2 mm could be reliably judged radiographically. Loosening of the talar component, as seen on the lateral radiograph, was defined as subsidence into the talar bone by greater than 5 mm or a change in position greater than 5° relative to a line drawn from the top of the talonavicular joint to the tuberosity of the calcaneus [12]. Evaluation of minor changes (ie, those less than 1 mm or 1.5°) in the position of the talar component on the AP radiograph was very difficult, and it was not possible to evaluate radiolucencies beneath the talar component on either view. All decisions were based on consensus. The interobserver reliability of the radiographic evaluation was 0.902 (95% confidence interval [CI], 0.843–0.958) for coronal alignment of the tibial component, 0.916 (95% CI, 0.866–0.961) for sagittal alignment of the tibial component, and 0.892 (95% CI, 0.812–0.934) for sagittal alignment of the talar component. The method and all reference lines and angles used for evaluation of alignment of the tibial and talar components were described previously [12]. For patients with suspicion of loosening or subsidence, we recommend performing CT or single photon emission CT (SPECT-CT) [29].

A Kolmogorov-Smirnov normality test was performed to verify whether our data met the assumptions of a parametric test. The prosthesis component stability was assessed by radiographic evaluation of prosthesis component alignment and migration. The intraclass correlation coefficients (ICC) and their 95% CI were used to summarize the interobserver reliability. An ICC of 1 indicated perfect reliability, and an ICC greater than 0.8 indicated excellent reliability [7]. We determined differences in VAS pain and SF-36 scores between preoperative and postoperative status using the Mann-Whitney rank-sum test. We determined differences in ROM between preoperative and postoperative status using Student's t-test. Data were analyzed using SPSS® Version 9.0 (SPSS Inc, Chicago, IL, USA) and SigmaPlot® 2004 for Windows® (Systat Software Inc, San Jose, CA, USA).

## Results

Wound healing occurred within 2 weeks of the surgery, free of adverse events. In one patient (Case 18) the dorsalis pedis artery was cut during tarsometatarsal-II arthrodesis. Vessel repair was performed with an ipsilateral autologous saphenous vein graft by a vascular surgeon. At the followup 3 months postoperatively, duplex sonography showed complete occlusion of the graft. However, we could not observe decreased blood circulation or sensitivity of the foot and toes.


**Fig. 1A–I** Preoperative (**A**) lateral and (**B**) AP radiographs show the left side (Case 14) in a 75-year-old man with painful bilateral ankle osteoarthritis. (**C**) Lateral and (**D**) AP radiographs show the right side of the same patient (Case 15). (**E**) A preoperative hindfoot view according to Saltzman and el-Khoury shows normal hindfoot alignment. Postoperative (**F**) lateral and (**G**) AP radiographs show proper position of both prosthesis components on the left side (Case 14; followup, 4.6 years). Postoperative (**H**) lateral and (**I**) AP radiographs of the right side also are shown (Case 15; followup. 4.0 years).


**Fig. 2A–B** Postoperative (**A**) lateral and (**B**) AP radiographs of a 56-year-old man taken at 3.3 years' followup show heterotopic ossification on the posteromedial side of the ankle.

Tibial and talar components were radiographically stable in all ankles (Fig. 1). On the tibial side, no radiolucent lines were seen and no migration of the talar component was detected. In one patient with a double arthrodesis (subtalar and talonavicular fusion) (Case 6) and in one patient with a tarsometatarsal-II arthrodesis (Case 18), a solid fusion on the site of arthrodesis was detected. Three patients (Cases 4, 10, 16) had heterotopic ossification on the posteromedial side (Fig. 2).

Four of the 16 patients were completely pain free and all patients experienced substantial pain relief. Overall, the average pain score (VAS) decreased ($p < 0.001$) from

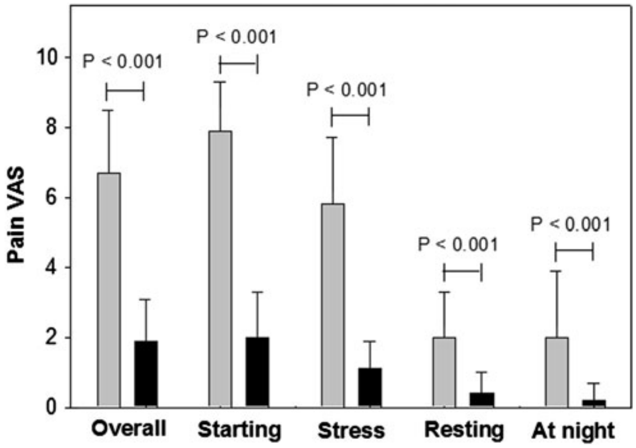


**Fig. 3** A graph shows preoperative (gray columns) and postoperative (black columns) levels of pain (VAS) for all patients. Data are presented as mean ± SD.

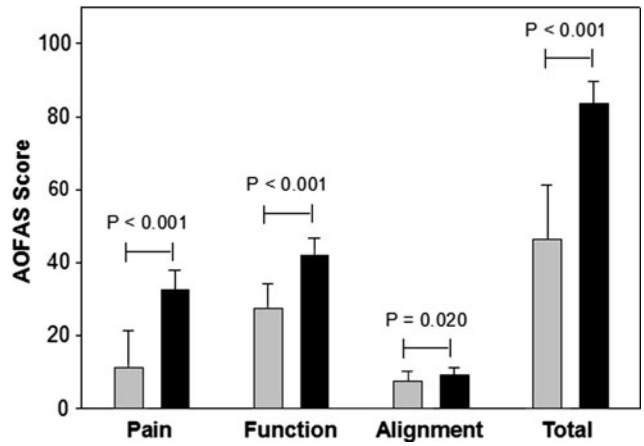


**Fig. 4** A graph shows preoperative (gray columns) and postoperative (black columns) AOFAS-hindfoot scores for all patients. Data are presented as mean ± SD.

6.7 ± 1.8 (range, 3–10) to 1.9 ± 1.2 (range, 0–4) (Fig. 3). Two patients (Cases 11, 13) had chronic pain develop attributable to arthrofibrosis. Therefore, 15 and 18 months after the primary surgery, respectively, open arthrolysis and percutaneous lengthening of the Achilles tendon were performed. Thereafter, one patient (Case 11) had a reduced pain level and the other patient (Case 13) was pain free. In both cases, motion was improved. Postoperatively a painful cyst developed on the tibial side in one patient (Case 2). Therefore, 36 months after primary surgery, an open cyst débridement was performed. Intraoperatively, no connection to the tibial prosthesis-bone interface was observed and both prosthesis components were considered stable. One patient (Case 19) complained of chronic pain on the lateral aspect of the ankle owing to subfibular impingement. For this patient, subfibular débridement and lateral ligament reconstruction were performed 19 months after the primary surgery. At 38 months' followup, no instability of the affected ankle was observed.

The AOFAS hindfoot score increased ($p < 0.001$) from 46 ± 15 (range, 22–67) preoperatively to 84 ± 6 (range, 74–94) (Fig. 4). Physical examination of the affected joints at latest followup showed no joint swelling, instability, or axial deformity of the affected joints. The average ROM increased ($p = 0.002$) from 29.6° ± 10.9° (range, 6°–45°) preoperatively to 39.3° ± 8.2° (range, 23°–56°) postoperatively (Table 3). An increase in ROM was observed for all patients; however, in 12 of the 21 ankles, ROM improvement was less than 10° (mean, 9.7° ± 7.2°; range, 1°–28°). All categories of SF-36 score showed improvements (Fig. 5). Also, the summarized components of the physical and mental outcomes scores improved from 30.5 ± 6.2 to 77.7 ± 4.7 ($p < 0.001$), and from 56.4 ± 4.9 to 78.8 ± 2.2 ($p < 0.001$), respectively. At latest followup, four of 19 patients had a normal level of sport activity and four of 19 patients had a moderate level, whereas preoperatively only two of 19 patients had a normal level of sports activities and two of 19 had a moderate level.

## Discussion

The role of TAA in patients with advanced painful arthropathy remains controversial. The use of TAA as an alternative to ankle fusion is continuously expanding, but reported results have been limited to small patient numbers and short-term followups. An important concern regarding TAA is the high rate of intraoperative complications [20, 27, 31, 35] and high rate of reoperations after this procedure [36]. We therefore determined (1) the intraoperative and postoperative complications, including surgical revision for any reason; (2) the prosthesis component stability; (3) whether TAA provides pain relief in the midterm; and (4) the midterm functional outcome, including ROM and quality of life.

Our study has some limitations. First, the surgeon who performed all of the procedures (BH) was involved with the design of the prosthesis, which raises concern regarding possible conflict of interest. However, the clinical evaluation was performed by two reviewers (AE, DH), and the radiographic evaluation was performed by two of the authors (AB, AE) who did not participate in any of operations. Second, low complication rates and good outcomes including ROM and quality of life observed in this study may relate to the surgeon's experience in performing TAA. Therefore, our results may not be generalizable for orthopaedic surgeons not familiar with the type of prosthesis used in this study. Third, the AOFAS score used for the clinical evaluation is not validated. Finally, followup in this study was limited to an average of 5.3 years. Patients

**Table 3.** Clinical results for 16 patients (21 TAAs) with HH

| Case | Followup (years) | ROM* (°) | | | AOFAS score | | SF-36 (maximum 100) | | | | Sports activity level | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | Preoperative | Postoperative | Change | Preoperative | Postoperative | Preoperative | | Postoperative | | Preoperative | Postoperative |
| | | | | | | | Physical health | Mental health | Physical health | Mental health | | |
| 1 | 8.6 | 7/13 | 11/26 | 17 | 28 | 93 | 33.8 | 59.3 | 83.8 | 83.5 | None | Moderate |
| 2 | 7.6 | 13/23 | 14/27 | 5 | 61 | 77 | 36.3 | 59.3 | 83.8 | 78.5 | None | Moderate |
| 3[†] | 7.6 | 7/27 | 12/36 | 14 | 65 | 87 | 29.9 | 49.7 | 78.5 | 75.3 | None | Moderate |
| 4 | 7.0 | 10/15 | 12/18 | 5 | 52 | 78 | 27.4 | 54.9 | 85.2 | 78.5 | None | None |
| 5 | 7.0 | 6/17 | 10/25 | 12 | 51 | 74 | 33.1 | 54.9 | 82.4 | 82.3 | None | None |
| 6 | 6.6 | 7/18 | 8/24 | 7 | 53 | 78 | 24.9 | 54.9 | 75.1 | 77.3 | None | Normal |
| 7 | 6.0 | 3/13 | 10/34 | 28 | 28 | 91 | 24.9 | 54.9 | 76.8 | 82.3 | None | Normal |
| 8 | 5.9 | 9/22 | 11/26 | 6 | 31 | 78 | 24.9 | 54.9 | 78.2 | 77.3 | None | None |
| 9 | 5.8 | 6/25 | 8/26 | 3 | 37 | 86 | 38.8 | 70.9 | 81.0 | 80.6 | None | None |
| 10 | 5.7 | 3/8 | 8/15 | 12 | 38 | 76 | 27.4 | 54.9 | 83.8 | 77.3 | Moderate | Moderate |
| 11[†] | 5.0 | 3/3 | 10/20 | 24 | 22 | 75 | 24.9 | 56.2 | 73.8 | 78.5 | None | None |
| 12 | 4.9 | 6/11 | 9/17 | 9 | 65 | 88 | 30.1 | 56.2 | 71.3 | 76.0 | None | None |
| 13[†] | 4.7 | 8/33 | 20/36 | 15 | 67 | 86 | 50.1 | 59.4 | 75.1 | 78.5 | None | Normal |
| 14[†] | 4.6 | 18/18 | 9/31 | 4 | 60 | 82 | 34.3 | 59.4 | 68.8 | 78.5 | Moderate | Moderate |
| 15[†] | 4.0 | 13/28 | 12/30 | 1 | 36 | 85 | 26.2 | 58.2 | 74.9 | 81.6 | Moderate | Moderate |
| 16[†] | 3.3 | 15/30 | 18/31 | 4 | 42 | 94 | 26.2 | 58.2 | 74.9 | 78.5 | None | None |
| 17[†] | 3.2 | 18/24 | 18/28 | 4 | 58 | 88 | 27.6 | 57.4 | 79.3 | 79.5 | Normal | Normal |
| 18 | 3.2 | 14/24 | 16/29 | 7 | 62 | 81 | 24.9 | 43.8 | 69.9 | 76.0 | None | None |
| 19[†] | 3.1 | 11/28 | 10/30 | 1 | 59 | 54 | 35.6 | 57.4 | 79.3 | 79.5 | Normal | Normal |
| 20[‡] | 3.1 | 9/22 | 14/29 | 12 | 29 | 87 | 29.9 | 54.9 | 78.2 | 77.5 | None | Normal |
| 21[‡] | 3.1 | 10/24 | 16/31 | 13 | 29 | 87 | 29.9 | 54.9 | 78.2 | 77.5 | None | Normal |
| Mean ± SD | 5.3 ± 1.7 | | | 9.7 ± 7.2 | 46 ± 15 | 84 ± 6 | 30.5 ± 6.2 | 56.4 ± 4.9 | 77.7 ± 4.7 | 78.8 ± 2.2 | | |

* Dorsiflexion/plantar flexion; [†]bilateral TAAs; [‡]simultaneous bilateral TAAs; HH = hereditary hemochromatosis; TAA = total ankle arthroplasty; AOFAS = American Orthopaedic Foot and Ankle Society.

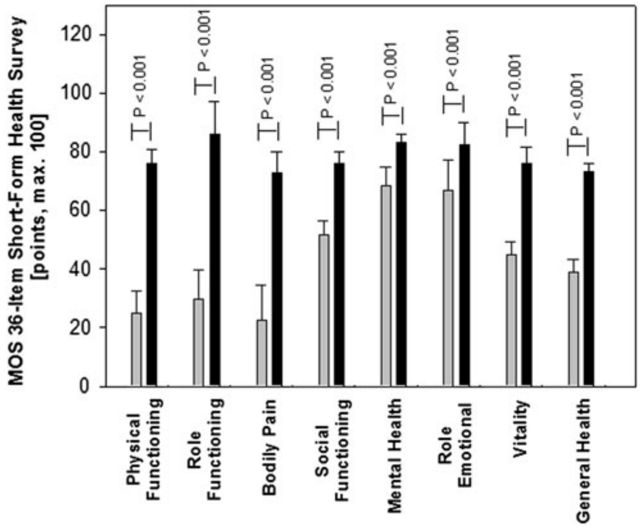


**Fig. 5** A graph shows preoperative (gray columns) and postoperative (black columns) quality of life for all patients assessed by the SF-36. Data are presented as mean ± SD.

should be evaluated at 10 years of followup for valid conclusions to be drawn regarding the long-term viability of TAA in patients with HH.

We observed a low rate of intraoperative complications. This low rate could be related to the surgeon's experience with the technique and implant design, as the senior author has performed more than 600 TAAs since 2001 using the HINTEGRA® implants. We found the cumulative incidence of revision for any reason was 19% (four ankles). These results are comparable to those reported in other studies [11, 36]. However, at the latest followup, both prosthesis components were radiographically stable, so that no component exchange was necessary during the study duration. Three patients (Cases 4, 10, 16) showed heterotopic, periarticular bone formation. However, these patients were asymptomatic and no excision of the ossifications was necessary. Heterotopic ossification, especially on the posteromedial side, is not uncommon and has been implicated as a cause of pain [3, 20, 37]. Valderrabano et al. [37] observed periarticular hypertrophic bone formation in 63% of patients undergoing implantation of the STAR prosthesis despite the use of an oral prophylaxis postoperatively. In 21% of cases, a secondary surgery (ossification removal and arthrolysis) was necessary [37]. As reported by Kim et al. [16], heterotopic ossifications were seen in 24% of patients who underwent implantation of an HINTEGRA® prosthesis, most commonly at the posterior aspect. However, they did not affect the VAS, AOFAS score, or ROM [16]. In the study by Wood and Deakin [40], reviewing results for 200 STAR replaced ankles, heterotopic bone formation appeared radiographically in 47% of the ankles. Additionally, there was no association between its presence and pain level [40].

It is well known pain is of particular relevance in the daily life of most patients with severe ankle osteoarthritis [9]. Joint pain also is one of the most common symptoms in patients with HH, with greater than 40% of patients reporting arthralgia [2, 24, 25, 30]. In our study, all patients experienced pain relief postoperatively, and 10 patients had a VAS score less than 2 at the latest followup.

The patients in our series all had symptomatic bilateral ankle arthropathy; bilateral procedures were performed in five patients. As more patients with HH have bilateral ankle arthropathy develop, therapeutic management may become challenging. Ankle fusion remains a valuable treatment option for end-stage osteoarthritis, providing pain relief, as seen at least in the short term [5]. However, short- and long-term problems for patients with ankle fusions have been described [19, 21, 22, 26]. One such problem is the development of degenerative changes in the adjacent joints [8]. Therefore, bilateral ankle fusion may not be the most suitable treatment option, given its detrimental influence on gait and functional ability of patients. In this study, all patients were treated by mobile-bearing TAA. Because of pain relief and preservation of ankle motion, four patients opted for contralateral ankle arthroplasty. In one patient, the bilateral TAAs were performed simultaneously. Recently, Karantana et al. [15] achieved postoperative pain relief and good function in five patients with simultaneous procedures using the STAR prosthesis. These results are comparable to our experience with 23 patients who underwent bilateral simultaneous TAAs in our clinic (unpublished data).

As expected, preoperative ROM was limited in most patients in our group. Postoperatively, ROM was increased in all patients. However, in 12 ankles, the ROM improvement was less than 10°. All patients reported the measured ROM was sufficient for their daily activities. With ankle arthroplasty, we do not necessarily expect an increased or physiologic ROM. The main goals are preservation of the functional arc motion, which is limited in patients with ankle fusion. These conclusions correspond with those of other reports addressing the results of ROM, where postoperatively measured ROM did not differ from preoperative findings [2, 15, 18, 40].

We observed pain relief and improved ROM after TAA in patients with HH. As most patients with HH have bilateral ankle osteoarthritis, it is our opinion TAA is superior to ankle fusion in these patients. However, we believe this type of surgery should be limited to foot and ankle surgeons with adequate experience in TAA. Long-term followup of additional patients is needed to confirm our findings.

**Acknowledgments** We thank Richard L. Amendola for assistance in preparing the paper, Heath B. Henninger PhD for helpful discussions, and Christine L. Abraham and Michael D. Harris for correcting the manuscript. We also thank the reviewers and editors (Richard A.

Brand MD and Paul A. Lotke MD) for their suggestions for strengthening the work.

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