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## Functional outcomes following ankle arthrodesis in males with haemophilia: analyses using the CDC's Universal Data Collection surveillance project

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### Summary

In persons with haemophilia (PWH), repeated ankle haemarthroses lead to pain, loss of joint range of motion (ROM), and limitations in activity and participation in society. PWH are offered ankle arthrodesis (AA) to eliminate pain. In our experience, PWH are hesitant to proceed to AA due to concerns regarding gait anomalies, functional decline and complete loss of ROM. The aim of this study was to report outcomes in ROM, assistive device (AD)/wheelchair use, activity scale and work/school absenteeism for participants in the CDC's Universal Data Collection surveillance project (UDC) pre- and post- AA. Males with haemophilia enrolled in the UDC with first report of AA (1998–2010) were selected. Descriptive statistics were calculated using data from the annual study visit pre-AA and the follow-up visit (~12–24 months) post-AA. The 68 subjects who fulfilled the criteria were: mean age 36.9 years (SD = 12.9); 85.3% white; 85.3% haemophilia A; 72% severe, 20.6% moderate; and 10.3% with inhibitor once during the study period. Mean loss in total arc of ankle motion was 17.02° (SD = 21.8,  $P = 0.01$ ) pre- compared to post-AA. For 61.8%, there was no change in use of AD for ambulation/mobility. For 85.3%, there was no change in use of a wheelchair. On a self-reported activity scale, 11.8% improved, 8.8% worsened and 79.4% did not change. Work/school absenteeism averaged 2.7 (SD = 6.4) pre- and 1.5 (SD = 6.4,  $P = 0.26$ )

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#### Author contributions

Heidi Lane and Azfar Siddiqi wrote the paper. Heidi Lane, Scott Ward and Azfar Siddiqi designed the research study. Scott Ward, Patricia Tobase and Robina Ingram-Rich contributed to authorship of the paper and assisted in designing the research study. Azfar Siddiqi performed the statistical analysis.

#### Disclosures

Heidi Lane has acted as a paid consultant to Bayer Pharmaceuticals, Inc. and Baxter Healthcare Corporation. Pattye Tobase has acted as a paid consultant to Bayer Pharmaceuticals, Inc. AEA Siddiqi, R Ingram-Rich, P Tobase, R. Scott Ward stated that they had no interests which might be perceived as posing a conflict or bias.

days per year post-AA. While ankle ROM was significantly reduced post-AA, for most subjects, there was no change in use of AD/wheelchair for ambulation/mobility. Physical activity was maintained and work/school absenteeism remained stable.

## Keywords

ankle arthrodesis; ankle fusion; arthropathy; haemophilia; outcomes; range of motion

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## Introduction

Bleeding into joints and muscles is the most common clinical manifestation of haemophilia, with approximately 80–85% of bleeding episodes occurring in the joints [1–3]; the ankle is more commonly affected in the first 5 years, while the knee and elbow are more commonly affected thereafter [3]. In a randomized controlled trial of prophylaxis vs. enhanced episodic factor replacement therapy, Manco-Johnson *et al.* [4] found 18 abnormal joints in 15 of 65 children enrolled, all under the age of 6 years. Of the 15 children, 2 were on prophylaxis and 13 were on the episodic treatment arm. Thirteen of the 18 joints found to be abnormal by MRI or X-ray, were in the ankle joints. Despite prophylaxis or aggressive factor replacement, we have observed that persons with haemophilia (PWH) continue to be at risk for joint arthropathy. Advanced stage haemophilic arthropathy, due to repeated joint haemarthroses, is characterized by pain, joint range of motion (ROM) loss, strength loss and deformity, culminating in loss of mobility [1–3].

Surgical ankle arthrodesis (AA) or ankle fusion to eliminate ROM at the fused joint has been the preferred procedure for painful end-stage haemophilic arthropathy (Grade IV – Modified Arnold-Hilgartner classification of haemophilic arthropathy) in this joint [5]. Reported indications for AA include severe pain, recurrent haemarthrosis, chronic synovitis, equinus contracture, extensive joint incongruence, or loss of ability to walk [6,7]. Despite reports of effective surgical outcomes in reducing pain, eliminating further haemarthroses and correcting deformity [6–9], in our clinical experience, PWH are reluctant to undergo AA, reporting fear of loss of all ankle motion and becoming more limited in physical activity. PWH often postpone the procedure until pain becomes incapacitating. End-stage haemophilic arthropathy often results in loss of quality of life and disability [3,10].

The literature contains few reports of functional outcomes related to AA in PWH. Existing studies are largely limited to case reports and primarily include outcomes related to surgical procedures such as successful joint fusion rates, infection rates, pain and joint haemarthroses recurrence [6–9].

The purpose of this study was to report selected outcomes available through the Center for Disease Control and Prevention's (CDC) Universal Data Collection (UDC) project in PWH who have undergone AA from 1998 to 2010. Surveillance data from the UDC were used to describe changes in ankle joint ROM and physical functioning [use of an assistive device (AD) and/or wheelchair for mobility, self-reported activity level and absenteeism from work/school] as a result of AA. Patient characteristics and joint infection were also reported.

Pain, an important AA outcome, was not collected in the UDC and therefore is not available for analysis.

## Materials and methods

From 1998 to 2010, data were collected at ~130 federally funded haemophilia treatment centres (HTCs) as part of the CDC-funded UDC. This project has been described elsewhere [11]. Each participating HTC and the CDC provided institutional review board oversight. Data were collected annually from participants, typically during comprehensive clinic visits.

A subset of UDC data fitting the inclusion and exclusion criteria was created for the analysis reported here. Male subjects with factor VIII or IX deficiency, who reported undergoing AA at least 1 year after enrolling in the UDC and had completed at least one UDC follow-up visit post-AA report, were included. This study was limited to subjects with first report of AA. Data were analysed from two annual UDC visits: (i) visit immediately prior to report of AA and (ii) the next follow-up visit post-AA (Fig. 1). This approach allowed for at least 9–12 months of recovery post-AA.

The initial report of AA, when first performed, was included in the analysis. In a few cases of bilateral AA, only information about the first AA was analysed.

## Measures

Outcomes of interest included change in measured ankle ROM and self-reported use of an AD (cane, crutches or walker), use of a wheelchair, activity level and number of days missed from work/school due to lower extremity problems. Presence of a physician-diagnosed joint infection in any joint in the year preceding follow-up visits was included.

Ankle dorsiflexion (DF) and plantarflexion (PF) ROM was measured, as described by Norkin and White [12], by a physical therapist (PT) or a trained medical provider using a standard goniometer and passively moving the ankle to its full extent, recording the measurement to the nearest degree. Using the same methods described by Norkin and White [12], Soucie *et al.* [13] found 5° difference among nine licensed PTs on a subset of 10 individuals. Ankle ROM values in otherwise healthy 20- to 44-year-old males were 12.7° (SD 11.6–13.8) DF and 54.6° (SD 53.2–56.0) PF [13].

Use of an AD or wheelchair since the last annual HTC visit was recorded as ‘never’, ‘intermittent’ or ‘always’. Self-reported activity level at time of HTC visit was recorded by presenting the patient with five choices, on a Likert-like scale, that best described their overall activity level and ranged from ‘requiring assistance for self-care’ representing ‘most limited’, to ‘unrestricted work/school and recreational activities’ representing ‘least limited’. In UDC, self-reported absenteeism from work/school was recorded as number of days missed since the last annual HTC visit. Physician-diagnosed joint infection, in any joint in the previous year, was recorded as a yes/no variable.

For analysis, categories were created within existing UDC variables: for use of an AD or wheelchair, ‘intermittent’ and ‘always’ categories were merged. This categorization improved statistical stability of the test statistics that were otherwise unstable and less

reliable. For example, cross-tabulations of pre- and post-AA wheelchair and AD usage as three level variables resulted in more than 50% of the cells having 'expected' values less than 5 that made the chi-square test questionable. Additionally, the stated categorization provided a two-group variable that reflected 'no usage' vs. 'any usage'. However, for interested readers, a description of movement of people to and from the 'intermittent' usage category is provided in the results for qualitative/clinical assessment. For the self-reported activity scale, a dichotomous variable, representing high and low activity levels, was created by merging the top two 'least' limited and the remaining three 'more' limited options respectively; for absenteeism from work/school since their previous HTC visit, mean number of days missed pre- and post-AA and 'none' vs. 'at least' 1 day missed were evaluated.

Covariates of interest included age at AA, race, ethnicity, body mass index (BMI), haemophilia type and severity, presence of factor replacement therapy inhibitor, and history of target joint (per UDC definition, 4 bleeds in a 6-month period) or invasive procedures performed on any other joint.

Subjects' age in years at AA was estimated by subtracting birth date from visit date of reported AA, with differences rounded to the highest complete year. Self-reported patient race was recorded per U.S. census categories, including Hispanic/non-Hispanic ethnicity.

Body mass index was calculated at each visit. Based on BMI, each subject was categorized according to the year 2000 CDC U.S. Growth Charts [14] as being obese, overweight or normal and defined as weighing above the 95th, within the 85th–95th, and below the 85th percentile for gender and age respectively. BMI data, collected at the time of follow-up visit post-AA report, were analysed in the multivariate model.

Haemophilia type was documented as either 'A' or 'B' depending on the deficient clotting factor (VIII or IX respectively). The site of the first bleed was recorded. Haemophilia severity was noted and expressed as a per cent of normally expected factor activity level and categorized as severe (<1%), moderate (1% to 5%) or mild (>5% to <50%).

Factor inhibitor antibodies were categorized as either positive (patient had at least one recorded inhibitor titre of 0.5 Bethesda units) or negative. Participants with no recorded antibody measurements were assumed to be negative.

We adjusted for the possible effect of previous history of, or current joint arthropathy, in our multivariate regression model by creating a dichotomous variable. Subjects who reported a target joint or any invasive orthopaedic procedure on any major joint other than the ankle (shoulder, elbow, hip or knee) at any annual visit before reporting AA were coded 'positive' with others coded 'negative'.

### Statistical analysis

Frequency distributions of select demographic and clinical variables at baseline were calculated. Ankle joint ROM measurements pre- and post-AA were summarized. ROM means were computed for PF, DF and the complete arc, and were calculated separately for

right and left ankles. Statistical tests of significance (paired *t*-test) were done to identify significant changes in mean ROM post-AA. Frequency distribution of changes in functional outcomes post-AA was performed. Because of small cell frequencies, statistical tests of significance were not done and the results are reported as frequencies. All analyses were performed in SAS<sup>®</sup> software version 9 (SAS Institute Inc., Cary, NC, USA). Generalized linear model (GLM) is a generalization of ordinary linear regression, proposed by Nelder and Wedderburn [15] that can accommodate response variables with non-normal distribution.

To give readers some sense of the independent associations between various characteristics of interest and the outcome; a single multivariate GLM without formal model building steps was used. We deliberately refrained from formal multivariate GLM modelling and analysis for several reasons – the total sample size was small ( $n = 51$ ); only one variable attained statistical significance in univariate analysis; and the outcome variable showed considerably large variations (overall mean change in ROM  $-17.02$ , SD 21.8). These factors would have resulted in unpredictable model building and an unstable multivariate model.

## Results

Between June 1998 and September 2010, 150 individuals with haemophilia who underwent AA were identified in UDC data. After excluding individuals who underwent AA prior to their first UDC visit and those who had not had at least 2 post-AA follow-up visits, 68 subjects were included.

The mean age at AA was 36.9 (SD 12.9) years. The majority (58, 85.3%) of the study subjects were white, with four (8.8%) reported as Hispanic. Fifty-eight (85.3%) had haemophilia A, 49 (72.1%) had severe at least once during the study period. Most individuals were diagnosed with haemophilia early in life, with 27 (39.7%) diagnosed prior to their first birthday (mean 14.9 months, SD 31.5 months). Most subjects experienced their first bleed at an early age (mean 18 months, SD 33.9 months). Table 1 includes the distribution of these and other demographic and clinical characteristics.

Statistically significant reductions in mean joint ROM post-AA for all measurements were observed. Overall, a reduction of  $17.02^\circ$  was observed in the complete arc of ankle motion post-AA, of which  $13.12^\circ$  (77%) of the total reduction was seen in PF and  $3.9^\circ$  (23%) in DF (Table 2). Post-AA, remaining PF and DF averaged  $14.57^\circ$  (SD = 15.01) and  $-1.63^\circ$  (SD = 7.17) respectively. Most observed reductions in ROM measurements were statistically significantly different from zero (Table 3).

Functional outcomes (AD use, wheelchair use, activity level, missed days from work/school) and reported occurrence of physician-diagnosed joint infection are summarized in Table 4. Forty-two (61.8%) individuals reported no change in use of an AD for mobility, 23 (55%) of which did not use an AD pre- or post-AA. Fifty-eight (85.3%) subjects reported no change in use of a wheelchair with 57 (98%, 90% of the total) reporting no use of a wheelchair pre- or post-AA. Analysis taking movements across the three categories of wheelchair/AD use showed that four ‘intermittent’ wheelchair users and one ‘always’

wheelchair user stopped using the wheelchair altogether. Another five individuals who did not use a wheelchair pre-AA started using it ‘intermittently’ post-AA. None of the non-wheelchair users moved to ‘always’ a use wheelchair user post-AA. A somewhat similar pattern was observed in terms of use of AD. Two ‘always’ and 11 ‘intermittent’ users stopped using ADs altogether post-AA and two ‘always’ AD users became ‘intermittent’ users. On the other hand, 13 non-AD users became intermittent users. None of the non-AD users moved to the always use category. Small numbers prevented evaluation by formal statistical tests of significance and the data are presented to allow readers qualitative/clinical assessment of the impact of AA on pre- and post-AA wheelchair/AD use.

Fifty-four subjects reported no change in activity level post-AA [8 (11.8%) improved and 6 (8.8%) declined]. Seventeen (35%) subjects who reported missing at least one work/school day pre-AA reported not missing any post-AA, and seven (10.3%) first missed work/school post-AA. Days missed averaged 2.7 (SD 6.4) before and 1.5 (6.4) post-AA ( $P = 0.3$ ). Two (2.9%) subjects experienced at least one joint infection (unspecified location) during the 2 years post-AA.

Bivariate comparisons were used to identify significant demographic or clinical characteristics (Table 5) associated with loss in joint ROM post-AA. These analyses showed no statistical significance except haemophilia severity, which was significantly associated with ROM loss. Age at assessment reached marginal significance ( $P = 0.06$ ). Formal multivariate analysis was not carried out for reasons stated earlier. However, to give the readers a glimpse of what a multivariate adjusted model from the data would look like, we evaluated all variables we considered biologically relevant (based on contemporary literature and authors’ experience) and those with a  $P$ -value of 0.02 or less in univariate assessment, in a single multivariate GLM model, without going through formal steps of multivariate model building. The multivariate model consisted of the following variables: age at AA, ethnicity, BMI, type and severity of haemophilia, presence of factor inhibitors and history of target joints or invasive orthopaedic procedures (Table 5). After adjusting for other variables in the model (stated above), individuals younger than 25 years experienced a larger reduction in joint ROM than subjects 35 year or older ( $-20.09^\circ$ , SD  $8.49^\circ$ ,  $P = 0.02$ ). However, overall, the age variable was only marginally statistically significant ( $P = 0.06$ ) and did not reach the cut-off of 0.05 for consideration as statistically significant. People with mild and moderate haemophilia experienced greater loss in joint ROM than people with severe haemophilia (mild  $-18.35^\circ$ , SD  $14.21$ ,  $P = 0.2$ ; moderate  $-20.82^\circ$ , SD  $7.73^\circ$ ,  $P = 0.01$ ). Haemophilia severity was also found to be statistically significant overall ( $P = 0.02$ ). No other statistically significant associations were found.

## Discussion

The 68 individuals with haemophilia studied had a mean age at first AA of 36.9 years, similar to previous reports (mean age range of 25–41.8 years) [6–9]. Overall, functional outcomes did not change. This should reassure patients about proceeding with AA when the indication for it is firm.

Ankle arthrodesis aims to eliminate ROM of the involved joint in painful end-stage haemophilic arthropathy. We expected subjects to exhibit loss in ROM, but not total loss as ankle ROM encompasses multiple joints. There was a significant reduction in mean joint ROM of the ankle, resulting in loss of arc of motion of 17.02° (23% decrease in DF; 77% decrease in PF). The type of AA and joints involved may influence changes in ROM [6–9]. Haemophilia-related pre- and post- AA ROM data in degrees have not been reported previously. In the non-haemophilia literature, Gellman *et al.* [16], via *in vitro* analysis, reported varying percentage losses in DF (50–62.8%) and PF (70.3–82.2%) depending on the type of AA. Data on the type of AA were not collected in the UDC. Single, double, and triple AA may have been included in the analysis and potentially contributed to the relatively large SD for ROM loss.

Age and severity of haemophilia were the only characteristics significantly associated with joint ROM loss following AA. The type of AA performed is probably the biggest factor in the amount of loss in ankle ROM in PWH and demographic or clinical characteristics most likely only minimally influence this change. Our finding that age and severity were significant factors in influencing change in ROM was somewhat surprising; however, data from previously published studies may provide an explanation. First, persons with more severe haemophilia, due to more severe joint disease, have greater limitations in ROM prior to arthrodesis [17]. Second, healthy individuals without haemophilia have decreased ankle joint ROM with increasing age [13]. In this study, individuals less than 25 years and/or with moderate haemophilia experienced greater ROM loss compared to those older than 35 years and/or with severe haemophilia respectively. Thus, prior to arthrodesis, younger and/or persons with less severe haemophilia had more available joint ROM that could be lost following surgery, as compared to older and/or persons with more severe haemophilia. The average total ROM recorded prior to AA was 39.3°, 33.0° and 25.21° for persons younger than 25, 25–34 and 35 years or older, respectively, and 50.0°, 43.55° and 24.30° for people with mild, moderate and severe haemophilia respectively.

Individuals in this study did not experience an overall change in AD or wheelchair use pre- and post-AA. The lack of overall change in AD use is not altogether surprising as PWH commonly have more than one joint involved. If the AD was used to address a problem with a joint other than the ankle, we expect no change. We anticipated our findings to be consistent with reports from the contemporary literature. Gamble *et al.* [6] reported wheelchair use in four of eight individuals pre-AA; three of four discontinued wheelchair use post-AA, and the fourth, due to severe pain in the knee, required continued use of the wheelchair. Panatopoulos *et al.* [7] reported on four individuals in whom walking improved and AD use became unnecessary in all individuals. The UDC does not link use of AD with a specific joint or disability. It is plausible the people started AD use post-AA for reasons unrelated to AA.

In this study, the average number of days missed from work/school, since the last annual visit, went from 2.7 to 1.5 days pre- vs. post-AA. This change represented a nearly 45% reduction in days missed post-AA. Due to the small sample size, the difference did not attain statistical significance. These results are consistent with observations within the authors'

respective institutions. If the ankle is the primary issue limiting participation in school/work, post-AA, this outcome is improved.

Two (2.9%) subjects experienced a physician-diagnosed infection in the 2-year follow-up period. Because the UDC does not collect data indicating the infected joint, the site is unspecified and a 0–2.9% rate of infection is possible for this cohort. In the haemophilia AA literature, during a mean follow-up of 2.7–9.4 years, an infection rate of 0–5% was reported [6–9,18].

We recognize the small sample size and thus the resultant low power as a limitation of this study. Nonetheless, given the relative scarcity of longitudinal data on PWH who have undergone AA, we consider our results informative. We applied a strict set of criteria in defining an analytic dataset from the larger UDC data to ensure inclusion only of individuals with complete and unambiguous information available. Therefore, while our results did not attain statistical significance, we expect the results reported here to be valid and unbiased. We were able to show some large differences that are considered significant in a clinical sense, despite being unable to attain the desired 0.05 *P*-value. Most subjects reported maintenance or improved functional outcomes. Despite consistent report of reduction in pain post-AA [6–9], lack of pain data is considered a limitation of this study, as it is the primary reason PWH consider AA. We caution our readers to consider the limitations stated earlier, when attempting to draw inferences from the multivariate model.

## Conclusion

Ankle arthrodesis resulted in significant ankle joint ROM loss, but not complete loss, for both DF and PF among these UDC subjects in the ~12–24 month period, post-AA. Persons under 25 years or with moderate haemophilia experienced the largest loss in arc of motion compared to persons over 35 years or with severe haemophilia. PWH commonly perceive AA as a debilitating procedure that results in functional decline. Despite loss in ankle ROM, the aim of AA, selected UDC functional outcomes were no worse post-AA. Though this study reports relatively short-term outcomes, the results may provide reassurance to PWH who are reluctant to undergo this procedure. Though pain was not included as an outcome in the UDC, previous studies [8,9] strongly support the efficacy of AA in reducing pain. It is likely that pain was also reduced in this cohort. In counselling PWH, the type of procedure, the presence of additional lower extremity haemophilic arthropathies, and patient-desired goals, all need to be considered when discussing individual patient expectations post-AA. The less than optimal statistical power and the short-term nature of the data in this study both point to the need for additional research on this subject.

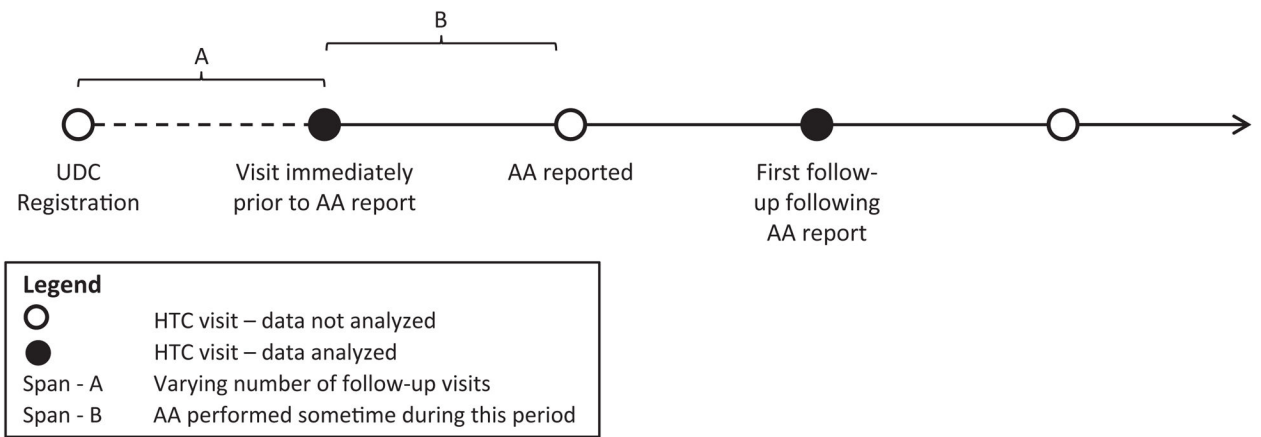
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**Fig. 1.** Graphic presentation of UDC visits timeline, highlighting time of ankle arthrodesis (AA) and the UDC visits selected for data analysis.

**Table 1**

Demographic and clinical characteristics of males with haemophilia undergoing ankle arthrodesis.

| Characteristic  | n(%)       |
|---|------------|
| Age at diagnosis (months)   |            |
| <1  | 27 (39.7)  |
| 1–6   | 13 (19.1)  |
| 6–11  | 10 (14.7)  |
| 12+   | 18 (26.5)  |
| Age at ankle arthrodesis (years)                                  |            |
| 24  | 15 (22.1)  |
| 25–34   | 13 (19.1)  |
| 35–44   | 20 (29.4)  |
| 45+   | 20 (29.4)  |
| Age in months at first bleed (n = 51)*                            |            |
| <1  | 14 (27.5)  |
| 1–6   | 7 (13.7)   |
| 6–11  | 12 (23.5)  |
| 12+   | 18 (35.3)  |
| Site of first bleed (n = 48)*                                     |            |
| Head (intra or extra cranial)                                     | 3 (<0.1)   |
| Oral mucosa   | 3 (<0.1)   |
| Circumcision  | 14 (0.2)   |
| Joint   | 8 (0.1)    |
| Others  | 20 (0.4)   |
| Race  |            |
| White   | 58 (85.2)  |
| All minorities  | 10 (14.8)  |
| Type of haemophilia   |            |
| Haemophilia A   | 58 (85.3)  |
| Haemophilia B   | 10 (14.7)  |
| Haemophilia severity  |            |
| Mild  | 5 (7.3)    |
| Moderate  | 14 (20.6)  |
| Severe  | 49 (72.1)  |
| Factor inhibitors (ever)  |            |
| Positive  | 7 (10.3)   |
| Negative  | 61 (89.7)  |
| History of target joints or an invasive procedure in other joints |            |
| Yes   | 46 (32.35) |
| No  | 22 (67.65) |
| Body mass index at arthrodesis                                    |            |
| Underweight   | 2 (2.9)    |

| Characteristic | n(%)      |
|----------------|-----------|
| Normal         | 20 (29.4) |
| Overweight     | 26 (38.2) |
| Obese          | 20 (29.4) |

\*  
n of <68 due to missing data.

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**Table 2**

Comparison of average range of motion (degrees) of the ankle joint pre- and post-ankle arthrodesis.

| Plane of motion | Before arthrodesis ( <i>n</i> = 51)* |                              | After arthrodesis ( <i>n</i> = 51)* |                              |
|-----------------|--------------------------------------|------------------------------|-------------------------------------|------------------------------|
|                 | Mean (STD)                           |                              | Mean (STD)                          |                              |
| Complete arc    | 29.96 (20.89)                        |                              | 12.94 (15.5)                        |                              |
| Dorsiflexion    | 2.27 (9.85)                          |                              | -1.63 (7.17)                        |                              |
| Plantarflexion  | 27.69 (17.6)                         |                              | 14.57 (15.01)                       |                              |
|                 | Left ankle ( <i>n</i> = 29)          | Right ankle ( <i>n</i> = 22) | Left ankle ( <i>n</i> = 29)         | Right ankle ( <i>n</i> = 22) |
| Complete arc    | 28.76 (19.45)                        | 31.55 (23.04)                | 16.93 (16.95)                       | 7.68 (11.73)                 |
| Dorsiflexion    | 0.28 (10.66)                         | 4.91 (8.17)                  | -2.83 (8.42)                        | -0.05 (4.84)                 |
| Plantarflexion  | 28.48 (18.07)                        | 26.64 (17.33)                | 19.76 (16.22)                       | 7.73 (9.96)                  |

\* Comparisons restricted to patient with complete set of measurements available (pre- and post-ankle arthrodesis).

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**Table 3**

Average change in range of motion (degrees) of the ankle joint pre- and post-ankle arthrodesis.

|         |                | Mean (SE) change in ROM | P-value* |
|---------|----------------|-------------------------|----------|
| Overall | Complete arc   | -17.02 (3.05)           | <0.01    |
|         | Dorsiflexion   | -3.9 (1.42)             | <0.01    |
|         | Plantarflexion | -13.12 (2.73)           | <0.01    |
| Left    | Complete arc   | -11.83 (3.34)           | <0.01    |
|         | Dorsiflexion   | -3.10 (2.12)            | 0.15     |
|         | Plantarflexion | -8.72 (3.45)            | 0.02     |
| Right   | Complete arc   | -23.86 (5.28)           | <0.01    |
|         | Dorsiflexion   | -4.95 (1.75)            | 0.01     |
|         | Plantarflexion | -18.91 (4.16)           | <0.01    |

\* Test that change is not different than zero.

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**Table 4**

Changes in physical functioning and joint infection outcomes post-ankle arthrodesis.

| <b>Outcome</b>                                  | <b>n(%)</b> |
|---|-------------|
| Use of assistive device for ambulation/mobility |             |
| Stopped using after fusion                      | 13 (19.1)   |
| Started using after fusion                      | 13 (19.1)   |
| No change                                       | 42 (61.8)   |
| Use of a wheelchair for mobility                |             |
| Stopped using after fusion                      | 5 (7.4)     |
| Started using after fusion                      | 5 (7.4)     |
| No change                                       | 58 (85.3)   |
| Activity level                                  |             |
| Improved after fusion                           | 8 (11.8)    |
| Got worse after fusion                          | 6 (8.8)     |
| No change                                       | 54 (79.4)   |
| Missed at least one day from work/school        |             |
| No days missed after fusion (improved)          | 17 (25)     |
| Missed for the first time after fusion (worse)  | 7 (10.3)    |
| No change                                       | 44 (64.7)   |
| Average number of work/school days missed       | Mean (SD)   |
| Before arthrodesis                              | 2.7 (6.4)   |
| After arthrodesis                               | 1.5 (6.4)   |
| Joint infections                                |             |
| Repeat experience after fusion                  | 0 (0.0)     |
| First experienced after fusion                  | 2 (2.9)     |
| No change                                       | 66 (97.1)   |

**Table 5**

Multivariate GLM analysis of the effect of characteristics of interest on the drop in overall range of motion on ankle joint post-ankle arthrodesis ( $F = 1.73$ ,  $P = 0.11$ ).

| Characteristic  | Parameter estimate | Standard error | <i>P</i> |
|---|--------------------|----------------|----------|
| Age at procedure  |                    |                |          |
| <25   | -20.09             | 8.49           | 0.02     |
| 25–34   | -8.95              | 7.32           | 0.23     |
| 35  | Reference          |                |          |
| Race  |                    |                |          |
| Minorities  | -2.68              | 8.96           | 0.77     |
| White   | Reference          |                |          |
| BMI   |                    |                |          |
| Obese   | -4.55              | 15.64          | 0.77     |
| Overweight  | -11.01             | 16.10          | 0.50     |
| Normal  | -7.11              | 16.47          | 0.67     |
| Underweight   | Reference          |                |          |
| Type of haemophilia   |                    |                |          |
| Haemophilia A   | -3.53              | 8.35           | 0.67     |
| Haemophilia B   | Reference          |                |          |
| Haemophilia severity  |                    |                |          |
| Moderate  | -20.82             | 7.73           | 0.01     |
| Mild  | -18.35             | 14.21          | 0.20     |
| Severe  | Reference          |                |          |
| Inhibitors  |                    |                |          |
| Positive  | -8.27              | 9.97           | 0.41     |
| Negative  | Reference          |                |          |
| History of target joints or invasive orthopaedic procedures |                    |                |          |
| No  | -1.46              | 7.02           | 0.84     |
| Yes   | Reference          |                |          |