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ORIGINAL STUDY

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Ankle arthrodesis in patients with haemophilia-associated ankle arthropathy - does the technique influence the outcome?

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Management of symptomatic osteoarthritis (OA) of the ankle in patients with haemophilia can be challenging. Arthroscopic ankle arthrodesis has been shown in non-haemophiliac patients to provide similar or superior rates of fusion to open ankle fusion. However, the literature regarding ankle arthrodesis in patients with haemophilia is limited. Our aim was to compare the rate of successful fusion between open and arthroscopic assisted ankle arthrodesis in patients with haemophilia. A retrospective study was performed. All patients with haemophilia who underwent ankle arthrodesis at our centre were included. Outcomes including peri- and post-operative complications, and lengths of stay were extracted from patients' records. Radiographs were reviewed for signs of successful arthrodesis.

Seventeen arthrodesis procedures were performed in 13 patients between 1980 and 2017. Nine procedures were performed arthroscopically and eight were open. Ten patients were diagnosed with haemophilia A and three with haemophilia B. The success rates of arthroscopic and open tibiotalar arthrodesis were 100% and 87.5% respectively. Four complications occurred. In the open technique group, there was one non-union. The same patient also developed subsequent haematoma after revision surgery. One patient developed a superficial wound infection which resolved with antibiotics. In the arthroscopic group, one patient developed a pseudoarthrosis of the distal tibiofibular joint which required a revision procedure. The results of this study suggest that arthroscopic ankle fusion for haemophilia-associated arthropathy is a viable option, with the rate of successful fusion being comparable to open procedures.

Keywords: Haemophilia; ankle arthrodesis

INTRODUCTION

Roughly two-thirds of patients with haemophilia have associated joint pain or arthropathy *(1, 2)*. The mechanism of arthropathy is thought to be the result of recurrent intra-articular bleeding, leading to cartilage degeneration secondary iron-catalysed

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free radical formation, as well as synovitis triggered by haemosiderin deposition *(3, 4)*.

Management of symptomatic arthropathy in haemophiliac patients consists of non-operative measures such as analgesia and physiotherapy, as well as surgical options including radiosynovectomy, arthroscopic or open debridement, arthrode¬sis and arthroplasty. The ankle is the most commonly involved joint in patients with haemophilia associated arthropathy *(5, 6)* however there is limited information on the optimal management of ankle arthropathy in this group of patients *(7)*.

For non-haemophilia related end-stage ankle arthropathy, arthrodesis remains the gold standard treatment despite improvements in ankle replacement8. This is traditionally performed as an open procedure, although arthroscopic ankle arthrodesis has increased in popularity in recent years *(9, 10)*. Proposed benefits of arthroscopic ankle arthrodesis include shorter intra-operative tourniquet time, reduced length of hospital stay, and improved validated functional ankle outcome scores at 2-year follow-up11. Radiographic rates of fusion are comparable to those observed with open arthrodesis *(11)*.

In patients with haemophilia associated ankle arthropathy (HAAA) however there is a paucity of information relating to clinical function or radiographic rates of union of arthrodesis performed with either technique. Tsailas and Wiedel reported encouraging rates of radiological union (95%) following open arthrodesis for end-stage ankle arthropathy in haemophilia patients *(12)*. Two case series have reported satisfactory outcome in ankle arthrodesis in haemophiliacs using arthroscopic techniques *(13, 14)*. Significant anatomic deformity of the ankle joint has been proposed to be a risk factor for non-union following arthrodesis *(15)*. It has been suggested that open arthrodesis allows better visualisation of the joint surfaces, and optimises the conditions for successful fusion, however the increased risks of bleeding and soft tissue injury associated with an open procedure may equally increase the risk of infection, delayed wound healing and reoperation *(9)*. To our knowledge there has been no previous study comparing open and arthroscopic ankle arthrodesis in haemophilia from a single centre.

The aim of this study was to report the outcome of patients with haemophilia who were managed by open or arthroscopic ankle fusion over a period of 30 years, to identify any significant variance in success or failure of fusion as well as significant post-operative complications.

MATERIALS AND METHODS

All patients with HAAA who underwent open or arthroscopic surgical fusion of the tibiotalar were included in this study. Tibiotalocalcaneal fusions were excluded from the study. Data on patient demographics, type and severity of haemophilia, surgical approach and surgical outcomes were extracted from chart review and Electronic Patient Records (EPR).

Radiographic images were retrieved from our Picture Archiving and Communications System (PACS) and reviewed by two orthopaedic surgeons. Signs of radiographic union were defined as presence of bridging trabeculae in at least two planes. Delayed union following ankle arthrodesis was defined as the lack of radiological signs of bone union at or beyond 3 months post-operatively, whilst non-union was defined as absence of radiological evidence of bone healing at 6 months or more.

The rate of successful fusion, peri- and post-operative complications, and the length of hospital stay were compared between open and arthroscopic techniques.

Statistical analysis was performed using the Mann Whitney U test using GraphPad software (Graphpad Prism, CA, USA).

Open procedures were performed either using an anterior approach to the ankle or a lateral approach if a fibula osteotomy was deemed to be required by the operating surgeon. Tibiotalar compression was achieved with at least two partially threaded screws (6.5mm) in both open and arthroscopic cases with screws placed from the medial aspect of the tibia into the talus. A third and/or fourth screw was inserted from the tibia into the talus depending on the surgeon's preference. Plate fixation was not used in any of the cases and fibula fixation was not undertaken. Bone graft augmentation was not required in any of the cases.

Patient received a pre and post-op management plan with regards to Haemophilia from our Haematology team. Plans were tailored according to each patient and varied according to severity of Haemophilia. In general, patients were given tranexamic acid commencing 7 days prior to surgery. They then received a bolus of the deficient factor 1 hour pre surgery and with subsequent factor replacement recommenced at 12 hours post surgery.

RESULTS

Two hundred and twenty eight patient with haemophilia A or B were identified. Of these 93 patients (41%) had ankle arthropathy and/or subtalar arthropathy. Seventeen ankle fusion procedures (16 primary and 1 revision) were performed in 13 male patients. Ten patients had haemophilia A (8 severe and 2 moderate) and 3 had haemophilia B (2 severe and 1 moderate). The mean age at the time of the primary fusion was 39 years (range 20 - 59). Table I shows the demographics of our patient cohort.

Nine procedures were carried out arthroscopically and 7 open primary procedures were carried out, one open ankle arthrodesis did not fuse and was later revised via another open procedure. The mean follow-up period was 7.6 years (range 0.5 – 33.4 years).

Mean tourniquet times were 89.5 minutes in the arthroscopic group (range 55 – 120 minutes) and 86.9 minutes in open procedure group (range 65 – 117 minutes) (p = 0.86). The mean length of stay (LOS) in the arthroscopic group was 3.6 days (range 3-5 days). Mean LOS in the open procedure group was 10.6 days (range 7-15 days) (p = 0.032).

Successful radiological arthrodesis was achieved by 3 months in 100% and 85.7% for arthroscopic and open tibiotalar arthrodesis respectively. There was a single case of revision tibiotalar arthrodesis procedure for painful delayed union at 6 months following an open procedure. Successful union occurred in this case at 3 months post revision surgery.

Complications were noted in 3 patients (Table II). In the open procedure group, one patient had painful delayed union which required revision surgery at 6 months. The same patient developed wound haematoma requiring surgical evacuation. Union was achieved and pain resolved after the revision procedure. There one case of superficial wound infection which was treated successfully with a course of oral antibiotics. One patient from the arthroscopic group developed a painful tibiofibular pseudoarthrosis. As this did not resolve with non-operative management, surgical resection of the pseudoarthrosis was undertaken.

DISCUSSION

Our results demonstrate that arthroscopic ankle fusion for end-stage haemophilia-associated arthropathy is at least comparable with, and probably superior to, the rate of successful fusion when performed as an open procedure. Furthermore, there appears to be a significant reduction in length of

Table I. — Demographics of patients who had arthroscopic and open arthrodesis, the rate of successful fusion, length of follow-up and number of complication

Group	Mean Age	Type of Haemophilia	Joint Operated	Rate of Fusion	Mean Length of Follow-up	Number of Complications encountered
Arthroscopic	38	Haemophilia A Severe: 4 Moderate: 2 Haemophilia B Severe: 1 Moderate: 1	Tibiotalar: 9 (Primary: 9)	Tibiotalar: 100%	1.6 years	1
Open	38	Haemophilia A Severe: 5 Haemophilia B Severe: 1	Tibiotalar: 8 (Primary: 7) (Revision: 1)	Tibiotalar: 87.5%	15.1 years	3

Table II. — Complications following arthroscopic and open tibiotalar arthrodesis

Patient	Method of fusion	Complication	Outcome
2	Left primary Open	Failed fusion	Revision surgery
	Left revision Open	Haematoma	Surgical evacuation
4	Left primary Open	Wound infection	Resolved with antibiotics
6	Left primary arthroscopic	Tibia/fibula pseudo-arthrosis	Tibia/fibula fusion surgery

hospital stay and no significant difference in duration of intra-operative tourniquet use.

There is a limited number of studies reporting arthroscopic ankle arthrodesis in haemophilia patients. Within an early case series of 10 patients who underwent arthroscopic ankle fusions, De Vriese reported on 1 patient with haemophilia *(16)*. More recently, Kats et al. described 4 haemophiliac patients within a case series of 15 patients in total who had undergone arthroscopic ankle fusion *(17)*. Tsukamoto et al. reported 3 ankle fusions using an arthroscopic technique in 2 hemophiliac patients *(14)*. A more recent study by Bai et al. was the largest patient set of haemophilia patients with ankle arthropathy managed by arthroscopic arthrodesis with a total of 10 patients *(13)*. All patients experienced successful fusion and there was minimal complications of postoperative pain in two patients *(13)*. To our knowledge, however, this is the first study comparing the results of open and arthroscopic ankle arthrodesis in the haemophiliacs from a single orthopaedic unit.

Combining existing evidence in the literature with our results, arthroscopic arthrodesis offers high successful fusion (100% in our cohort) which were superior to our cohort that underwent open ankle arthrodesis. These data also indicate that there is a comparable or lower complication rate. Infections may be less likely due to decreased amount of trauma to the surrounding soft tissue envelop, which may also enhance the rate of fusion and reduce bleeding complications.

The length of stay in hospital amongst the patients who had arthroscopic fusion was shorter, and may be attributable to an earlier return of clotting factor to therapeutic level. This could be accompanied by a reduced cost in patient management in arthroscopic versus open ankle arthrodesis as detailed by Peterson et al previously *(18)*, and particularly in the haemophilia setting, the majority of the cost can be related to the expense of clotting factor concentrates.

There are several limitations in this study, with small sample size in particular. However, as haemophilia is a group of uncommon genetic conditions, patient care requires significant amount multi-disciplinary input. At our unit, this involves extensive and careful pre-surgical planning and peri-operative management, followed by close post-operative monitoring and rehabilitation. These patients are seen pre-operatively in clinic, jointly by the orthopaedic surgeon and the haematologist who will provide support throughout the patients' admission. Orthopaedic procedure(s) is / are performed by a joint-specific consultant surgeon, while both the surgeon and the anaesthetist have had experience in the peri- and post-operative management of haemophiliac patients. During the rehabilitation period, their progress will be followed by the same physiotherapist(s), who also has / have previous experience working with these patients, on a regular basis. As such, a single centre study of a single joint procedure with a large sample size would not be feasible, without collaboration from a wider region.

We also acknowledge the lack of validated patient outcome scores used in this study. Unfortunately, as this is a case-control study, it was not possible to obtain such data retrospective from patients without introducing significant recall bias. However, aim of this study has been achieved, which was to identify and compare the success rate of fusion between the two operative techniques and associated risks and complications. Furthermore, all surgical procedures were performed by two surgeons, one of whom performed only open arthrodesis of the tibiotalar and joint, while the other performed both open and arthroscopic arthrodesis. However, both are experienced consultant foot and ankle surgeons, and utilised the same intra- and post-operative

approach for the open tibiotalar arthrodesis to minimise the difference in surgical outcome that may be attributable to their techniques.

CONCLUSION

While haemophilia is an uncommon condition, ankle arthropathy with associated morbidities is a common complaint in this population. Ankle arthrodesis remains the gold standard in the treatment of symptomatic, end-stage ankle arthropathy.

Despite the small patient population, our results, together with existing evidence in the literature, indicates that arthroscopic arthrodesis provides potential advantages in management to the haemophilia population with a rate of successful fusion comparable to open procedures, and minimal complications.

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