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A case of arthroscopic ankle arthrodesis for hemophilic arthropathy of the bilateral ankles

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ABSTRACT

INTRODUCTION: Hemophilic arthropathy can affect multiple joints including ankle. However, only one report has been published regarding both arthroscopic ankle arthrodesis with hemophilic arthropathy.
PRESENTATION OF CASE: The patient was a 23-year-old man with hemophilia A and a 3-year history of recurrent hemarthrosis in both ankles. We undertook surgery to treat arthropathy. His left ankle was treated first and the right ankle 6 months later. In both ankles, the cartilage was worn and eburnated. The remaining cartilage was removed and more dimples were created to fuse the tibia and talus. The ankle was fixed using 3 cannulated screws. Postoperatively, the patient wore an immobilization cast with no weight-bearing for 2 weeks. Thereafter, weight-bearing was allowed and the cast was removed 4 weeks after surgery.
DISCUSSION: At the 1-year follow-up, bony union was satisfactory, functional outcome was acceptable, and pain relief was good. The Japanese Society for Surgery of the Foot ankle-hindfoot scale score increased from 24 preoperatively to 87 postoperatively.
CONCLUSION: We report successful treatment with arthroscopic arthrodesis in a case of hemophilic arthropathy in both ankles.

1. Introduction

Hemophilia A is an inherited bleeding disorder caused by deficiency or absence of clotting factor VIII. It is an X-linked condition and mainly affects males, with a prevalence of 1 in 5000 [1,2]. It is characterized by spontaneous bleeding into the joints, muscles, and internal organs [3]. Hemarthrosis secondary to a clotting factor disorder is common [3]. The tibiotalar joint is often the first site involved, with onset of hemarthrosis when children start to walk [4–6]. The ankle is commonly affected in the second decade of life, and recurrent joint bleeding eventually results in end-stage hemophilic arthropathy [4–6].

Various surgical options are available to treat hemophilic ankle disease, including debridement, arthrodesis, and arthroplasty. The standard treatment for end-stage osteoarthritis of the ankle joint in hemophilic patients is joint fusion, which is associated with better long-term results than ankle arthroplasty [7,8]. The benefits of arthrodesis include pain reduction and elimination of hemarthrosis. Arthroscopic arthrodesis has several advantages over open arthrodesis, including smaller skin incision and less damage to soft tissue around the joint [9–15]. Although some reports have described arthroscopic arthrodesis for hemarthrosis of the ankle [16–20], we are aware of only one report on bilateral arthroscopic ankle arthrodesis for hemophilic arthropathy [21].

Here, we report a case of successful arthroscopic ankle arthrodesis in a young man with hemophilic arthropathy in both ankles. This has been reported in line with the SCARE criteria [22].

2. Presentation of case

The patient provided written informed consent for publication of this case report.

The patient was diagnosed with hemophilia 7 months after birth. He developed bilateral ankle pain and swelling at age 10 years and increasing pain in the left ankle at age 16, which was aggravated by prolonged standing. He was initially managed conservatively with analgesia and orthotics. He was referred to us at age 23. At that time, a subcutaneous injection of 2000 U of coagulation factor VIII (Cross Eight MC, Japan Blood Products Organization) was administered twice a week.

On presentation, he complained of grinding and clicking in the ankles, a minimal range of ankle motion, and severe pain when ambulating. Physical examination revealed swelling and tender-

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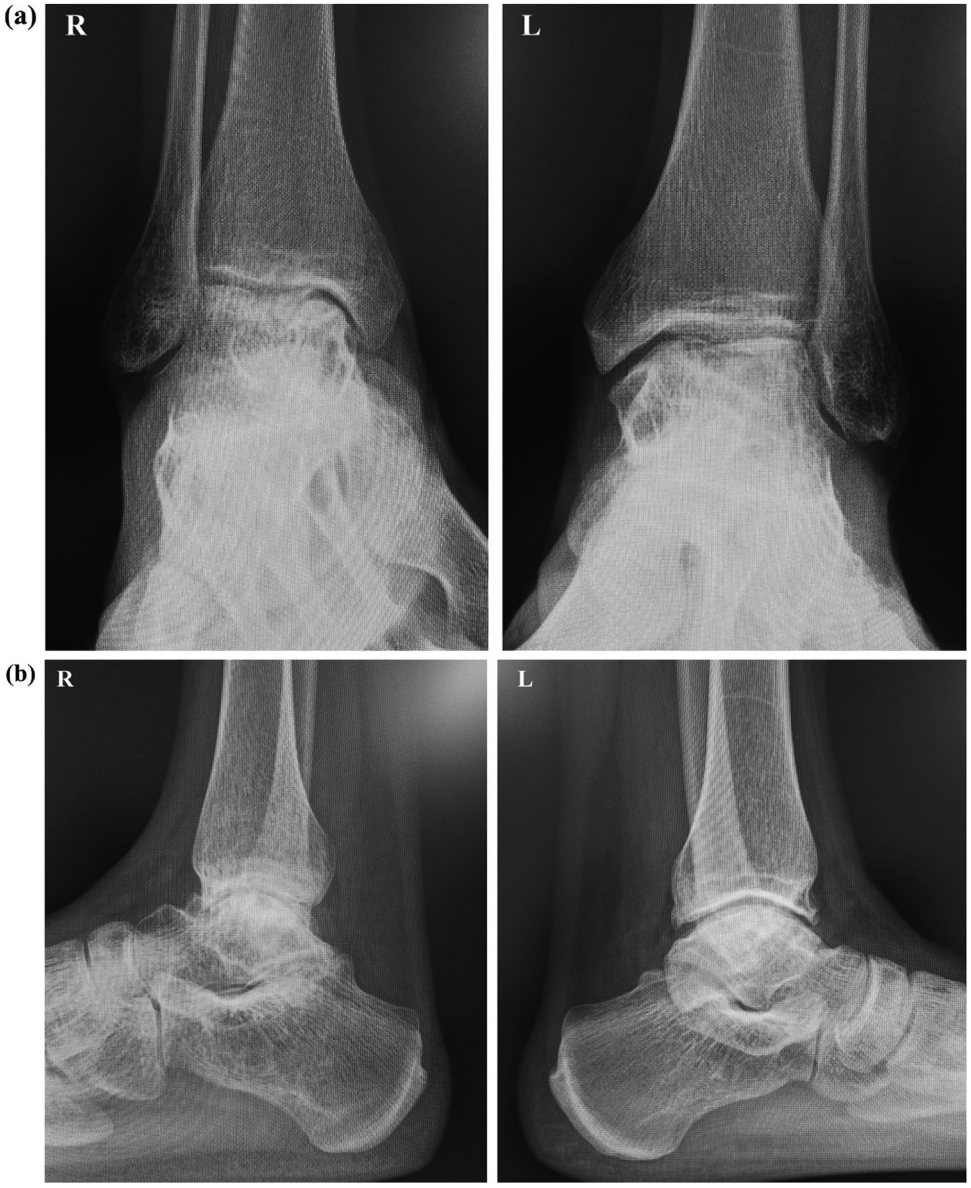

Fig. 1. Preoperative standing radiographs of both ankles showing advanced arthropathy with narrowing of the joint space, erosions of the articular margins, and incongruity of the joint surfaces in (a) anteroposterior view and (b) lateral view.

ness over both ankles and crepitus and pain with passive range of motion. His total range of motion at the ankle was <20° and maximum dorsiflexion was 5°. Weightbearing radiographs revealed narrowed joint spaces in weightbearing areas in both ankles (Fig. 1a, b), although ankle alignment was almost neutral. Computed tomography (CT) showed cystic regions in the weightbearing areas of the tibia and talus and an irregular articular surface (Fig. 2a, b). Given the long-term failure of conservative measures, we proposed surgical intervention. The plan was for ankle arthroscopic arthrodesis, which offers high fusion rate, outcomes comparable or superior to open procedures, and low complication rate. Preoperative and postoperative functional levels were assessed using the Japanese Society for Surgery of the Foot (JSSF) ankle-hindfoot scale score. Preoperative JFFS score was 24/100 (pain 0/40, function 14/50, alignment 0/10).

Ankle arthroscopy was performed by I.T. under traction via standard anteromedial and anterolateral portals. An abundance of scar tissue and hemorrhagic synovial tissue was encountered and removed manually. Cartilage remnants at the talus and tibia (Fig. 3a) were easily removed using a shaver and curette. The upper surface of the talus and lower surface of the tibial plafond were then abraded to remove sclerotic bone, creating dimples on the tibial and talar subchondral bone that were confirmed to bleed (Fig. 3b). After confirming all subchondral bone was removed, guidewires were inserted under fluoroscopic control through a small incision at the proximal aspect of the medial malleolus. Three cannulated 6.0-mm screws were placed to compress the tibia onto the talus. The ankle was fixed in the neutral position. Six months later, the same surgery was performed for the right ankle. Arthroscopic findings of the right ankle were very similar to those at the left ankle (Fig. 3a, b). The screws were inserted from the medial condyle of the distal tibia to the lateral process, neck, and back of the talus. The clotting factor level of at least 80% was maintained by a continuous infusion during ankle arthroscopic surgery and a few days after surgery. The ankle was immobilized in a below-the-knee cast for 4 months; partial weight-bearing was allowed 2 months after surgery. Bony union was confirmed 8 weeks after surgery.

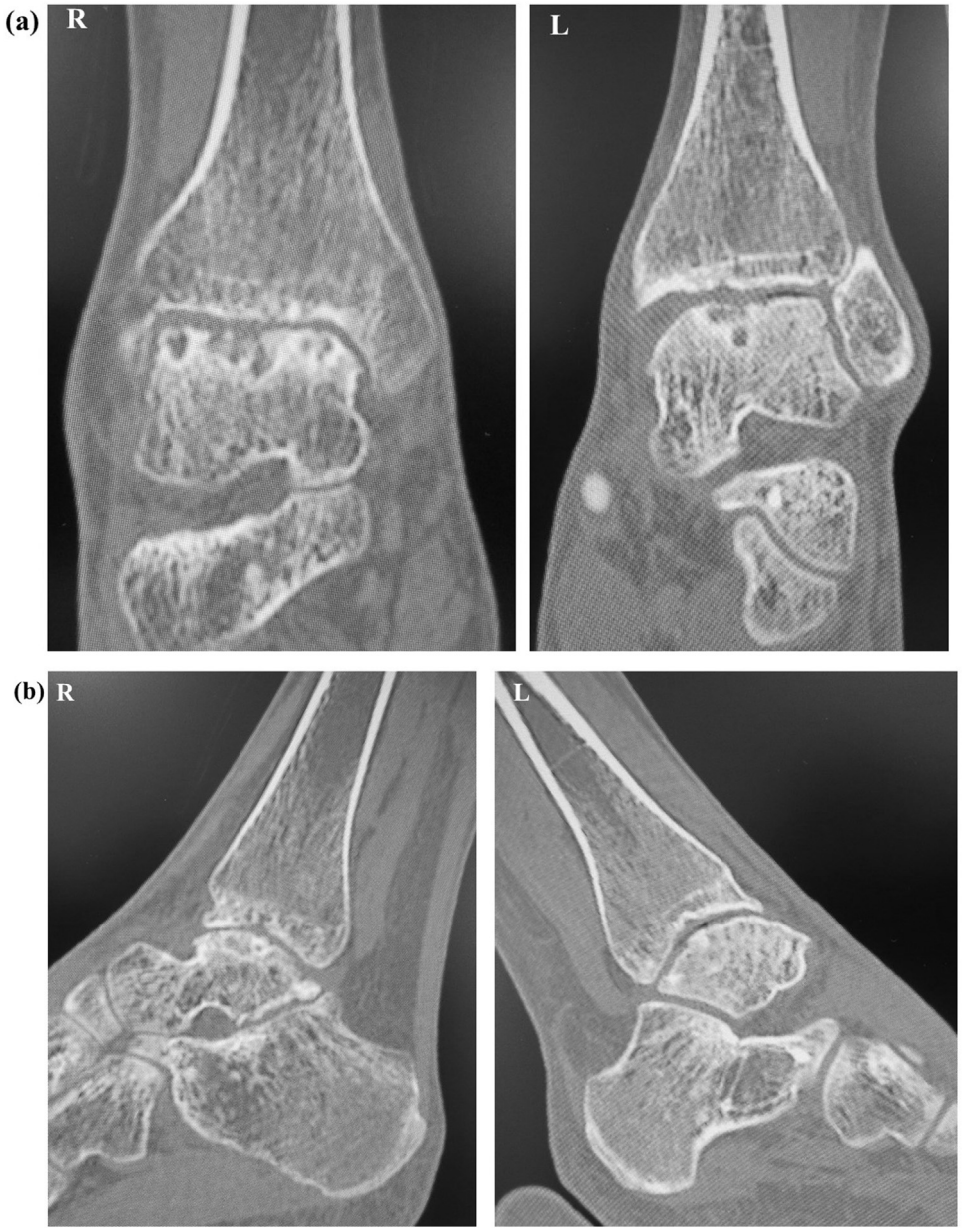

Fig. 2. Computed tomography shows cystic lesions and an irregular articular surface at both the tibia and talus on (a) coronal view and (b) sagittal view.

At the 1-year follow-up visit, bony union was adequate and there was no radiologic evidence of screw loosening, defined as the presence of bridging trabeculae across the arthrodesis (Fig. 4a, b). He is presently pain-free and working without limitations in daily activities but has limitations in recreation activity because both ankles were fixed. The JFFS score has improved to 87/100 (pain 30/40, function 37/50, alignment 10/10).

3. Discussion

We successfully treated a 23-year-old man with bilateral hemophilic ankle arthropathy by arthroscopic arthrodesis. Tsukamoto et al. [21] reported 3 cases of ankle fusion using an arthroscopic technique in 2 patients with hemophilia. Kats et al. [18] also described 15 cases of arthroscopic ankle arthrodesis, including 4 ankles in 4 patients with hemophilia. De Vriese et al. [17] reported 10 cases of arthroscopic ankle fusion, one in a patient with hemophilia. Gamble et al. reported 10 tibiotalar fusions in 8 patients using a variety of techniques; ankle fusion was performed for hemophilic arthropathy in 8 of these ankles [16]. Baker et al. reported 4 ankles in 4 patients with hemophilic arthropathy treated with arthroscopic arthrodesis of the tibiotalar articulation [20]. Finally, a series reported by Bai et al. included 10 patients with hemophilia A (10 ankle joints) who underwent arthroscopically-assisted ankle arthrodesis for end-stage arthritis [19]. However, there has been only one report on both arthroscopic ankle arthrodesis and hemophilic arthropathy [21], making our case rare.

Arthroscopy is an attractive method for achieving ankle fusion in hemophilic arthropathy. Arthroscopic arthrodesis is less invasive, requires less periosteal stripping, and preserves the normal contours of the ankle joint [23]. It is also associated with less postoperative pain and fewer complications such as blood loss, neurovascular problems, infection, and thrombosis [23,24]. Therefore, we selected arthroscopic ankle arthrodesis in this case. Glick et al. recommended ankle arthroscopic arthrodesis if the ankle defor-

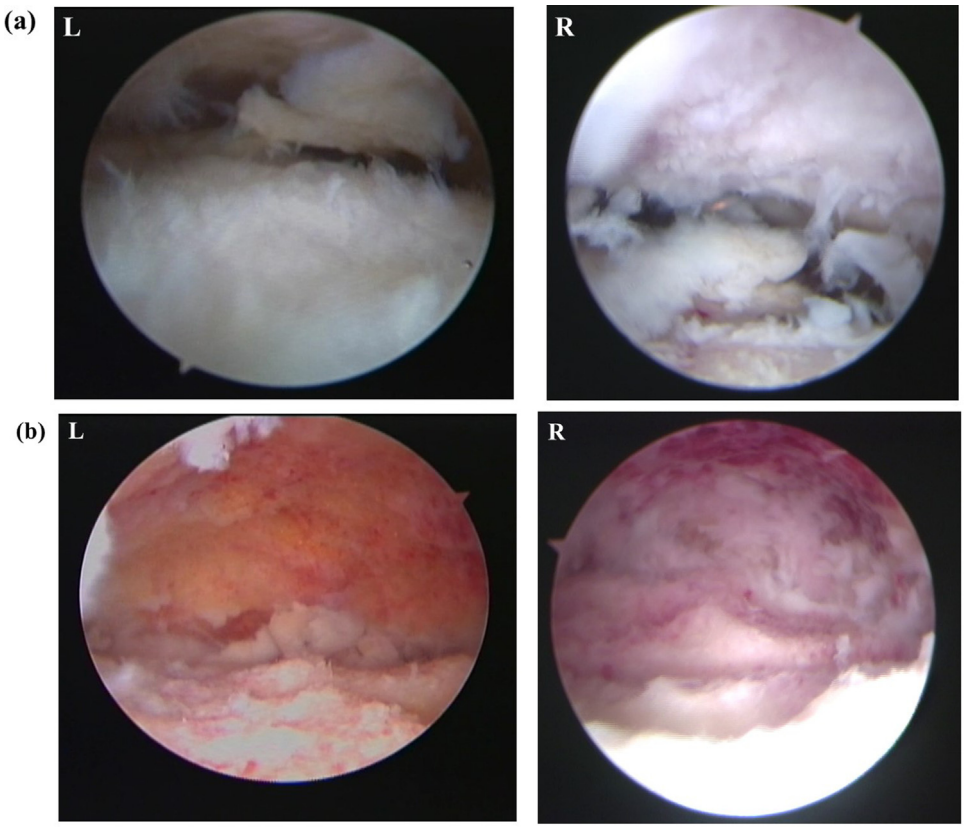

Fig. 3. Intraoperative findings show (a) peeling of the cartilage from the tibia and talus in both ankles and (b) bleeding from numerous dimples on the talus and tibial plafond.

mity was varus or valgus to <15° and <1 cm of anteroposterior translation of the talus [10]. In such cases, varus or valgus malalignment and anterior or posterior tibiotalar translation are usually mild, allowing ankle alignment to be corrected arthroscopically.

There were other advantages of arthroscopic ankle arthrodesis for hemophilic ankle arthropathy. Bai et al. reported that reduced time to union makes arthroscopic arthrodesis advantageous for patients with hemophilic arthropathy because this procedure decreases the need for clotting factors and reduces the cost of the surgery [19]. The decreased cost is an advantage to the use of arthroscopic arthrodesis compared to open procedure and is also worth considering with reference to the cost in hemophilia, like clotting factor therapy. In patients with hemophilia, a minimally invasive surgery allows decreased postoperative pain, less healing time, shorter hospitalization, and fewer doses of clotting factor [19].

However, this technique also has disadvantages. Unlike in primary osteoarthritis, the joint is rigid in hemophilic arthropathy, meaning that debridement of hyaline cartilage and subchondral bone is difficult to perform by arthroscopy. Also, as with any arthroscopic technique, arthroscopic ankle fusion is technically demanding and involves a significant learning curve [23]. In hemophilic arthropathy, little articular cartilage remains because of recurrent bleeding episodes, and a higher concentration of sclerotic bone in or adjacent to the subchondral cortex can be seen, which may contribute to the higher rate of nonunion and reinforces the importance of aggressive resection of subchondral sclerotic bone [12,17,21]. Thus, we aggressively resected the subchondral sclerotic bone in this case (Fig. 3a, b) and early bony union was achieved in both ankles.

There were other disadvantages of surgery for patients with hemophilic ankle arthropathy. The rate of postoperative infection in ankle arthroscopy in hemophilia is high compared to other joints [8], and Bai reported that a contributing factor might include the fact that the foot and ankle have a thin surrounding soft tissue envelope as a cause of the higher rate of postoperative infection [19], although we could not find any sign of postoperative infection in this patient.

Some patients with hemophilia require perioperative hemostatic management with supplementation of the deficient coagulation factor, depending on the extent of surgery and related treatments. According to the Japanese Society on Thrombosis and Hemostasis, if arthroscopic surgery is planned in a patient with congenital hemophilia, the target peak clotting factor level should be maintained at ≥100%, and an additional infusion should generally be continued intravenously to maintain a minimum factor level of ≥80% [25–27]. Regarding management of hemophilic patients underwent arthroscopic arthrodesis, according to Bai et al., preoperatively, regular prophylactic transfusion of clotting factor was administered to ensure that the concentration of clotting factor reached 80% activity or above, and postoperatively, the concentration of clotting factor was kept 60% for 3 days after surgery and above 30% for 1 week [19]. According to Baker et al., severe factor deficient patients were managed with a continuous infusion for 5 days with subsequent factor replacement given on alternate days for 2 weeks, and mild Factor-deficient patients were managed with bolus administration to keep levels over 90% for the 5 days following surgery [20]. Although the clotting factor level of at least 80% was maintained by a continuous infusion during ankle arthroscopic surgery and a few days after surgery in this case, strict management for bleeding might be necessary when opting for the open procedure.

The main limitation of this report is the short follow-up. Although arthrodesis often results in significant pain reduction, inactivity leads to disturbance of the normal gait cycle and accelerated destruction of adjacent articulations [28]. Compensatory

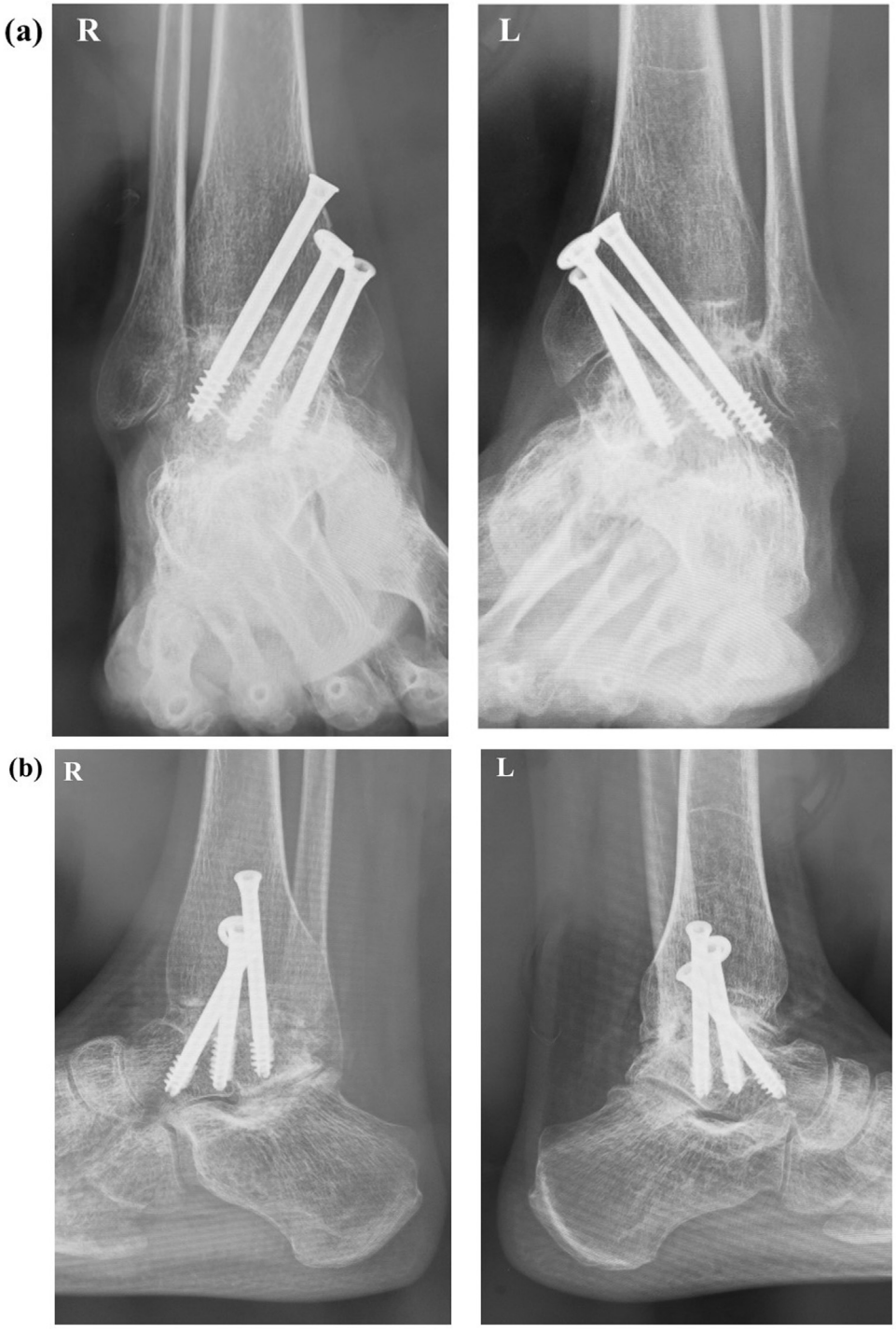

Fig. 4. Radiograph of the right ankle at 1 year after surgery shows bony fusion with complete obliteration of the joint space.

motion at the knee, subtalar, and Chopart joints leads to progressive erosive degeneration and arthritis. Although the patient had no ankle pain at the 1-year follow-up, longer term follow-up is necessary.

4. Conclusion

We have successfully treated a rare case of hemophilic arthropathy of both ankles by arthroscopic arthrodesis. In conclusion, we encountered a rare case of arthroscopic ankle arthrodesis for hemophilic arthropathy of the bilateral ankles successfully, suggesting arthroscopic arthrodesis was less invasive and effective even for hemophilic arthropathy of the bilateral ankles.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

A clinical case report is exempt from ethical approval in our institution.

Consent

A written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Ichiro Tonogai: data collection, writing the paper.
Koichi Sairyo: Interpretation.

Registration of research studies

None.

Guarantor

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Provenance and peer review

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