

# Total ankle arthroplasty (TAA) in a hemophiliac patient: case report

## Artroplastia total do tornozelo (ATT) em paciente hemofílico: relato de caso

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### ABSTRACT

Hemophilia is a genetically inherited disease linked to the X chromosome. The main alteration of this pathology affects the coagulation mechanisms. The most affected joints are the knees, elbows and ankles. Clinical manifestations include pain, joint bleeding and limited movement. The diagnosis takes into account the clinical picture and imaging tests. In this article, we present the case of a patient with restricted performance of activities of daily living and loss of quality of life due to hemophilic arthropathy in the right ankle. After total joint arthroplasty of the affected joint, there was improvement in the preoperative parameters of the patient.

**Level of Evidence V; Expert Opinion.**

**Keywords:** Ankle; Hemophilia; Hemarthrosis/surgery; Ankle replacement arthroplasty.

### RESUMO

A hemofilia é uma doença de herança genética ligada ao cromossomo X. A principal alteração dessa patologia afeta os mecanismos de coagulação. As articulações mais acometidas são os joelhos, cotovelos e tornozelos. As manifestações clínicas incluem dor, sangramento articular e limitação do movimento. O diagnóstico leva em consideração o quadro clínico e exames de imagem. Neste artigo, apresentamos o caso de um paciente com restrição das atividades diárias e perda da qualidade de vida devido à artropatia hemofílica no tornozelo direito. Após a realização da artroplastia total da articulação afetada, o paciente evoluiu com melhora dos parâmetros pré-operatórios.

**Nível de Evidência V; Opinião de Especialista.**

**Descritores:** Tornozelo; Hemofilia; Hemartrose/cirurgia; Artroplastia de substituição do tornozelo.

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### INTRODUCTION

Hemophilia is an X-linked recessive inheritance disease<sup>(1-4)</sup>. This disorder affects clotting factors and may manifest with spontaneous bleeding in the musculoskeletal system<sup>(2,3,5)</sup>. It is known that many cases may progress to

multiple-joint hemophilic arthropathy. The most affected joints are the knee, elbow and ankle<sup>(1)</sup>. The disease is diagnosed based on the clinical picture of the patient and imaging tests, which will also assist in the choice between conservative or surgical treatment<sup>(1,2,5,6)</sup>.

Work performed at Hospital de Clínicas da Universidade Federal do Paraná, Curitiba, PR, Brazil.

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There are few studies reporting the results and challenges related to total ankle arthroplasty (TAA) in hemophiliac patients because most studies address the hip and knee joints. Therefore, this case report aims to contribute to the improvement of TAA results in patients with hemophilia.

## CASE REPORT

This work was approved by the Ethics and Research Committee of the Hospital. The informed consent form was signed by the patient, according to the guidelines of the Declaration of Helsinki.

The patient (E.S.), a 40-year-old male who worked as a computer technician, sought the Orthopedics service because of a complaint of continuous and progressive pain in the right ankle. The patient has severe hemophilia A with a history of three other similar cases in the family. He used factor VIII for clotting and ibuprofen for pain control. On more than one occasion, the patient received corticosteroid infiltrations and radioactive isotopes of yttrium citrate in the right ankle (radiosynoviorthesis) without symptom relief. The patient had a history of previous surgeries: right ankle arthroscopy in 2010 and total right knee arthroplasty in 2011, both performed in our Orthopedics and Traumatology service.

With failure of conservative treatment, the patient was instructed to undergo right TAA. At the time, the patient had pain, joint swelling, crackling and limited range of mo-

tion in the right ankle. Figure 1 shows the preoperative X-rays of the right ankle in anteroposterior and lateral views, where a restriction of the joint space, osteophytosis and severe joint degeneration are observed.

A preoperative protocol was applied, with infusion of 4,000U of factor VIII and 2g of aminocaproic acid in the operating room<sup>(4)</sup>.

The surgery was performed with the patient in dorsal decubitus, under spinal anesthesia. Anterior universal surgical access of approximately 15 cm in the right ankle was used, followed by layer-by-layer dissection between the anterior tibial muscle and the long extensor muscle of the hallux with isolation of the neurovascular bundle.

Extramedullary guides were used for osteotomy of the distal tibia and talus. During surgery, tibial #1 and talar #1 components were used, as well as a 1/6 liner. After adequate placement of the prosthesis, layer-by-layer suturing was performed. A Zenith (Corin®) prosthesis was used.

In the immediate postoperative period, still in the surgical center, anteroposterior and lateral X-rays were taken of the patient's right ankle (Figure 2), showing proper placement of the tibial and talar components and liner.

During postoperative hospitalization, the patient used 2,000U factor VIII twice a day until the third day and then 2,000U per day until completing the seventh postoperative day<sup>(4)</sup>.



**Figure 1.** Preoperative X-ray of the right ankle. A: Anteroposterior view. B: Lateral view.  
**Source:** Author's personal archive.

In the present case, anamnesis, physical examination, the American Orthopedic Foot & Ankle Society (AOFAS) and 36-Item Short-Form Health Survey (SF-36) scales, and anteroposterior and lateral X-rays were used as parameters for the assessment and indication of the arthroplasty procedure. These resources evaluated joint function, quality of life, joint alignment and pre-operative joint conditions.

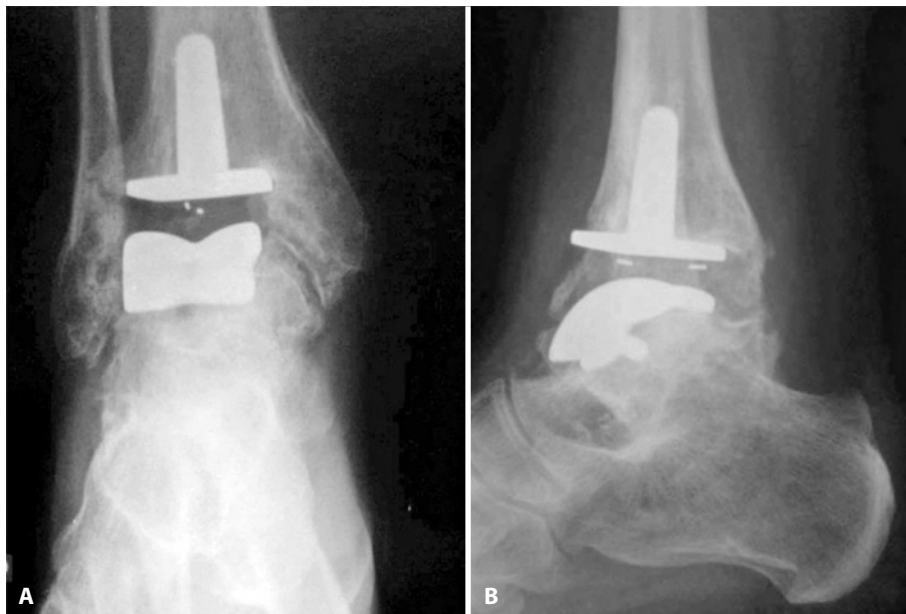
During the 3-year outpatient follow-up, the patient was assessed using the same criteria as in the preoperative period, plus static and dynamic computerized baropodometry.

In the clinical-functional and quality of life assessment before surgery, the patient scored 11 on the AOFAS scale and 83 on the SF-36 scale; after surgery, the scores obtained

were 87 and 93, respectively. Figure 3 shows anterior and posterior images of the hindfoot alignment (A, B) and the range of motion and degree of function (C, D).

Figure 4 shows the anteroposterior and lateral X-rays of the right ankle at 3 years follow-up, revealing good fixation and integration of the tibial and talar components as well as of the liner, with no signs of laxity.

The static baropodometric evaluation of the right foot (arthroplasty side) showed a plantigrade and neutral footprint with good load distribution. The plantar impression of the left foot (treated conservatively) showed a high instep. In the dynamic analysis of the right foot, a neutral footprint and a tendency toward supination on the left



**Figure 2.** Immediate postoperative X-ray of the right ankle. A: Anteroposterior view. B: Lateral view. **Source:** Author's personal archive.



**Figure 3.** Images from the late postoperative period (3 years). A: Anterior image of the ankle. B: Posterior image of the ankle. C: Plantar flexion in posterior view. D: Plantar flexion in lateral view. **Source:** Author's personal archive.

side were observed (Figure 5). Neither foot had calluses or ulcerations.

## DISCUSSION

Joint bleeding causes joint degeneration and stiffness and misalignment of the lower limbs, signs that are consistent with those reported in the literature<sup>(2)</sup>.

Conservative treatment includes analgesia, infusion of coagulation factors, physiotherapeutic rehabilitation and

chemical and radio synovectomy. In case of therapeutic failure, surgical procedures including synovectomy, arthroscopy, arthrodesis or joint arthroplasty are indicated<sup>(1-3,5,7-9)</sup>. This choice is difficult for the orthopedist because it is still a source of great controversy among specialists because it encompasses several factors.

Tibiotarsal arthrodesis was considered by many to be the gold standard surgical treatment for ankle arthritis until very recently. However, this treatment option may present worsening results over the years. One of the explanations is the overload involving adjacent joints that also evolve to painful arthrosis<sup>(8)</sup>. We believe, like other researchers, that arthroplasty may preserve or even improve joint mobility<sup>(5)</sup>.

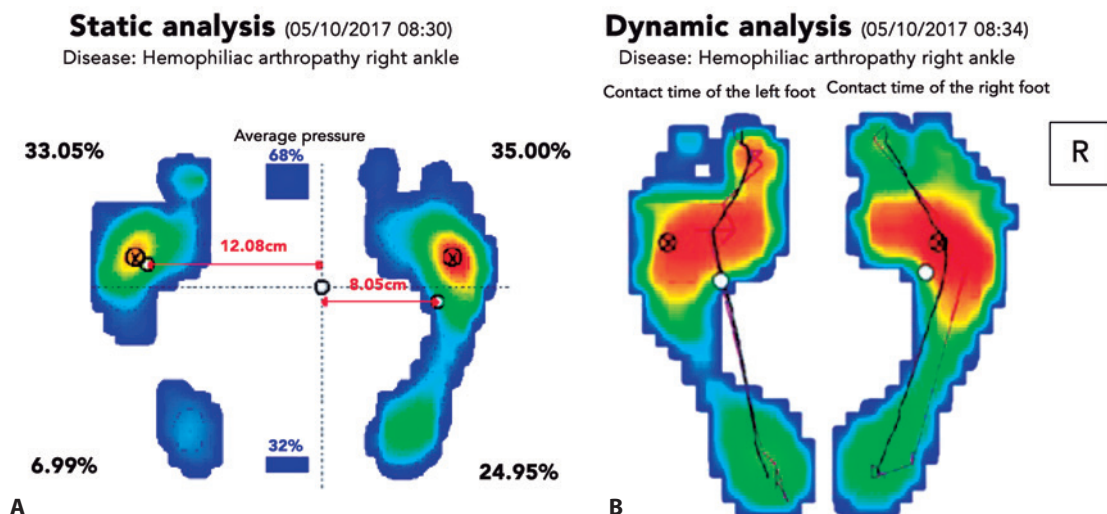
Some factors were essential in changing the perspectives related to the best management of these patients. Examples include the evolution of orthopedic materials and the patient's desire to maintain joint movement and perform early loading. These changes have altered the scenario, opening doors for total arthroplasty. Recent studies state that in adequately selected patients (good bone quality, non-practitioners of impact physical activities, with no history of previous or current infection in the surgical site), degrees of function and satisfaction are higher after TAA when compared with arthrodesis, after the application of the AOFAS and SF-36 questionnaires<sup>(7)</sup>.

The indication of TAA should be individualized, taking into account the patient's clinical context, refractoriness of alternative treatments, evolution of the arthropathy and deviations of the mechanical axis in the affected limb.



**Figure 4.** Three-year postoperative X-ray of the right ankle with load. A: Anteroposterior view. B: Lateral view.

**Source:** Author's personal archive.



**Figure 5.** Computerized baropodometry (2017). A: Static. B: Dynamic.

**Source:** Author's personal archive.

The main contraindications to TAA are acute or chronic local infections, unreparable instability, talus necrosis, advanced osteoporosis and patients with a high functional demand due to sports<sup>(8,10)</sup>. The patient did not present any of these conditions that could contraindicate the procedure.

Static and dynamic computerized baropodometry tests were performed 3 years after TAA in both feet, and neutrality of the plantar support was found in both the static and dynamic evaluations of the foot subjected to TAA. This supports the hypothesis that TAA can improve the mechanics of the foot as a whole<sup>(3)</sup>. Unfortunately, we found no studies reporting baropodometric parameters in hemophilic patients who underwent TAA.

The performance of TAA demands a considerable learning curve from the orthopedist<sup>(7)</sup>. It is also necessary to consider the occurrence of complications, the most feared being implant infection and surgical wound/suture dehiscence. In patients with hemophilia, the occurrence of infection varies from 5 to 15%, according to cohort studies<sup>(7,8)</sup>. To date, no complications have been observed during the patient's clinical evolution.

In the event of TAA failure, surgical revision of the arthroplasty or salvage procedures, such as ankle arthrodesis, can be performed<sup>(3)</sup>.

We stress once again that arthrodesis is still considered by many as the best option for the treatment of severe ankle arthritis. However, new studies involving TAA cases show good results, altering future perspectives<sup>(4)</sup>. Further multicenter studies are needed to define the ideal cases that would benefit from arthroplasty<sup>(1,5,9,10)</sup>.

## CONCLUSION

We observe in this case that the result of the TAA has been good to date. The patient reports great improvement in quality of life and remains asymptomatic after surgery. However, a longer outpatient follow-up period and a larger number of patients are necessary to evaluate the durability and efficacy of the ankle arthroplasty.

We emphasize that when the patient is carefully selected, joint replacement surgery may present good results.

**Authors' Contribution:** Each author made significant individual contributions to this manuscript: JLVS (<https://orcid.org/0000-0002-9038-2895>)\* participated in the review process and has conceived and planned the activities that led to the study; LRLP (<https://orcid.org/0000-0002-5114-566X>)\* participated in the review process and has approved the final version; ERN (<https://orcid.org/0000-0003-2151-0758>)\* has written the article, participated in the review process and interpreted the study results; MP (0000-0002-8144-0029)\* has written the article, has conducted the bibliographical research and reviewed the international literature. \*ORCID (Open Researcher and Contributor ID).

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