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Received: 2016.02.16 Accepted: 2016.05.01 Published: 2017.01.27		Perioperative Physiotherapy for Total Ankle Replacement in Patients with Inherited Bleeding Disorders: Outline of an Algorithm			
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		The treatment of end-stage hemophilic arthropathy c	of the ankle joint remains a controversial problem, and to-		

tal ankle replacement (TAR) is considered to be a valuable management option. Physiotherapy continues to be an extremely important part of TAR and has a tremendous impact on the outcomes of this procedure. Given the lack of data on the latter, this study details a protocol of perioperative physiotherapy in TAR in patients with inherited bleeding disorders (IBD).

The protocol outlined in this paper was devised via consultations within an interdisciplinary group, the authors' own experiences with TAR in hemophilic and non-hemophilic patients, previous reports on this issue in the literature, and patient opinions. Our working group followed the criteria of the International Classification of Functioning, Disability and Health.

The algorithm includes 4 physiotherapy phases with specified time frames, aims, interventions, and examples of exercises for each phase. We emphasize the importance of preoperative rehabilitation, and recommend introducing intensive physiotherapy immediately after the surgery, with regard to the wound protection and avoiding full weight-bearing in the first weeks. The intensity of physiotherapy should be adjusted individually depending on individual patient progress.

This study details a rehabilitation protocol for TAR in patients with IBDs, which can be equally applicable to clinicians and researchers. Further scientific studies are required to investigate the beneficial effect of different protocols as well as to clarify the effectiveness of various frequencies, durations, and intensities of selected interventions.

MeSH Keywords: Ankle Joint • Arthroplasty, Replacement, Ankle • Rehabilitation

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### Background

Inherited bleeding disorders (IBD) constitute a group of rarely occurring coagulation disturbances caused by blood clotting cascade dysfunctions and are characterized by spontaneous bleeding [1–5].

Based on the pathophysiology, the 3 most common IBDs are hemophilia A, hemophilia B (Christmas disease), and von Willebrand disease (VWD), whose intensity may range from mild to severe. The clinical course of hemophilia A depends on the plasma level of factor VIII (FVIII). Hemophilia B is caused by the deficiency or impaired function of coagulation factor IX (FIX). VWD is the most common IBD caused by the deficiency or dysfunction of von Willebrand factor, a plasma protein which mediates platelet hemostatic function and stabilizes FVIII. Defects of VWF can thus cause bleeding by impairing platelet adhesion or by reducing the concentration of FVIII. Severe forms of hemophilia (defined as <1% of normal factor activity; plasma level <1IU/dL) result in spontaneous bleeding into joints and muscles. Moderate hemophilia (1-5%; 1-5 IU/dL) is characterized by bleeding after minor injuries, with spontaneous bleeding episodes occurring rarely. In mild forms of hemophilia (5-50%; 5-50 IU/dL), an increased risk of bleeding is observed only in the case of injury or surgery. By contrast, VWD is usually divided into 3 types: Type 1 with a decreased level of VWF and a reduction in FVIII; Type 2, which is an abnormality of VWF itself; and the most severe, Type 3, characterized by a total lack of VWF and severe reduction of the FVIII plasma level [1–6].

### Discussion

The clinical course of IBDs is dominated by spontaneous bleeding episodes. Recurrent joint and muscle bleeding leads to irreversible destruction of the affected tissues. As a result, clinically symptomatic arthropathy with synovitis, cartilage deterioration, extensive bone destruction, and subsequent advanced disablement is observed in nearly all patients with severe forms of the disease. It should be noted that hemophilic arthropathy remains multi-articular in most cases and is the major cause of morbidity in hemophilia. In severe forms of hemophilia, the first musculoskeletal hemorrhage usually occurs at the age of 2 or 3 years and advanced disability may be present even at 20–30 years of age.

It was previously estimated that more than 90% of spontaneous bleeding episodes in hemophiliac patients affect the musculoskeletal system and 80% of these occur within joints. Almost 80% of all articular hemorrhages affect the knees, elbows, and ankle joints [1,7]. Windyga et al. [8] assessed the orthopedic status of 92 severe Polish hemophiliacs aged 20–30 years. In this population, physical examination revealed at least 1 affected ankle joint in about 85% of the patients, and radiological evaluation confirmed ankle joint involvement in over 90% of the patients. Ankle pain was reported by 43 patients (46.7%), and 47.8% of the patients experienced bleeds in the ankle joint in the 12-month period prior to the study.

The treatment of end-stage hemophilic ankle arthropathy remains a controversial problem and 2 surgical approaches – total ankle replacement (TAR) and ankle arthrodesis – have been proposed [9–11]. Despite numerous controversies, TAR continues to evolve with increasingly better outcomes and survival rates [12] and is considered to be a valuable alternative to ankle fusion for patients with hemophilic osteoarthritis. Recently, Barg et al. [10] performed a study to assess midterm results of 18 TARs performed in patients with VWD and found very encouraging outcomes. Asencio et al. [11] reported on a retrospective study of 21 patients with hemarthropathy who had undergone 32 TARs. The authors concluded that ankle arthroplasty is a promising alternative to arthrodesis in this group of patients.

We believe that physiotherapy (physical therapy, PT) remains an extremely important part of TAR and has a tremendous impact on the outcomes of this procedure. It is especially significant in the group of young hemophilic patients, who might expect normal activity with a pain-free ankle joint, functional independence, and the ability to participate in social activities without constraints. This emphasizes the importance of accurate and well-aimed rehabilitation in this population.

The main goal of this study was to outline and discuss an algorithm of PT procedures for TAR performed in patients with IBDs. In our opinion, the course of pre- and postoperative PT forms an integral part of total ankle arthroplasty and should be considered as important as the surgery itself. To the best of our knowledge, this is the first report of the protocol of a rehabilitation course for hemophilic patients undergoing TAR.

The protocol described in this paper applies to patients with IBDs undergoing TAR and was outlined at the Department of Orthopedic Surgery and Traumatology of the Central Clinical Hospital of the Ministry of Interior in Warsaw in cooperation with a convened multi-center working group. It was aimed at allowing the clinicians to improve the outcomes of TAR procedures in hemophilic patients and was devised via: 1) consultations within an interdisciplinary group (including orthopedic surgeons, physiotherapists, hematologists, and psychologists), 2) the authors' own experiences with TAR in hemophilic and non-hemophilic patients, 3) previous reports on this issue in the literature, and 4) patient opinion. Our working group followed the criteria of the International Classification of Functioning, Disability, and Health (ICF) [13–15], which serves

as a framework for planning the rehabilitation process as well as for the assessment of the structure, function, activity, and participation level.

The main aim of this article is to share our experience with regard to PT administered as an element of management of hemophilic patients undergoing TAR. As yet, there has been no discussion on this issue in the literature. We therefore decided to report on our approach in this matter and to propose an algorithm of a PT program for this difficult patient population. The outlined protocol is presented in the form of a Table 1 and supplemented by further remarks specified in the text below.

The importance of preoperative rehabilitation should be emphasized and strongly recommended since strength and movement amelioration in the ankle before surgery improves recovery after the implantation of the prosthesis and decreases the risk of postoperative stiffness.

We suggest introducing an intensive physiotherapy program no later than 2 weeks prior to the surgery, accompanied by appropriate factor replacement treatment [16]. We acknowledge that enforcing such a preparation phase may be problematic and difficult to organize, especially in the context of financial constraints faced by many health systems. Nevertheless, the benefits doubtlessly outweigh the demerits in that respect and we believe that exercises should be introduced as soon as possible.

The role of broad preoperative patient education should be emphasized. This population requires extensive information about the ongoing debate on TAR vs. ankle arthrodesis as well as the increased risk of various complications, such as intraarticular and periarticular bleeding, anaemia, development of an inhibitor, delayed wound healing, superficial and deep periprosthetic infections, and implant loosening, which are considered to be the most serious adverse events complicating joint arthroplasty procedures in hemophiliacs [17-19]. In addition, most individuals undergoing TAR in this group of patients are young and thus may require further operations in the future, which they should be informed about. The aim of reliable preoperative patient education is to prepare and motivate the patient for the demanding rehabilitation process and, above all, to teach the correctness of exercise performance. According to our observations, hemophilic patients are usually highly satisfied and appreciate ankle replacement procedures, but are more likely to accept possible postoperative deficiencies and rehabilitation difficulties if they are informed in a detailed manner before the operation. As a result, this approach reduces the feeling of fear and anxiety, which in turn increases trust and patient involvement in the rehabilitation process [20].

The main intent of PT during this preoperative period is to reduce pain and swelling, increase the range of motion in the ankle and other affected joints, and improve lower limb strength, flexibility, and balance, as is reflected in our regimen. We believe that in the case of significant preoperative pain, the protocol should be supplemented by local cryotherapy and physical procedures (e.g., magnetic field therapy) as they can significantly facilitate the results, thus improving the preoperative ankle joint condition [21-23]. It is sometimes recommended to exercise both legs simultaneously and activities improving general physical fitness may be taken into consideration in some cases. Crutch walking with partial weight-bearing, going up and down the stairs, and upper limb strengthening exercises should always be implemented. The intensity of exercises should be adjusted individually to achieve the best possible limb function before the surgery. In our opinion, such preoperative rehabilitation is not to be overestimated.

The rehabilitation process should be initiated immediately after the implantation of the prosthesis, primarily involving breathing exercises, isometric and synergistic contralateral exercises, lower limb elevation, and local cryotherapy in the early stage. This is aimed at preventing muscle atrophy, swelling, and thrombosis, and additionally reduces pain. The patients are routinely supplied with a posterior splint directly after the surgery, which allows for controlling edema and the wound healing process, and enables the early initiation of electrotherapy, if required. On the day of the intervention, cold therapy and limb elevation is typically implemented. Gradual patient verticalization and walking with crutches or a walking frame is advisable on the first postoperative day. In our protocol, we recommend absolutely no weight-bearing on the operated limb for 2 weeks after the surgery. Typically, an operated joint is immobilized for several weeks after the surgery, preferably in a controlled ankle motion (CAM) boot, which allows for performing exercises and ankle CPM machine application (if it is available). Because wound healing problems are grave complications of ankle replacement and may have perilous consequences, we suggest far-reaching caution and introducing the first joint exercises at about 2 weeks after the surgery, once the pain and swelling decreases and the wound has healed properly. At our department, we usually remove sutures on the 14<sup>th</sup>-16<sup>th</sup> postoperative day. From this point on, we recommend partial weight-bearing according to the pain tolerance. Mastering the correct gait pattern, with facilitation of each gait phase, is a very important element of rehabilitation that should be emphasized. Throughout the rehabilitation process, electrotherapy (e.g., magnetic field therapy) and dynamic taping may also be used, as specified in our protocol. Basically, assuming that the previous stages of rehabilitation were carried out properly and no significant complications occurred, full weight-bearing is allowed at 6 weeks after surgery; the rehabilitation should be continued for up to 12

500

# Table 1. A proposal of peri-operative physiotherapy guidelines for total ankle replacement in patients with inherited bleeding disorders.

Phase and aims	Interventions	Examples of exercises, dose
<ul> <li>Phase 1</li> <li>(2-4 weeks before surgery)</li> <li>Aims: <ul> <li>comprehensive patient education</li> <li>decreasing pain, inflammation and aedema</li> <li>improving range of motion</li> <li>muscles strengthening</li> <li>teaching patient walking with crutches</li> <li>improving general physical condition before surgery</li> </ul> </li> </ul>	<ul> <li>explaining the plan of physiotherapy and time scheadule of returning to work and daily activities</li> <li>teaching patient how to do exercises correctly after surgery</li> <li>walking with crutches on the floor and on the stairs with half weight bearing</li> <li>ankle range of motion exercises</li> <li>lower limb muscle strenghtening (around hip, knee and ankle joints)</li> <li>upper limb muscles and trunk muscles strenghtening</li> <li>proprioception and balance training</li> <li>local cryotherapy (20 minutes, several times a day)</li> </ul>	– exercises from phases 2 and 3
<ul> <li>Phase 2</li> <li>(0-2 weeks after surgery)</li> <li>Aims: <ul> <li>wound protection</li> <li>minimizing the consequences of immobilisation</li> <li>antithrombotic prophylaxis</li> <li>decreasing pain, inflammation and aedema</li> <li>patient verticalisation in 2<sup>nd</sup> day after surgery</li> <li>walking with crutches</li> </ul> </li> <li>* Patient should avoid weight bearing on operated limb in first two weeks after surgery</li> </ul>	<ul> <li>lower limb elevation</li> <li>breathing exercises</li> <li>synergistic contralateral exercises (nonoperated leg)</li> <li>training of proper gait pattern with crutches</li> <li>magnetic field therapy</li> <li>local cryotherapy</li> </ul>	<ul> <li>deep inspiration and slow expiration with upper limbs movements, 5 repetitions between exercises</li> <li>active ankle extension and flexion of opposite leg, 30 repetitions each direction</li> <li>active toes flexion and extension of operated limb, 30 repetitions each direction</li> <li>isometric contractions of gluteal muscles (hold 10 seconds, 30 times)</li> <li>gait phases facilitation</li> <li>magnetic field therapy 20 minutes</li> <li>application of an ice-pack (wrapped in a towel to protect the skin) for 20 minutes every 2 hours</li> </ul>
<ul> <li>Phase 3 <ul> <li>(2-6 weeks after surgery)</li> </ul> </li> <li>Aims: <ul> <li>wound protection</li> <li>minimizing the consequences of immobilisation</li> <li>antithrombotic prophylaxis</li> <li>decreasing pain, inflammation and aedema</li> <li>restoration of range of motion (without pain)</li> <li>muscles strengthening</li> <li>walking with crutches</li> </ul> </li> </ul>	<ul> <li>lower limb elevation</li> <li>breathing exercises</li> <li>passive and assistive exercises</li> <li>active exercises</li> <li>isometric contractions</li> <li>synergistic contralateral exercises</li> <li>lower limb muscle strenghtening (around hip, knee and ankle joints)</li> <li>training of each gait phase with partial weight bearing (regarding pain tolerance, 3<sup>rd</sup> week: 20% of body weight, 4<sup>th</sup> week: 40% of body weight, 5<sup>th</sup> week: 60% of body weight, 6<sup>th</sup> week 80% of body weight)</li> <li>proprioceptive neuromuscular facilitation techniques</li> <li>proper gait pattern training</li> <li>magnetic field therapy</li> <li>local cryotherapy</li> <li>dynamic taping</li> </ul>	<ul> <li>active knee flexion and extension against gravity (knee flexors and extensors strengthening)</li> <li>ankle range of motion (foot writing): 2 sets</li> <li>ankle flexion and extension with elastic stretch band (Theraband): 2 sets of 10 repetitions</li> <li>ankle inversion/eversion with elastic stretch band (Theraband): 2 sets of 10 repetitions</li> <li>ankle inversion/eversion with elastic</li> <li>stretch band (Theraband): 2 sets of 10 repetitions</li> <li>heel/toe walking: 2 set for 30 steps</li> <li>golf ball roll - 2 sets for 2 minutes</li> <li>towel gather (curls): 2 sets of 10 repetitions</li> <li>slant board stretch: 2 set of 10 (hold the stretch for 20 seconds and relax for 20 seconds)</li> <li>proprioceptive neuromuscular facilitation (PNF) techniqes for lower limb</li> </ul>

501

 Table 1 continued. A proposal of peri-operative physiotherapy guidelines for total ankle replacement in patients with inherited bleeding disorders.

Phase and aims	Interventions	Examples of exercises, dose
Phase 3 (continued)		<ul> <li>gait phases facilitation</li> <li>magnetic field therapy 20 minutes</li> <li>application of an ice-pack (wrapped in a towel to protect the skin) for 20 minutes every 2 hours</li> <li>lymphatic application of taping</li> </ul>
<ul> <li>Phase 4</li> <li>(6-12 weeks after surgery)</li> <li>Aims: <ul> <li>physiotherapy adjusted to individual patient's needs (depending on the previous condition and age)</li> <li>balance training</li> <li>improving general physical condition</li> <li>whole body strengtening</li> </ul> </li> </ul>	<ul> <li>continuation of interventions from phase 3</li> <li>resistive exercises</li> <li>coordination exercises</li> <li>proprioceptive neuromuscular facilitation techniques</li> <li>walking up and down stairs</li> <li>balance and proprioception training</li> <li>advanced strenghtening training</li> <li>endurance exercises</li> </ul>	<ul> <li>continuation of exercises from phase 3</li> <li>riding a stationary bicycle: 5–10 minutes</li> <li>ball pick-up: 2 sets of 10 repetitions</li> <li>calf raises: 3 sets of 10 raises (hold for 5 seconds)</li> <li>complex steps training, f.ex. over obstacles and in various directions</li> <li>walking on various surfaces</li> <li>single leg stance with open and closed eyes, with straight and bent knees: 3 times for 30 seconds</li> <li>one leg stance on trampoline, another leg has to touch to 4 objects: 5 minutes</li> <li>stepper training: 10 min</li> <li>tiltboard with knees straight and bent: 5 minutes</li> <li>standing on wobble cushion with various tasks for upper limbs and opposite lower limb: 5 minutes</li> <li>swimming: 10–30 minutes</li> </ul>

weeks after surgery in accordance with the patient's individual needs and expectations in order to regain the best possible physical function. We believe that partial weight-bearing and maintaining immobilization for 6 weeks is a reasonable approach facilitating bone ongrowth. It is advisable to remember the remark by Prusinowska et al. [24] that supination and pronation exercises should be started only when complete active motions of flexion and extension are restored, as failure to follow that principle may lead to the destabilization of the ankle joint and damage of the prosthesis fixing. It is particularly important to intensify PT after removing the walker boot and to implement balance and proprioception training during this rehabilitation phase. Of course, the patient should be precisely instructed about further stages of at-home rehabilitation and adequate precautions should be made. In general, we discourage all types of repetitive high-impact activities such as running. The use of an athletic brace should be considered for risky situations [25].

Precise control of the wound healing process and edema, particularly in the first postoperative period, is crucial to introduce the subsequent stages of rehabilitation without any problems. In our hospital, we apply intravenous administration of mannitol, which has not been hitherto proposed in the literature. According to our observations, it is a safe and effective way to control edema. We have never observed any type of complications associated with this treatment at our department; consequently, we believe it to be a recommendable treatment option.

At the very beginning, it should be noted that extensive PT is exceptionally desirable but difficult to perform in patients with IBDs because recurrent bleeding typically leads to massive destruction of several joints simultaneously, resulting in a major reduction of the range of motion of the affected joints and significant impairment of gait ability. Hemophilic patients scheduled to undergo TAR are generally young and would expect a considerable improvement after the surgery. Therefore, much effort has to be made to achieve satisfactory results.

Several concerns should be addressed before further discussion. Firstly, the intent of this protocol was to provide clinicians with a guideline of the PT course, and is by no means intended to be a substitute for clinical decision making. Each part of the devised protocol should therefore be adapted or supplemented based on physical findings and the patient's individual progress, if necessary. General state of health, course of the operation, wound healing process, acute hemarthrosis, personal commitment and motivation, to name only a few factors, should all be taken into consideration while making decisions. In case of multi-articular arthropathy, all affected joints should be included in the rehabilitation process and, generally, the progression to the next phase of PT should be based more on the clinical criteria than on the time frames established in this protocol. The rehabilitation process should take place in strict cooperation and under supervision of an orthopedic surgeon and hematologist, with adequate factor replacement therapy. Finally, the protocol refers to isolated total ankle arthroplasty without simultaneously performed ancillary procedures, which would usually demand significant modification and individualization of the rehabilitation regimen.

As stated above, after the TAR procedure, the operated joint is usually immediately immobilized with the recommendation of no weight-bearing for several weeks, but there is no consensus as to the optimal type and duration of such immobilization and the progression from non-weight-bearing to weight-bearing, and various management strategies have been proposed in the literature. In fact, the number of reports on this issue is very limited and they all refer to non-hemophiliac patient populations. In 2015, a discussion paper was published with the aim to present the opinions of a group of experts on the postoperative course for TAR [25]; in addition, papers by Barg et al. [26], Schipper et al. [27], Koczy et al. [28] and Asencio et al. [11] should be mentioned. A detailed description of approaches published in particular reports is far beyond the scope of this article, but it should be strongly emphasized that significant differences in the approach to the topic under discussion were demonstrated in particular papers and that currently there is no evidence to support any specific approach. At our department, patients are usually discharged home between the 5<sup>th</sup> and 7<sup>th</sup> postoperative day and on that day the posterior splint is typically changed to a CAM boot worn for 6 weeks. Partial and full weight-bearing is allowed in the 2<sup>nd</sup> and 6<sup>th</sup> week after the surgery, respectively, to avoid disrupting bone growth into the implant. In our opinion, special caution should be taken in this group of patients; however, this does not exclude the possibility of early introduction of the rehabilitation process. We believe our algorithm reconciles these issues.

According to the literature, impaired wound healing is the fourth most commonly reported TAR complication [29], and significant postoperative ankle and hindfoot edema can increase the risk of wound complications, including infection [30,31]. The approach should be therefore comprehensive, and effective pharmacotherapy should be always considered as a part of the treatment protocol in cases of worrying local condition. Consistently, in our protocol we propose for the first time the intravenous application of mannitol, which to be a safe and effective way of controlling edema in our clinical practice.

Based on previous reports, it can be assumed that a particularly difficult aspect of the rehabilitation process of patients with hemophilia following TAR would be increasing ROM. Barg et al. [1] reported on 8 hemophilic patients with 10 TARs followed up for an average of 5.6 years (range 2.7-7.6). In this study, the mean American Orthopedic Foot and Ankle Society's scale (AOFAS) score increased significantly, from 38 preoperatively to 81 postoperatively, the pain level decreased from 7.1 preoperatively to 0.8 postoperatively, and all domains of the SF-36 score showed significant improvement in the quality of life. Nevertheless, the improvement in terms of ROM was slight and not statistically significant. In turn, Strauss et al. [32] presented the results of 11 TAR in 10 hemophilic patients at a mean follow-up of 3.0 years (range 1.2-5.4). In their group, the mean AOFAS score improved significantly, from 21.5 to 68.0 points, the VAS score decreased significantly from 7.6 to 1.9 points, and ROM increased from 23.2 to 25.0 degrees, but without statistical significance. In addition, Ajis et al. [3] assessed 119 TARs of 3 different prosthetic designs performed in a non-hemophilic population and found no notable improvement in the ankle ROM at 6 months after total ankle arthroplasty. In our opinion, it is therefore appropriate to use such a postoperative rehabilitation program in which one of the basic techniques of intervention is used to improve the mobility of the treated joint.

The final point to be emphasized is that electrotherapy is usually overlooked when discussing different rehabilitation algorithms for TAR procedures and one might think that its role in this respect is either insufficient or undervalued. By contrast, electrotherapy is an integral part of our clinical practice and regimen prepared for hemophilic patients. The usefulness of various forms of electrotherapy was confirmed in earlier studies [21,33].

We believe TAR to be a valuable alternative to ankle fusion for advanced ankle arthropathy in patients with IBDs. According to our experience, it can alleviate the progressive pain and improve quality of life. Nevertheless, selected requirements have to be met to obtain satisfying results, including proper qualification process, precise surgical technique, multidisciplinary approach, sufficient treatment of pain and edema, appropriate factor replacement and, finally, well-thought-out, complex, consistently implemented, and individualized physiotherapeutic treatment.

Given the lack of data on the latter, we chose to initiate discussion and propose a rehabilitation algorithm. We believe that this protocol effectively addresses the issue and can be widely implemented into clinical practice because it was proved to be effective in our clinical practice. We acknowledge that further work is required in this area. As the effectiveness of the treatment will be higher if the most appropriate physical therapy is applied, we hope our remarks will provoke discussion and stimulate consensus for a standardized, well-aimed protocol, and that future studies will benefit from using our protocol.

### Conclusions

Preoperative and postoperative physiotherapy remains an integral part of TAR. It is related to superior functional outcomes, which is especially important in patients with IBDs suffering from disabling multi-articular arthropathy.

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This study details a rehabilitation protocol for TAR in patients with IBDs, which can be used by clinicians and researchers. Further scientific studies are required to investigate the beneficial effect of different protocols, as well as to clarify the effectiveness of various frequencies, durations, and intensities of selected interventions.

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504