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The role of physical activity in the rehabilitation of hemophilic arthropathy patients

Adina TRĂILĂ [1], Margit ŞERBAN [2], Andreea-Ancuţa VĂTĂMAN (TĂLÎNGĂ) [3,4], Ada-Maria CODREANU [3,4,5], Roxana-Ramona ONOFREI[6*], Oana SUCIU [7]

Abstract

Introduction. Hemophilia is a rare, x-linked inherited genetic disease. All types of hemophilia are characterised by a tendency to hemorrhage, which is sometimes of extreme, life-threatening severity in the absence of adequate treatment with clotting factor deficiency. Physical activity, regular training and sports help the hemophilia patient to develop basic motor skills such as coordination, flexibility, endurance and strength, keeping the musculoskeletal system in a good status. *Aim*.The present study aims to assess the outcomes of kinetotherapy and recreational sports in hemophilic arthropathy patients, using the Hemophilia Joint Health score (HJHS) and Functional Indepencence score. *Material and methods*. Thirteen male subjects aged between 13 and 61 years were selected and 23.1% were diagnosed with hemophilia B and 76.9% with hemophilia A. In our study group 38.46% of the patients received daily kinetotherapy sessions, and 61.54% of them received analgesic treatment with electrotherapy in addition to kinetotherapy. In the Medical Centre for Evaluation, Therapy, Specific Medical Education and Recovery for Children and Young People "Cristian Serban" in Buzias, patients included in this study performed different sports and recreational activities such as ping-pong and football. *Results.* The HJHS score significantly improved after therapy, from 53.07 ± 19.80 before treatment to 40.15 ± 17.37 after treatment ($p<0.0001$). The most affected score from HJHS was for the left knee flexion and extension loss. Only the score for extension have significantly improved ($p=0.04$). *Conclusions*. Incorporating physical activity and low-impact sports into the treatment plan of a hemophilia patient should be a balanced decision, taking into account the individual's specific condition, the severity of the disease, and their overall health profile.

***Key words**: physical activity, kinetotherapy, Hemophilia Joint Health Score, Functional Independence Score, hemophilic arthropathy*

Rezumat

Introducere. Hemofilia este o boală genetică rară, x-linkată. Toate tipurile de hemofilie se caracterizează printr-o tendinţă la hemoragie, care este uneori de o gravitate extremă, care pune viaţa în pericol, în absenţa unui tratament adecvat cu factori de coagulare. Activitatea fizică, antrenamentul regulat şi sportul ajută pacientul cu hemofilie să îşi dezvolte abilităţile motorii de bază, cum ar fi coordonarea, flexibilitatea, rezistenţa şi forţa, menţinând sistemul musculo-scheletic într-o stare bună. *Scop*. Studiul de faţă îşi propune să evalueze efectele kinetoterapiei şi ale sportului recreativ la pacienţii cu artropatie hemofilică,

[1]Hematologist, MD "Cristian Şerban" Buziaş Medical Centre for Evaluation, Therapy, Specific Medical Education and Recovery for Children and Young People, European Haemophilia Treatment Centre, Buzias, Romania

[2] Professor, MD PhD, "Louis Ţurcanu" Children's Emergency Clinical Hospital Timisoara, European Treatment Centre for Haemophilia, Romania

[3] PhD student, Doctoral School, "Victor Babeş" University of Medicine and Pharmacy, Timişoara, Romania

[4] PhD student, Department of Functional Sciences – Pathophysiology, Center for Translational Research and Systems Medicine, "Victor Babeş" University of Medicine and Pharmacy, Timişoara, Romania

[5] PhD student, Department of Medicine, Faculty of Medicine, „Vasile Goldis" Western University, Arad, Romania

[6*] Associate Professor, MD, PhD, Hab. Department of Rehabilitation, Physical Medicine and Rheumatology, Research Center for Assessment of Human Motion, Functionality and Disability, "Victor Babes" University of Medicine and Pharmacy Timisoara, Timisoara, Romania. Corresponding author: *e-mail:onofrei.roxana@umft.ro*

[7] Associate Professor, MD, PhD. Department of Rehabilitation, Physical Medicine and Rheumatology, Research Center for Assessment of Human Motion, Functionality and Disability "Victor Babes" University of Medicine and Pharmacy Timisoara, Timisoara, Romania

utilizând scorul Hemophilia Joint Health (HJHS) şi scorul Functional Indepencence. *Material şi metodă.* Au fost selectaţi 13 subiecţi de sex masculin cu vârste cuprinse între 13 şi 61 de ani, iar 23,1% au fost diagnosticaţi cu hemofilie B şi 76,9% cu hemofilie A. În grupul nostru de studiu, 38,46% dintre pacienţi au beneficiat de şedinţe zilnice de kinetoterapie, iar 61,54% dintre ei au primit tratament fizioterapeutic cu electroterapie antalgică pe lângă kinetoterapie. În cadrul Centrului Medical de Evaluare, Terapie, Educaţie Medicală Specifică şi Recuperare pentru Copii şi Tineri "Cristian Şerban" din Buziaş, pacienţii incluşi în studiu au desfăşurat diferite activităţi sportive şi recreative, precum ping-pong şi fotbal. *Rezultate.* Scorul HJHS s-a îmbunătăţit semnificativ după terapie, de la 53,07 ± 19,80 înainte de tratament la 40,15 ± 17,37 după tratament ($p<0,0001$). Cel mai afectat scor din HJHS a fost referitor la pierderea flexiei şi extensiei genunchiului stâng. Doar scorul pentru extensie s-a îmbunătăţit semnificativ ($p=0,04$). *Concluzii*. Participarea în programe de activitate fizică şi sporturi cu impact scăzut ca parte a tratamentului pacientului hemofilic trebuie să fie o decizie bazată pe profilul individual al pacientului, severitatea bolii.

Cuvinte cheie*: activitate fizică, kinetoterapie, scorul de sănătate articulară în hemofilie, scorul de independenţă funcţională în hemofilie, artropatia hemofilică*

Introduction

Hemophilia is a rare, x-linked inherited genetic disease. Worldwide, it is distributed regardless of geographic area or race, and is characterised by a deficiency of coagulation factors VIII, IX or XI. Depending on the type of coagulation factor affected, hemophilia is classified as hemophilia A (coagulation factor VIII deficiency), hemophilia B (impaired synthesis of factor IX) and hemophilia C or Christmas disease defined by quantitative or qualitative synthesis defect of coagulation factor XI (Iorio A. et al., 2019).

All types of hemophilia are characterised by a tendency to haemorrhage, which is sometimes of extreme, life-threatening severity in the absence of adequate treatment with clotting factor deficiency.

Manifestations are dependent on the severity of the disease, and residual clotting factor VIII or IX activity determines the frequency, severity and character of bleeding (White GC II et al., 2001).

The most common clinical manifestation of hemophilia is intraarticular haemorrhage or hemarthrosis. This occurs at the age of mobilisation, in the knees and ankles, and is caused by weight bearing, walking or running. In patients with hemophilia, pain is usually a sign of a hemorrhagic episode, preceded by local symptoms such as stinging, pressure, burning or pulsation. Repeated joint bleeding leads to synovial damage, initially acute, then chronic with degenerative effects on the articular cartilage and destructive effects on the subchondral bone tissue. Both inflammatory reactions induced by proinflammatory cytokines and cartilage degenerative changes triggered by erythrocyte degradation products are involved in the pathogenesis of hemophilic arthropathy (Hermans C. et al., 2011).

All these articular changes, along with pain, whether acute or chronic, affect the quality of life of patients with hemophilia, which is why a multidisciplinary team approached therapeutic and recreational physical activity methods in the management of hemophilia complications.

Hemophilia is a disease that, untreated, becomes a disability through the progression of hemophilic arthropathy (Timmer MA et al.,2015).

The introduction of prophylactic substitution has been an important moment in the prognosis of patients with hemophilia, allowing them to lead a normal life with the possibility of performing different types of sports. Prophylaxis is the standard of treatment for hemophilia in many countries, aiming to prevent bleeding, disabling arthropathy and life-threatening hemorrhages (Schrijvers LH. et al., 2013).

Recently, prophylaxis targets minimal levels to optimise treatment outcomes and allow the patient to lead a normal life (Oldenburg J. et al.,2015). The World Federation of Hemophilia recommends target levels for different situations, depending on the patient's needs and lifestyle, with optimisation of intervals between administrations and reduction of the annual bleeding rate (Blanchette VS et al., 2014). Moreover, prophylaxis allows patients with hemophilia to lead an active life, including participation in physical and social activities, similar to healthy people (Nilsson IM et al., 1992).

Thus, exercise and sport are an integrated part of hemophilia patient management. Physical activity, regular training and sports help the hemophilia patient to develop basic motor skills such as coordination, flexibility, endurance and strength, keeping the musculoskeletal system in a good status. The present study aims to assess the outcomes of kinetotherapy and recreational sports in hemophilic arthropathy patients, using the Hemophilia Joint Health score.

Materials and methods

Patients

The study included thirteen male patients, 10 patients with hemophilia A with coagulation factor VIII deficiency and three patients with haemophilia B respectively coagulation factor IX deficiency, admitted to the Medical Centre for Evaluation, Therapy, Specific Medical Education and Recovery for Children and Young People "Cristian Serban" in Buzias. The inclusion criteria were: a) age over 18 years; b) presence of hemophilia; c) history of hemophilic arthropathy. All patients received individualized physical therapy.

Study protocol was explained to all participants and those who fulfilled the inclusion criteria and agreed to participate in the study signed an informed consent. The study was conducted in accordance with the Declaration of Helsinki.

Assessments

Demographic data were collected, including age, weight and height. Clinical assessment was performed at the first visit, including anamnesis (history of disease diagnosis, duration of symptoms, prophylactic treatment received) and physical examination including assessment of posture and joint mobility, palpation and gait evaluation (Figure1-2).

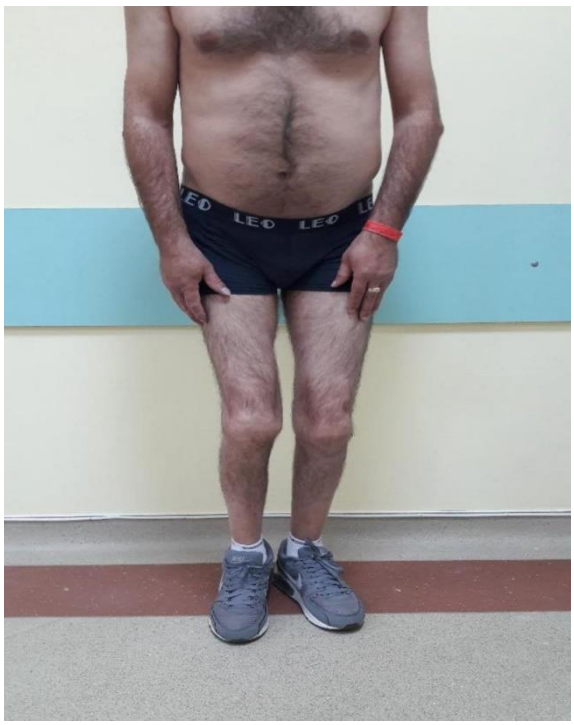
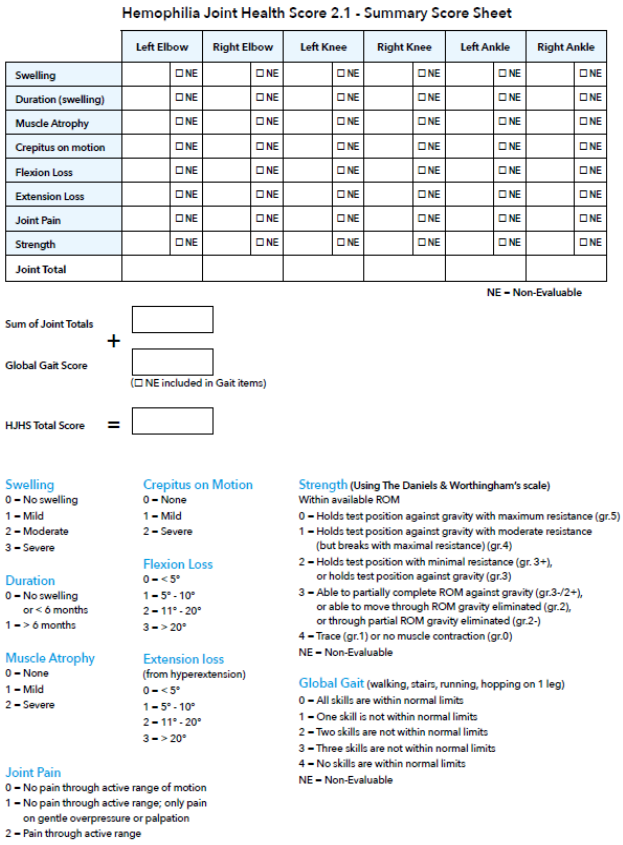


Figure 1. Postural assessment in a patient with hemophilic arthropathy (anterior view).

Figure 2. Postural assessment in a patient with hemophilic arthropathy (lateral view).

All patients included in this study were evaluated also with the Hemophilia Joint Health Score and the Functional Independence Score.

The Hemophilia Joint Health Score (HJHS), which is an assessment that takes an average of 45-60 minutes. This score is used to assess joint damage and monitor changes over time, particularly in response to treatment like prophylaxis or physiotherapy. It helps in the early detection of joint disease and in planning appropriate interventions (Feldman et al., 2011). The HJHS measures the impact of recurrent bleeding on the elbows, knees and ankles through 11 symptoms: swelling, duration of swelling, muscle atrophy, axial alignment, crepitus on movement, loss of flexion, instability, joint pain, strength and gait. Each of the 11 parameters was rated on a Likert-type scale from 0-1 to 0-4 (St-Louis J. et al., 2022). The scores from the individual parameters were summed to get a joint score, a gait score and a total score (Figure 3).



Hemophilia Joint Health Score 2.1 - Summary Score Sheet

	Left Elbow		Right Elbow		Left Knee		Right Knee		Left Ankle		Right Ankle	
Swelling		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Duration (swelling)		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Muscle Atrophy		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Crepitus on motion		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Flexion Loss		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Extension Loss		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Joint Pain		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Strength		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE		☐ NE
Joint Total												

NE = Non-Evaluable

Sum of Joint Totals +

Global Gait Score

(☐ NE included in Gait items)

HJHS Total Score =

Swelling
0 = No swelling
1 = Mild
2 = Moderate
3 = Severe

Duration
0 = No swelling or < 6 months
1 = > 6 months

Muscle Atrophy
0 = None
1 = Mild
2 = Severe

Joint Pain
0 = No pain through active range of motion
1 = No pain through active range; only pain on gentle overpressure or palpation
2 = Pain through active range

Crepitus on Motion
0 = None
1 = Mild
2 = Severe

Flexion Loss
0 = < 5°
1 = 5° - 10°
2 = 11° - 20°
3 = > 20°

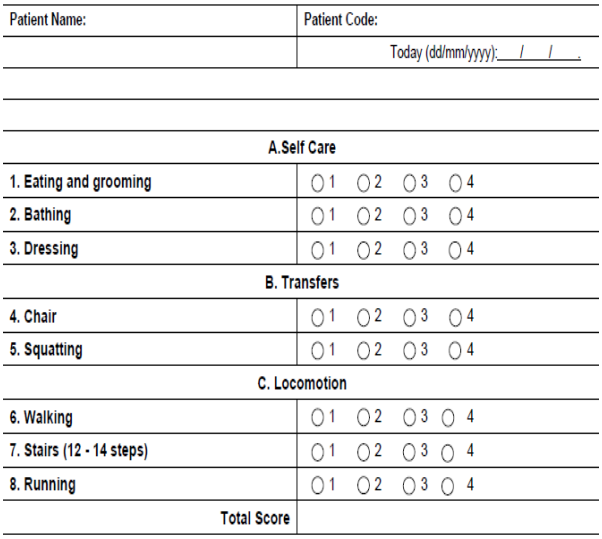
Extension loss
(from hyperextension)
0 = < 5°
1 = 5° - 10°
2 = 11° - 20°
3 = > 20°

Strength (Using The Daniels & Worthingham's scale)
Within available ROM
0 = Holds test position against gravity with maximum resistance (gr.5)
1 = Holds test position against gravity with moderate resistance (but breaks with maximal resistance) (gr.4)
2 = Holds test position with minimal resistance (gr. 3+), or holds test position against gravity (gr.3)
3 = Able to partially complete ROM against gravity (gr.3-/2+), or able to move through ROM gravity eliminated (gr.2), or through partial ROM gravity eliminated (gr.2-)
4 = Trace (gr.1) or no muscle contraction (gr.0)
NE = Non-Evaluable

Global Gait (walking, stairs, running, hopping on 1 leg)
0 = All skills are within normal limits
1 = One skill is not within normal limits
2 = Two skills are not within normal limits
3 = Three skills are not within normal limits
4 = No skills are within normal limits
NE = Non-Evaluable

Figure 3. Hemophilia Joint Health Score

Functional Independence Score was also used prior to physical therapy (Figure 4). Functional Independence in hemophilia is a score designed to measure a patient's autonomy in day-to-day activities such as grooming, eating, bathing and dressing, transfers and mobility like walking, climbing steps and running (Poonnoose PM et al., 2005).



Patient Name:	Patient Code:
	Today (dd/mm/yyyy): ___/___/___
A.Self Care	
1. Eating and grooming	○1 ○2 ○3 ○4
2. Bathing	○1 ○2 ○3 ○4
3. Dressing	○1 ○2 ○3 ○4
B. Transfers	
4. Chair	○1 ○2 ○3 ○4
5. Squatting	○1 ○2 ○3 ○4
C. Locomotion	
6. Walking	○1 ○2 ○3 ○ 4
7. Stairs (12 - 14 steps)	○1 ○2 ○3 ○ 4
8. Running	○1 ○2 ○3 ○ 4
Total Score	

Figure 4. Functional Independence Score in Hemophilia

Pain intensity was also quantified on the visual analogue scale (VAS) from 0 (no pain) to 100 (maximal pain) before and after physiotherapy programme.

All patients included in the study underwent a daily kinetotherapy protocol, which included warm-up exercises with 1 set of 8-10 repetitions, strengthening exercises with 1 set of 3 repetitions and gradually increasing to 5 repetitions, maintaining 10 seconds of contraction with 5 second gaps between each repetition, balance and proprioceptive exercises and cool-down containing 10 repetitions of 10 seconds of contraction with 5 second gaps between each repetition in the pain-free interval. Duration of the kinetotherapy program was 45 minutes. All patients also participated in recreational sports, like ping-pong and football.

Data were analyzed with MedCalc Statistical Software version 22.013 (MedCalc Software Ltd, Ostend, Belgium). Normal distributed data are presented as mean ± standard deviation and non-normal distributed data as median [interquartile range]. Paired t-test and Wilcoxon tests were used to compare data before and after treatment. A significance level of 0.05 was set.

Results

The study included 13 male subjects aged between 13 and 61 years who met the inclusion criteria and agreed to participate in this study. The mean age was

31.39 ± 13.28 years, all subjects were normal weight (BMI - 25.54 ± 5.32 kg/m^2; weight 75.6 ± 16 kg; height 172.3 ± 9.18 cm), and 23.1% were diagnosed with hemophilia B and 76.9% with hemophilia A. Patients characteristics are presented in Table 1.

Table 1. Patients characteristics

	Patients (n=13)
Age (years), mean±SD	31.39 ± 13.28
Weight (kg), mean±SD	75.6 ± 16
Height (cm), mean±SD	172.3 ± 9.18
BMI (kg/m^2), mean±SD	25.54 ± 5.32

The mean duration of rehabilitation therapy was 12.39± 4.47 days. In our study group 38.46% of the patients received daily kinetotherapy sessions, and 61.54% of them received analgesic treatment with electrotherapy in addition to kinetotherapy. Pain intensity was 43.85 ± 12.6 on the VAS scale at the beginning of the physical therapy session; later, after completing the procedures, it was significantly improved, with a decrease of VAS score to 16.15±7.68.

At the beginnning of the treatment, the FISH score for self care was 3.76 ± 0.43, while the score for locomotion was 2.69 ± 0.75. The total FISH score was 9.23 ± 1.83.

The HJHS score significantly improved after therapy, from 53.07 ± 19.80 before treatment to 40.15 ± 17.37 after treatment ($p<0.0001$). The most affected score from HJHS was for the left knee flexion and extension loss, with a value before treatment of 3 [1-3] for both motions. Only the score for extension have significantly improved 2 [1-2.25] ($p=0.04$).

Discussions

Hemophilia is a lifelong disease that predisposes to joint and muscle bleeding which, if left untreated, leads to disability and locomotor disability. When there is significant musculoskeletal dysfunction, activities that promote the development and maintenance of good bone density are recommended, as far as joint health allows. Each patient should select favourite physical activities that reflect physical condition and ability, local contexts and available resources (Iorio A. et al., 2010).

In our study, physical exercise was administered to all patients who met the inclusion criteria, and electrotherapy with analgesic effect was added to 61.54%. The role of physical exercise and its benefits, both physical and mental, have been known since ancient times. Today, regular physical exercise has found its usefulness for prevention, treatment and recovery in many diseases. Exercise has also been shown to improve bone density, reducing the risk of developing osteoporosis in patients with hemophilia, and it is well documented that sport can influence the dynamics of blood clotting factors. Individuals with hemophilia may be at increased risk of low bone mineral density compared to the general population due to risk factors including severity of hemophilia and hemophilic arthropathy and resulting immobility (Sossa Melo CL. et al., 2018). There are various ways to promote bone health that include regular exercise and optimal intake of calcium and vitamin D (Kempton CL. et al., 2015).

Gomis M et al. in his study revealed that physical activity is important in promoting normal neuromuscular development and fitness. Kinetotherapy aims to maintain muscle strength and functional capacity in patients with haemophilic arthropathy. The intensity of physical therapy should be gradually increased and adapted according to the availability of prophylaxis; but it should be less intense in patients with limited access to specific treatment (Gomis M et al., 2009).

In our study patients had a program based on strenghtening exercises, proprioceptive and balance exercises, avoiding high-impact exercises. Exercises programs were established according to the individual's health status, age, severity of joint damage, and hemophilia type. There are some studies which indicate that strength training with low weights and high repetitions can be beneficial. They increase stability providing better support and reducing the load on the joint (Konkle BA et al., 2009).

Strike K. and Co. demonstrated in their study of 233 men with haemophilia A and B, age between eight years to 49 years, that functional exercises such as treadmill walking for strengthening the knee flexors and extensors and partial weight-bearing exercises are more effective than static exercises. Other complementary physical therapy modalities such as

manual therapy, electrotherapy and hydrotherapy were also used in this study (Strike K et al., 2016).
Physiotherapy sessions on a regular base can help improve mobility, and reduce pain. A regular physical activity is becoming one of the most important outcomes for people with hemophilia (Wells AJ et al., 2020). Non-contact sports such as swimming, walking, jogging, badminton, cycling and ping-pong should be encouraged for patients with hemophilia (Mulder K et al., 2006). These exercises help maintain muscle strength and joint range of motion without exacerbating joint damage (Khair K. et al., 2012).
In the Medical Centre for Evaluation, Therapy, Specific Medical Education and Recovery for Children and Young People "Cristian Serban" in Buzias, patients included in the present study performed different sports and recreational activities such as ping-pong and football.

Figure 5. Patients with hemophilic arthropathy playing football

Figure 6. Patients with hemophilic arthropathy playing football

All patients included in the study and who participated in sports activities were carefully examined and evaluated using HJHS and FISH scores and underwent suitable prophylactic treatment.
Both the HJHS and FISH scores allow for more precise monitoring and management of joint health and functional abilities and contribute significantly to personalized kinetotherapy programs, aiming to improve quality of life and reduce the burden of disease in individuals with hemophilia (Srivastava A. et al., 2013).
Psychological effects during physical activity include a sense of well-being, improving confidence, promoting the release of endorphins and reducing anxiety and stress levels (Mackensen S. et al., 2016).
Meanwhile physiological aspects reffers to maintainig muscle and joint activity, training effect on the heart muscle, increasing collateral circulation and maintaing lower blood presure (Blamey G. et al., 2010).

Conclusions

While physical activity might initially seem risky for hemophilic patients, it actually offers several significant benefits. Physical activity and kinetotherapy plays an important role, offering benefits like maintaining joint function, muscle strength, and overall physical health.
Incorporating physical activity and low-impact sports into the treatment plan of a hemophilia patient should be a balanced decision, taking into account the individual's specific condition, the

severity of the disease, and their overall health profile.

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