Original Article



The influence of physiotherapeutic treatment on the functional independence of patients with hemophilia submitted to radioactive synovectomy

Influência do tratamento fisioterapêutico na independência funcional de pessoas com hemofilia submetidas à sinovectomia radioativa

Cecília do Socorro Sousa da Silva¹, Luzielma Macêdo Glória¹, Roberta Bentes de Melo²

ABSTRACT

Hemophilia is a recessive hereditary coagulopathy associated to the X chromosome that causes bleeding of varied severity, especially in the form of hemarthroses. If the hemarthroses are recurrent, they may cause chronic synovitis and consequently hemophilic arthropathy. Radioactive synovectomy (RS) is considered the treatment of choice for controlling chronic synovitis unresponsive to conservative treatment, and the physiotherapeutic treatment is recommended to improve functionality. Objective: To verify the influence of physiotherapeutic treatment on the functional independence and joint health of patients with hemophilia after treatment with RS. Method: This is a descriptive, retrospective, analytical study with qualitative and quantitative analysis methodology. A physiotherapeutic assessment was conducted. The HJHS scale and the FISH Score were applied to assess joint health and functional independence, respectively. The participants were divided into two groups according to whether or not they underwent physical therapy after RS. Results: Eight people with hemophilia A participated in the study. They were all male, with mean age of 19±5.3 years. There were 12 joints subjected to RS (41.67% elbows, 33.33% knees, and 25% ankles). When comparing both groups, there was no statistical differences between the groups considering either joint health or Functional Independence. Conclusion: This study is the first attempt to describe the impact of physiotherapy treatment on the functional independence and joint health of patients with hemophilia who undergo RS. It was possible to observe that the group that did not undergo physiotherapy had better joint health and better functional independence prior to RS compared to the group that underwent physiotherapy, however, the physiotherapy group presented worse overall condition, with functionality impacted by other joints and not just the one treated with RS, having wider number of target joints.

Keywords: Hemophilia A, Blood Coagulation Disorders, Synovectomy, Physical Therapy Specialty, Rehabilitation

RESUMO

A hemofilia é uma coagulopatia hereditária recessiva ligada ao cromossomo X, os sangramentos são de gravidade variável principalmente em forma de hemartroses, se recorrentes causam sinovite crônica e artropatia hemofílica. A sinovectomia radioativa (SR) é considerada o tratamento de eleição no controle da sinovite crônica não responsiva ao tratamento conservador, sendo recomendado tratamento fisioterapêutico para a melhora da funcionalidade. Objetivo: Verificar a influência do tratamento fisioterapêutico na independência funcional e saúde articular de hemofílicos após tratamento com SR. Método: Trata-se de um estudo descritivo, retrospectivo, analítico e metodologia quali-quantitativa. Realizou-se avaliação fisioterapêutica, aplicação do HJHS para avaliação da saúde articular e Escore FISH para avaliação da independência funcional. Os participantes foram subdivididos em dois grupos de acordo com a realização ou não da fisioterapia após a SR. Resultados: Participaram do estudo 8 pessoas com hemofilia A, sexo masculino, média de idade de 19±5,3 anos. Foram 12 articulações submetidas a SR, dessas 41,67% cotovelos, 33,33% joelhos e 25% tornozelos. Na comparação dos grupos, não houve diferença estatística entre os eles nas variáveis: saúde articular e a Independência Funcional. Conclusão: O estudo é uma primeira tentativa de descrever o impacto da fisioterapia na independência funcional e saúde articular de hemofílicos submetidos à SR. Embora possua limitações, foi possível observar que o grupo que não realizou fisioterapia apresentava melhor saúde articular e melhor independência funcional previamente à SR em comparação ao grupo que realizou fisioterapia; porém, o grupo fisioterapia apresentava pior quadro global, com a funcionalidade impactada por outras articulações e não somente aquela tratada com SR, apresentando maior número de articulações alvo.

Palavras-chaves: Hemofilia A, Transtornos da Coagulação Sanguínea, Sinovectomia, Especialidade de Fisioterapia, Reabilitação

¹Universidade do Estado do Pará – UEPA ²Fundação Centro de Hemoterapia e Hematologia do Pará – HEMOPA

Corresponding author

Cecília do Socorro Sousa da Silva E-mail: cecilia.silva21@yahoo.com

Conflicts of interest

None declared

Submitted: September 2nd, 2021 Accepted: September 27th, 2023

How to cite

Silva CSS, Glória LM, Melo RB. The influence of physiotherapeutic treatment on the functional independence of patients with hemophilia submitted to radioactive synovectomy. Acta Fisiátr. 2023;30(3):180-186.

DOI: 10.11606/issn.23170190.v30i3a181719

ISSN 2317-0190 | Copyright © 2023 | Acta Fisiátrica Instituto de Medicina Física e Reabilitação - HCFMUSP



This article is licensed under a license Creative Commons - Attribution 4.0 International

INTRODUCTION

Hemophilia is an X-linked recessive hereditary coagulopathy that results in a deficiency of clotting factors, such that hemophilia A regards the factor VIII deficiency and hemophilia B the factor IX. Considering severity, hemophilia is severe, moderate, or mild when the level of circulating factor is below 1%, between 1 and 5%, and between 5 and < 40% of the average level, respectively. 1,2

It is estimated that the disease affects around 1 to 2 of every 10,000 born males. In Brazil, there were 12.983 registered patients with hemophilia in blood banks in 2020, and 80% of these patients had hemophilia A and 20% had hemophilia $\rm B.^3$

Due to this deficiency of clotting factors, spontaneous or traumatic hemorrhages of various severity are the most frequent complications among patients with hemophilia. Hemorrhagic events occur mainly in the osteoarticular-muscular system in the form of hemarthrosis (joint bleeding) or hematoma (intramuscular bleeding). Generally, hemarthrosis is the most common clinical characteristic of hemophilia, affecting around 80% of patients at the large synovial joints, especially the knees, elbows, and ankles. A

The presence of blood in the joint cavity is always harmful, as it leads to intra-articular iron deposition, causing the proliferation of synovial and vascular cells leading to chronic synovitis, a change in the synovial membrane. Consequently, chronic synovitis stimulates new intra-articular hemorrhages, generating more and more lesions due to iron deposition in a vicious circle of hemarthrosis-synovitis-hemarthrosis.⁶ Therefore, chronic synovitis and recurrent hemarthroses generate a clinical condition of pain and increased joint volume, evolving into stiffness, muscle weakness, deformities, reduced mobility, irreversible damage to the cartilage, and significant functional loss. All these complications characterize the hemophilic arthropathy.^{8,9}

In this perspective, the best strategy to avoid hemophilic arthropathy is the prevention of recurrent hemarthroses, and the central aspect of hemophilia management is the regular prophylactic intravenous administration of clotting factor concentrate and physiotherapy interventions to avoid repetitive bleeding and prevent progressive joint degeneration. ^{7,10}

Physiotherapy treatment is essential for patients with hemophilia as recurrent hemarthroses trigger chronic synovitis and, subsequently, hemophilic arthropathy, causing major musculoskeletal complications, such as joint movement restriction, contractures, deformities, gait changes, muscle strength reduction, and others sequelae of significant functional impairment.⁴

However, some patients are refractory to conservative treatment. They develop chronic synovitis and recurrent hemarthroses after three to six months of therapy and the therapeutic option is the removal of the hypertrophied and inflamed synovium, a synovectomy. ^{1,10} Radioactive Synovectomy (RS) is the first option for controlling refractory chronic synovitis. It consists of a minimally invasive procedure of intra-articular injection of radioactive drugs for reducing synovial hypertrophy.

This treatment causes the sub synovial vessels plexus sclerosis, consequently reducing the inflammatory process. ^{6,11} This treatment can be performed three times at most, and its main objective is to prevent the appearance of new hemarthroses and the progression of chronic synovitis to hemophilic

arthropathy.7,8

The beneficial effects of RS, demonstrating significant reduction of pain and bleeding events and increasing the quality of life of patients with hemophilia, have been reported by several authors. 7,8,12,13 It is a simple, safe, minimally invasive, low-cost technique, achieving satisfactory results with more than a 75% reduction in hemarthroses in the long term. Therefore, this intervention presents high effectiveness in preventing hemophilic arthropathy. 14

Physiotherapy is an excellent ally to the RS procedure, being highly recommended after the injection due to the musculoskeletal complications that chronic synovitis and hemarthroses may cause, such as movement restriction.⁷

Physical therapy allows for gains or maintenance of muscle strength, mobility, flexibility, balance, and functionality, improving the daily life activities of those with hemophilia. Furthermore, it is highly effective in reducing muscle and joint bleeding, improving the emotional and social well-being of these patients.¹⁵

However, the literature still lacks clinical results regarding the functionality of RS⁸ and scientific sources that discuss and explore physiotherapeutic treatment after the procedure.

OBJECTIVE

The present study aims to establish the influence of physiotherapeutic intervention on the functional independence and joint health of patients with Hemophilia after treatment with radioactive synovectomy (RS).

METHODS

This study has a descriptive, retrospective, analytical design with a qualitative and quantitative methodology. It was conducted from 2018 to 2020 and included patients with chronic synovitis due to hemophilia, and assisted at the Hemotherapy and Hematology Center of Pará (HEMOPA). Radioactive Synovectomy (RS) had been delivered at another clinical center to all participants, regardless the physiotherapeutic treatment, which could be delivered by the HEMOPA professionals or in other facilities.

Patients with Hemophilia A or B admitted at the HEMOPA, of both sexes who underwent an RS procedure, aged between 10 and 30 years, were included. Patients were excluded if they had acute hemarthrosis that compromised the assessments, with a history of concomitant treatments in addition to RS at the evaluated joint, and those who underwent physiotherapy six months after RS.

This study was conducted following the Declaration of Helsinki, the Nuremberg Code, and the Brazilian regulations for clinical research (Resolution 466/12 CNS) of the Brazilian National Health Council. The study protocol was submitted to and approved by the Ethical Review Board of the State University of Pará and was registered with the CAAE number 07801219.7.0000.5174.

The sampling was non-probabilistic, being selected intentionally. Patients found in the HEMOPA Foundation database were initially recruited by phone and briefly informed about the objectives and procedures of the research. Then, they were invited for a visit to the institution for further detailing and, in case the patient was willing to participate, signing the Informed Consent Form or Informed Consent Agreement Form for participants under 18 years of age. After signing Consent forms, the participants undertook physiotherapeutic assessments.

The baseline assessments were based on the participant's

medical records. The researchers collected sociodemographic data (age, sex, marital status, household region, formal education, profession, and income), clinical data (type and severity of hemophilia, age at diagnosis, prophylaxis, presence of inhibitor, synovitis), SR treatment data (joints treated, number of times SR was delivered, self-reported improvement), number of hemarthroses in the six months before and after SR, hemarthroses in the last six months (before the physiotherapeutic assessments), presence of concomitant physiotherapeutic treatments, number of sessions before and after SR and type of physiotherapeutic intervention. If any data was missing in the medical record, the researchers collected directly from the participants during the physiotherapeutic assessment.

The researchers developed the physiotherapeutic assessment. Pain intensity was quantified with a Visual Analogue Scale, joint health with the Hemophilia Joint Health Score version 2.1 (HJHS-2.1), and functional independence with the Functional Independence Score in Hemophilia (FISH).

The HJHS is a scale for assessing the joint health of patients with hemophilia. This evaluation assesses the presence of swelling and its duration, muscle atrophy, crepitus on motion, flexion and extension losses, joint pain, muscle strength, and gait. Each joint has a score range from 0 to 20, and gait has a score from 0 to 4. The total score ranges from 0 to 124. The higher the score obtained, the greater the joint impairment. Our study analyzed the joint that underwent RS (from 0 to 20 points) and the general score.

FISH is a score that combines seven tasks, divided into three domains: self-care, transfers, and mobility. The total score ranges from 7 to 28, and 28 indicates the best level of functionality.

After evaluation, participants were divided into two groups for comparison: those who did and did not undergo physiotherapy after the procedure.

With the data collected, an exploratory and descriptive analysis was conducted with Microsoft Windows Excel 2007[®]. The descriptive statistical analysis of categorical variables was presented in absolute and relative frequencies. The small sample size made the application of statistical inferences unfeasible.

RESULTS

There were 448 people with Hemophilia in the HEMOPA database, of which only 11 underwent RS, and 8 participants were included in the study. The patient sample corresponds to 12 joints subjected to SR, and the physiotherapeutic assessment occurred 1.5 years on average after the last RS infusion.

The participants' ages ranged between 12 and 29 years, with a mean of 19±5.3 years. All participants were male, seven patients were single (87.5%), and five lived in the countryside of the state (62.5%). Regarding the type of Hemophilia, all participants had Hemophilia A (100%), and six of them (75%) had a severe form of the disease. The distribution between the groups with and without physiotherapeutic treatment was equivalent. Four patients (50%) received the diagnosis of Hemophilia before the 1st year of age.

All patients were undergoing treatment with secondary prophylaxis and there was no presence of inhibitors use among the participants. The characterization of the study population is described in Table 1.

Table 2 describes the distribution of patients regarding the physical therapy treatment, showing that 50% of the participants underwent physical therapy before the SR procedure.

Table 1. Sociodemographic characterization of participants

Variables	Absolute frequency	Relative frequency (%)	95%CI
Sex			
Male	8	100	100 - 100
Marital status			
Single	1	12.5	0.3 - 52.7
Married	7	87.5	47.3 - 99.7
Schooling			
Ongoing elementary school	2	25	3.2 - 65.1
Ongoing high-school	3	37.5	8.5 - 75.5
High-school completed	1	12.5	0.3 - 52.7
Undergraduate	2	25	3.2 - 65.1
Occupation			
Self-employed work	1	12.5	0.3 - 52.7
Student	7	87.5	47.3 - 99.7
Region			
Inner-city areas of Belém	2	28.6	3.7 - 71.0
Countryside	5	71.4	29.0 - 96.3
Income			
1 minimum wage	3	37.5	8.5 - 75.5
2 minimum wages	2	25	3.2 - 65.1
≥ 3 minimum wages	3	37.5	8.5 - 75.5

95%CI: 95% Confidence Interval

The same patients also underwent physical therapy treatment after SR, as recommended. Regarding the number of sessions after SR, it was observed that this group had more than ten sessions and that the type of treatment of the majority was electrothermal phototherapy, cryotherapy, and kinesiotherapy. Regarding the other group, all participants declared not taking physical therapy treatments due to obstacles since all of this group were residents of the countryside of the State of Pará.

Regarding the location of the SR, of the 12 joints subjected to the procedure, 41.67% were elbows, 33.33% were knees, and 25% were ankles, considering the total sample. Also, equivalent data was found in the number of treated lower limbs (LL) and upper limbs (UL). However, considering the participants who received physiotherapy interventions, the knee was the lower limb joint subjected to RS, while in the control group, RS was performed on the ankle. Furthermore, three participants in the physical therapy group had two or more target joints, and two performed the procedure three or more times. One had three SR injections in the same joint (Table 3).

Table 2. Clinical description of the participants

Variables	Absolute Frequency	Relative frequency(%)	95%CI
Hemophilia type			
Hemophilia A	8	100	100 - 100
Hemophilia B	0	0	0 - 0
Severity			
Mild	0	0	0 - 0
Moderate	2	25	3.2 - 65.1
Severe	6	75	34.9 - 96.8
Age at diagnosis			
<1 year	4	50	15.7 - 84.3
1 - 2 years	2	25	3.2 - 65.1
≥ 2 years	2	25	3.2 - 65.1
Secondary			
Prophylaxis	8	100	100 - 100
Inhibitor			
Yes	0	0	0 - 0
No	8	100	100 - 100

95%CI, 95% Confidence Interval

Table 3. Distribution of patients according to the presence of physiotherapeutic intervention before and after RS injections

Variables	Absolute frequency	Relative frequency (%)	
Physiotherapeutic intervention after RS			
Yes	4	100	
Number of sessions before RS			
≤10	1	25	
> 10	3	75	
Number of sessions after RS			
> 10	4	100	
Intervention			
Electrotherapy and kinesiotherapy	1	25	
Electrotherapy, cryotherapy, and kinesiotherapy	3	75	

RS, Radioactive synovectomy

Most participants (6 - 75%) reported they improved after the RS injections (Table 4).

 Table 4. Participant distribution regarding the interventions with

Variables	Physiotherapy group n= 4	Control group n= 4
	n (%)	n (%)
RS site		
UL joints	1 (25%)	1 (25%)
LL joints	2 (50%)	2 (50%)
UL and LL joints	1 (25%)	1 (25%)
Number of joints		
1	1 (25%)	3 (75%)
2	3 (75%)	1 (25%)
Number of RS injections		
1	1 (25%)	2 (50%)
2	1 (25%)	1 (25%)
≥ 3	2 (50%)	1 (25%)
Self-reported improvement		
Yes	3 (75%)	4 (100%)
No	1 (25%)	0 (0%)

RS, Radioactive synovectomy; UL, upper limbs; LL, lower limbs

When analyzing the number of hemarthroses in the treated joint between the two groups, considering six months before and after RS, the results showed that those who underwent physiotherapy intervention had more significant bleeding before RS when compared to those who did not undergo physiotherapy treatment. It was impossible to observe such a significant reduction in bleeding six months after the procedure, regardless of the group. Nonetheless, both groups considerably reduced the number of hemarthroses in the six months before the assessments, determining that the joints studied had better responses to RS in the long term.

Furthermore, during the evaluation, three of the patients in the physiotherapy group (75%) had synovitis in one of the joints regardless of the treatment, compared to the control group. In the latter, synovitis was observed in two participants (50%). Nonetheless, pain was rarely reported at the time of evaluation, as there was no occurrence of acute hemarthrosis regardless of the studied group, as seen in Table 4.

Table 5 compares groups according to the performance of physiotherapy after RS, considering pain, joint health, and

Functional Independence. No statistically significant differences were found between the groups, indicating that they presented mild pain complaints (50% of the physiotherapy group and 25% of the non-physiotherapy group), joint impairment, and impacts on their functional independence.

However, it is essential to highlight that in the HJHS score, the lower the score, the better the patient's joint health, and in the FISH Score, higher values demonstrate a higher level of functionality. Hence, the results showed that the group who did not take physiotherapy intervention had better joint health and greater functional independence than the other group.

Table 5. Comparison of participants regarding the presence of physical therapy intervention, considering the number of hemarthrosis, pain intensity, and presence of synovitis.

Variables	Phisiotherapy intervention n= 4 n (%)	Without physiotherapy intervention n= 4 n (%)		
Hemarthrosis 6 months before RS				
≤3 times	2 (50%)	3 (75%)		
≥ 5 times	2 (50%)	1 (25%)		
Hemarthrosis 6 months before evaluation				
None	2 (50%)	3 (75%)		
1 to 3 three	1 (25%)	1 (25%)		
≥ 5 times	1 (25%)	0 (00%)		
Synovitis*	4 (100%)	1 (25%)		
Pain in the joint with RS	1 (25%)	1 (25%)		
Pain in other joint	2 (50%)	1 (25%)		

RS, Radioactive synovectomy; * considering synovitis in the same joint with RS or any Other after RS intervention

DISCUSSION

Despite recent advances in the treatment of people with hemophilia, including new therapeutic options and the availability of prophylaxis, chronic synovitis is still the main clinical problem in a significant portion of patients with hemophilia throughout the world. Physiotherapy is an essential part of the treatment, above all, due to the predisposition to bleeding of these patients and the appearance of severe sequelae in their osteoarticular system. These issues make physiotherapy intervention a necessary strategy for both prevention and recovery of functionality, 17 also an essential treatment after the RS procedure.

Although the present study has a very small number of participants, the epidemiological profile is similar to that described in the specialized literature, 8,12,18,19 as the participants aged between 16 and 22 years, with predominantly severe Hemophilia A. These characteristics are consistent with the disease's epidemiological and phenotypic characteristics, demonstrating the prevalence of recurrent hemarthrosis in a relatively young population.

Despite being recommended, only half of the participants underwent physiotherapeutic treatment before and after RS. Among the correlated causes, the barriers to physiotherapeutic treatment within the State are evident. The participants reported this issue as a geographical obstacle given the dimensions of Pará State hindering the monitoring of people with this coagulopathy.

Hemophilia is a complex and rare disease requiring constant monitoring by experienced professionals in specialized centers. Factors such as distance, travel time, limited public transport, travel costs, and reduced mobility due to arthropathy represent considerable barriers to regular participation in physiotherapeutic treatment for patients who dwell far from these centers.²⁰

In Pará State, only the coordinating blood center in the state capital, Belém, has a specialized physiotherapy service. This scarce clinical resource results in considerable difficulty in accessing specialized physiotherapeutic treatment for people with hemophilia living in the countryside. In the present study, half of the participants in the physiotherapy group underwent physiotherapeutic treatment in other facilities, limiting the study regarding the exact number of sessions they received and the types of resources used in the treatment, generating only categorical variables for analysis. Procedures such as electrotherapy, cryotherapy, and kinesiotherapy have been reported. Such interventions have evidence of efficacy for patients with hemophilia.⁹

Regarding the location of the RS, our total sample differs from studies in general. The studies commonly indicate the knee joint as the most prevalent site for RS.^{12,16,18} The elbow was the most prevalent joint treated with RS in our study. The literature reports that the results of RS for the knee joint are worse when compared to other smaller joints, such as the ankle and elbow. This understanding suggests that knee joints are more prevalent and require more than one application, often due to their size.²¹

In the inter-group analysis, it is observed that, compared to the control group, the physiotherapy group had disadvantages since that group received RS only in joints presented by the literature as highly responsive joints to this intervention, such as ankles and elbows. The patients of the physiotherapy group received RS in the knees only. It is also worth mentioning that three participants in the physiotherapy group (75%) had two joints subjected to RS, which could be both knees or knee and elbow. This treatment with two joints can impact the functionality and success of the therapy.

Even in this context, it was possible to observe that participants from both groups needed more than one application in the same joint. This issue allows for the inference that the chronic synovitis persisted in the treated joint even after treatment with RS. This result can be justified due to the type and severity of hemophilia in the studied sample. Hemophilia severity predisposes the patient to have more spontaneous bleeding, causing them to be more susceptible to the occurrence of chronic synovitis.²²

As expected, it was possible to observe the effectiveness of RS in reducing the incidence of hemarthroses and pain. These effects agree with data from the literature^{11,16,19,21} on the effectiveness of RS. Our data demonstrated that this improvement occurred, especially in the long term.

Some studies show the effectiveness of RS in the first six months after its administration^{12,19} and that it occurs mainly with the reduction of hemarthroses. Others find good results both in the first six months and after one year but with more significant results in the long term. ¹⁸

It was also observed that patients who underwent physiotherapy had more hemarthroses than the control group. It is necessary to emphasize that this group was mainly composed of the youngest participants of the sample. One study¹⁹ reported a correlation between mean age of 12 years and bleeding episodes after RS, suggesting that younger patients are more

prone to bleeding after such treatment, a characteristic that may be associated with the joint traumas to which younger patients are typically exposed.

There was no statistical difference between the groups concerning the total HJHS score. However, it was possible to observe that the present study found an average score of HJHS of 20.25 in the physiotherapy group, a score that supports the existence of chronic joint damage among these participants. Similar data was found in a Brazilian study¹⁹ that evaluated the joint health of 16 patients with hemophilia after RS. The authors found an average of 26.94 in HJHS and associated this high score with the delay in prophylactic treatment in Brazil, given that the median HJHS of European boys who undertake primary prophylaxis is 5 with an almost ideal functional capacity.²³

Prophylactic treatment in Brazil is recent, being reported only after 2012. Therefore, many patients showed some deterioration in their joint condition. ¹⁹ In addition, patients in the present study had access barriers, more than one target joint, and frequent bleeding episodes, diminishing the score observed.

Furthermore, the prevalence of hemarthroses in the physiotherapy group was more significant, both before and after RS. At the time of evaluation, synovitis was present in three patients (75%), regardless of the joint. This problem can lead to more significant joint damage and higher HJHS scores.

It is important to emphasize that, according to self-reported information, one participant in the physiotherapy group received deep heat, a contraindicated treatment, as well as severe levels of joint impairment, with hemophilic arthropathy in other joints, constant and more intense bleeding compared to the rest of the sample, generating a possible source of bias to the study, pushing the standard deviation, as shown by the maximum values.

A study found a correlation between the number of hemarthroses throughout life and the total HJHS score, such that a higher number of hemarthroses is associated with higher HJHS scores.²⁴ Nonetheless, this study is limited, as the number of hemarthroses is obtained by a subjective approximated measurement, according to the patient self-reported information of those who do not attend the blood center at each episode of hemarthrosis. This source of information jeopardizes the statistical comparison with healthy joints.

This issue was due to the difficulty in collecting the exact number of hemarthroses that occurred before and after treatment, as patients were unsure whether the event had occurred, and the information was not included in the medical records. Many patients take more than a year to attend routine appointments, which results in missing data, hindering the treatment.

Thomaz et al. ¹⁹ observed that most patients who received RS had an increased range of motion. However, no statistical significance was found when comparing the changes in range of motion between patients with and without access to physiotherapy. The loss of statistical power of the small sample size may justify it.

There were also no significant differences between the groups considering functional independence measured with the FISH score. The lack of difference between groups may demonstrate that physiotherapy did not influence the patients' functional capacity. On the other hand, at the time of evaluation, the control group presented lower pain intensity, better joint health, and better functional independence than the physiotherapy group.

The lack of HJHS and FISH data from previous physiotherapeutic assessments did not allow for quantifying participant improvement. Based on the collected data, it is possible to infer that the participants in the physiotherapy group had less joint health and functional independence prior to the procedure compared to the control group. Nonetheless, seven participants (75%) reported improvement after RS.

CONCLUSION

The present study is the first attempt to describe the impact of physiotherapy on the functional independence of hemophilic patients undergoing radioactive synovectomy (RS). Although this study has limitations regarding the subjectivity of the data and the difference between the groups, it was possible to observe that the group that underwent physiotherapy before and after RS had higher HJHS scores and more functional limitations compared to the group that did not have access to physiotherapy.

It is worth mentioning that patients who underwent physiotherapy had functionality benefits at other joints and not only at the one treated with RS, presenting a thorough context regarding hemophilia. Furthermore, the participants reported improvement after RS, leading us to conclude that it effectively reduced pain and the number of hemarthroses in most of these patients, possibly improving quality of life.

The small sample size did not allow for conventional statistical tests. Therefore, an exploratory, descriptive analysis was conducted based on the data collected. It was possible to observe the urgent need to intensify the early diagnosis of chronic synovitis in patients admitted at the HEMOPA, as the number of patients eligible for the procedure is still very small, partly due to the lack of regular medical visits.

The physiotherapeutic assessments were conducted at different time frames, in different settings for the treatment of the participants, the different physiotherapy professionals, the lack of information regarding the physical therapy delivered at other physiotherapy centers or clinics, and the different number of sessions received by the participants, as well as their treatment adherence are among the limitations of this study. These factors arise from the difficulty in accessing the blood center information due to the extensive geographical dimensions of Pará State.

Lastly, the small sample size does not allow for the generalizing of our results, therefore there must be other studies with larger samples to demonstrate the importance of physiotherapy and broaden the knowledge of the patients addressed in this research.

REFERENCES

- Atilla B, Güney-Deniz H. Musculoskeletal treatment in haemophilia. EFORT Open Rev. 2019;4(6):230-239. Doi: 10.1302/2058-5241.4.180068
- 2. García-Chávez J, Majluf-Cruz A. Hemofilia. Gac Med Mex. 2013;149(3):308-21.
- Brasil. Ministério da Saúde. Hemofilia: conheça doença que afeta quase exclusivamente homens [texto na Internet]. Brasília: Ministério da Saúde; c2020 [citado 2021 Jan 18]. Disponível em: https://www.gov.br/saude/pt-br/assuntos/noticias/2020/janeiro/hemofilia-conheca-doenca-que-afetaquase-exclusivamente-homens

- 4. Jorge MSG, Moreira IS, Felimberti G, Wibelinger LM. Abordagem fisioterápica na dor e na qualidade de vida de um indivíduo com artrite hemofílica. Relato de caso. Rev Dor. 2016;17(1):65-8. Doi: 10.5935/1806-0013.20160016
- 5. Cuesta-Barriuso R. Effectiveness of physiotherapy in the treatment of hemophilic arthropathy a systematic review. Ann Hematol Oncol. 2017; 4(9):1172.
- Querol-Giner M, Pérez-Alenda S, Aguilar-Rodríguez M, Carrasco JJ, Bonanad S, Querol F. Effect of radiosynoviorthesis on the progression of arthropathy and haemarthrosis reduction in haemophilic patients. Haemophilia. 2017;23(6):e497-e503. Doi: 10.1111/hae.13326
- van Vulpen LFD, Thomas S, Keny SA, Mohanty SS. Synovitis and synovectomy in haemophilia. Haemophilia. 2021;27 Suppl 3(Suppl 3):96-102. Doi: 10.1111/hae.14025
- Faracoa F, Uribeb A, Torob A, Lópezc J, Faracod M, Gómez Barrerae M. Efectos clínicos y funcionales de la radiosinoviortesis en pacientes con sinovitis crónica hemofílica. Un estudio prospectivo. Nivel de Evidencia: IV. Rev Colomb Ortop Traumatol. 2015;29(3):95-100. Doi: 10.1016/j.rccot.2016.02.006
- Hashem F, Bladen M, Carroll L, Dodd C, Drechsler WI, Lowery D, et al. Protocol for a feasibility randomised controlled trial of a musculoskeletal exercise intervention versus usual care for children with haemophilia. BMJ Open. 2019;9(8):e029474. Doi: 10.1136/bmjopen-2019-029474
- Querol F, Rodriguez-Merchan EC, Aznar JA, Lopez-Cabarcos C, Villar A. Post-synoviorthesis rehabilitation in haemophilia. Haemophilia. 2001;7 Suppl 2:54-8. Doi: 10.1046/j.1365-2516.2001.00111.x
- Thomas S, Mendes JD, Souza SA, Lorenzato CS, Assi PE, Pacheco LR, et al. Radioactive synovectomy with (90) yttrium and (153) samarium hydroxyapatite in haemophilic joints: preliminary study on radiation safety. Haemophilia. 2013;19(4):632-6. Doi: 10.1111/hae.12122
- 12. Thomas S, Gabriel MB, Assi PE, Barboza M, Perri ML, Land MG, et al. Radioactive synovectomy with Yttrium⁹⁰ citrate in haemophilic synovitis: Brazilian experience. Haemophilia. 2011;17(1):e211-6. Doi: 10.1111/j.1365-2516.2010.02379.x
- Alioglu B, Ozsoy H, Koca G, Sakaogullari A, Selver B, Ozdemir M, et al. The effectiveness of radioisotope synovectomy for chronic synovitis in Turkish paediatric haemophiliacs: Ankara experience. Haemophilia. 2010;16(6):932-6. Doi: 10.1111/j.1365-2516.2010.02276.x
- Tena-Sanabria ME, Rojas-Sato YF, Castañeda-Resendiz JC, Fuentes-Herrera G, Álvarez-Martínez FA, Tena-Gonzalez YI, et al. Treatment with radiosynoviorthesis in hemophilic patients with and without inhibitor. BMC Pediatr. 2020;20(1):173. Doi: 10.1186/s12887-020-02071-3
- Schäfer GS, Valderramas S, Gomes AR, Budib MB, Wolff ÁL, Ramos AA. Physical exercise, pain and musculoskeletal function in patients with haemophilia: a systematic review. Haemophilia. 2016;22(3):e119-29. Doi: 10.1111/hae.12909
- Rodriguez-Merchan EC, De la Corte-Rodriguez H, Jimenez-Yuste V. Radiosynovectomy in haemophilia: long-term results of 500 procedures performed in a 38-year period. Thromb Res. 2014;134(5):985-90. Doi: 10.1016/j.throm-res.2014.08.023

- Vinholte Júnior NS, Vieira WHB. Meios físicos na reabilitação do paciente portador de hemofilia – uma revisão de literatura. Rev UNI-RN. 2014;9(1/2):197.
- Sunny SS, Hephzibah J, Shanthly N, Oommen R, Mathew D, Abraham A. Yttrium-90 synovectomy in hemophilic arthropathy: an institutional experience for 15 years. Indian J Nucl Med. 2020;35(2):143-146. Doi: 10.4103/ijnm.IJNM_141_19
- Magalhães AF, Oliveira LCO, Pitella FA, Wichert-Ana L, Engel EE, Barbieri CH. Yttrium-90 radiosynovectomy in knees and ankles (25 joints in 22 hemophilic patients). Short-term results. Hematol Transfus Cell Ther. 202;43(1):15-20. Doi: 10.1016/j.htct.2019.11.001
- Wagner B, Seuser A, Krüger S, Herzig ML, Hilberg T, Ay C, et al. Establishing an online physical exercise program for people with hemophilia. Wien Klin Wochenschr. 2019;131(21-22):558-566. Doi: 10.1007/s00508-019-01548-1

- 21. Haje DDP, Ulisses J, Calegaro M, Machado J, Santa J, Almeida De C, et al. Sinovectomia com samário-hidroxiapatita nos tornozelos e cotovelos de hemofílicos. Rev ABTPé. 2011; 5(1): 28-35.
- 22. Galatti LR, Luiza M, Alves T, Duarte E. Exercício físico e hemofilia: conceitos e intervenção. Rev Mackenzie Educ Fís Esporte. 2012;11(2):96-109.
- Groen W, van der Net J, Bos K, Abad A, Bergstrom BM, Blanchette VS, et al. Joint health and functional ability in children with haemophilia who receive intensive replacement therapy. Haemophilia. 2011;17(5):783-90. Doi: 10.1111/j.1365-2516.2011.02606.x
- 24. Rampersad AG, Shapiro AD, Rodriguez-Merchan EC, Maahs JA, Akins S, Jimenez-Yuste V. Radiosynovectomy: review of the literature and report from two haemophilia treatment centers. Blood Coagul Fibrinolysis. 2013;24(5):465-70. Doi: 10.1097/MBC.0b013e3283602a28