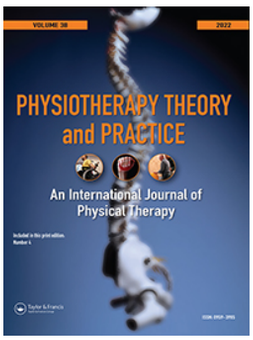

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Strength training program for an athlete with hemophilia A and an inhibitor while taking a new prophylactic drug treatment: a case report

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ABSTRACT

Background: Currently, patients with hemophilia and inhibitor are being offered therapy, including a tissue factor pathway inhibitor (TFPI). The new prophylactic drug treatment may allow for new opportunities for other interventions and overall improvement in quality of life. This case report assessed the safety and effects of a strength training program in a patient with an inhibitor on a specific new study drug

Description: A 20-year-old patient with severe hemophilia A with an inhibitor participated in a 6-week strength training program. The strengthening program consisted of 7 exercises involving limb and trunk muscles. A qualitative assessment of movement patterns was performed using the Functional Movement Test. Dynamic balance was measured by the Y-Balance Test, whereby the power of lower extremities was measured by Counter Movement Jump. The Quality of Life Index was done by survey to assess the perceived overall quality of the patient's life. The exercise fatigue after each training was measured with the Borg scale

Outcomes: After the intervention during treatment with the new drug, the patient's quality of life increased, especially in terms of health and function (from 15.6 to 29.1 points), also, the power of the lower limbs increased. There were no bleeding episodes during the intervention and after a 3-month follow-up

Conclusion: The proposed program during the application of the new prophylactic treatment seems to be effective in improving quality of life and increasing lower limb power in a hemophilic patient with an inhibitor. However, randomized clinical trials are needed to confirm the results.

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Hemophilia; inhibitor; quality of life; exercise training; joint bleeding

Introduction

Hemophilia (PWH) is a plasma hemorrhagic disorder caused by a deficiency of blood coagulation factor VIII (hemophilia A) or, less commonly, IX (hemophilia B) (Paredes et al., 2021). Hemophilia (type A) is a blood disease associated with coagulation factor VIII (FVIII) deficiency. Severe hemophilia is characterized by very low levels of clotting factor activity at <1%. Up to 30% of patients with severe hemophilia A will develop alloantibodies or FVIII inhibitors (Bossard et al., 2008; Forsyth and Zourikian, 2012; Luck et al., 2004). An alloantibody against FVIII is the most common form of a coagulation factor inhibitor. Persons with severe hemophilia experience bleeding episodes which may occur spontaneously or due to injuries. Most of them (about 85%) appear in the musculoskeletal system and appear as intra-articular hemorrhages and intramuscular hematomas (Bossard et al., 2008; Schäfer et al., 2016). Most often, intra-articular hemorrhages affect the knees, and ankles, but can occur in other joints as well (Bossard et al., 2008). Recurrent bleeding predisposes to chronic synovitis and, subsequently hemophilic arthropathy. Chronic pain, muscle atrophy, loss of joint motion, articular instability, and decreased motor skills in the long term, leading to reduced functional health status and quality of life (Bossard et al., 2008; Schäfer et al., 2016; Srivastava et al., 2013).

The underlying component of hemophilia treatment is bleeding prevention. The most severe impediment in hemophilia is the development of an inhibitor. Factor VIII inhibitor (FVIII) is an alloantibody that reveals itself after being exposed to FVIII concentrate. It may appear in early childhood in approximately 30% of patients with severe hemophilia A (Ehrenforth et al., 1992). Inhibitors can be considered strong or weak and it depends on the immune response (Meeks and Batsuli, 2016). Inhibitors significantly increase health care costs,

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create psychosocial stressors for patients and their families, and have negative consequences in disease morbidity and mortality due to bleeding episodes difficult to treat (Meeks and Batsuli, 2016).

Patients with inhibitors showed greater difficulty with mobility, increased joint pain, and a higher incidence of hemophilic arthropathy compared to patients without inhibitors (Bossard et al., 2008; Forsyth and Zourikian, 2012). The gold standard of treatments in patients with severe type A hemophilia is a prophylactic treatment with intravenous FVIII concentration, although FVIII inhibitors may appear, making it ineffective. Currently, primary treatment modalities for patients with inhibitor are bypassing agents (BPAs). BPAs help to achieve hemostasis that avoids the need for thrombin generation by factor VIII or IX. Currently available BPAs include recombinant factor VIIa and activated prothrombin complex concentrate (Cormier, Batty, Tarrant, and Lillicrap, 2020). Patients who exhibit low titer and poor inhibitor response (<5 Bethesda units [BU]/ml) may receive agent replacement therapy at higher doses for bleeding management and prophylaxis. High-titer inhibitors (>5 BU/ml) are considered a low-risk trait, but treatment of these patients with immune tolerance induction (ITI) should be implemented as soon as possible (Meeks and Batsuli, 2016). The few studies so far have shown that combined therapies can be more effective and thus bring the desired health effect (Zanon et al., 2020). For example, in patients with hemophilia, rehabilitation may be used in addition to pharmacological treatment.

The combination of prophylactic treatment and rehabilitation shows positive effects (Elnaggar, 2018; Zanon et al., 2020). Physical fitness and neuromuscular development concerning PWH is essential and regarded as a highly important part of life. Therefore, muscle strengthening, coordination, balance, and general fitness should be taken into account during rehabilitation (Elnaggar, 2018; Srivastava et al., 2013; Zanon et al., 2020).

Exercise can increase the production of synovial fluid and thus provide nutrients to the articular cartilage, potentially reducing joint degeneration (Hinman, Heywood, and Day, 2007). In addition, strength training supports cartilage lubrication, stimulates bone tissue, and increases the strength of periarticular muscles (Schäfer et al., 2016). A few studies have evaluated the efficacy and safety of implemented physiotherapy and training methods (Hilberg, 2018; Strike and Mulder, 2016). In one case report, researchers demonstrated improvements in range of movement of knees and ankles, pain in both knees, function of ankle joint and safety using a physiotherapy program based on manual therapy in a patient with hemophilia and an inhibitor (Cuesta-Barriuso and Trelles-Martínez, 2018).

The patient presented in this case report was treated in the Department of Pediatric and Adolescent Hematology in another city until the age of 18 years (Miklasz, Laguna, and Matysiak, 2020). After changing the place of residence the patient was referred to the Department of Hematology for Adults and to our Rehabilitation Clinic. The patient was a subject in an on-going drug trial for a new pharmacological intervention. The purpose of this report is to describe the use of an intensive strengthening program for this patient with hemophilia A with an inhibitor while taking a new drug designed to prevent joint hemorrhages and to assess the safety and efficacy of the protocol for this patient. A sports-related approach was proposed as a good fit for this patient's interests and abilities.

Case description

History

A 20-year-old student (height 179 cm, body mass 79.6 kg) with severe type A hemophilia and inhibitor volunteered for the report. During the interview and from medical history information on the patient's history was collected. The following is a brief history of the patient's hematologic treatment, which is described in detail in previous publication (Miklasz, Laguna, and Matysiak, 2020).

The first signs of hemophilia occurred after probing the nasolacrimal ducts for obstruction at three months of age, where there was prolonged oozing of blood from the tubules. This was followed by hemorrhage into the soft tissues of the hand at 6 months of age and a few months later by the first episode of hemorrhage into the right elbow joint. Laboratory tests of the dilated blood coagulation system showed abnormalities suggestive of hemophilia A severe form (i.e. prolonged APTT to 94s, FVIII < 1%). In early childhood, the patient underwent surgery to remove both left and right venous ports. At the age of 14, there was a serious intracranial hemorrhage (i.e. episode of increasing headache with vomiting and a seizure) due to an accident. Intensive therapy with aPCC was then necessary, where a good result was achieved. During this time, there were also bleeds and hemophilic arthropathy occurred. Therefore, the boy underwent radiosynvectomy of the right elbow joint, and after six months, the left elbow joint and the right knee joint.

The aPCC prophylaxis used effectively reduced the frequency and intensity of joint hemorrhages. During prophylaxis, FVIII inhibitor levels were maintained at

18–25 BU/mL. The patient's quality of life improved significantly after starting aPCC prophylaxis (Miklasz, Laguna, and Matysiak, 2020).

The right ankle joint was a target joint, it had a limited range of motion with crepitus and chronic synovitis. The patient's physical activity before the case report consisted of swimming, light strengthening program (i.e. 3 times a week, 1.5 hours each training unit session) using body weight and resistance bands. The patient, several months prior to our case report, had unplanned interruptions in physical activity once a month on average due to minor joint bleeds. The patient was interested in bodybuilding and strength training, but due to the risk of intra-articular hemorrhages, he could not perform such intensive strength training.

The patient was informed about the training protocol and the planned publication and gave written consent to participate. He was in constant contact with the Hematology and Transplantology Clinic in Gdansk through the duration of the case report. The report was approved by the Independent Bioethical Committee for Scientific Research.

New prophylactic treatment

Two months before the start of our training, the patient was included in a Phase II clinical trial investigating a new prophylactic treatment: Marstarcimab (previously PF-06741086). This study showed the potency of ex vivo Mastarcimab in restoring hemostasis in whole blood and plasma of hemophilia patients (NCT02974855) (Patel-Hett et al., 2019). The drug's clinical trials are currently in Phase III (NCT03938792).

This prophylaxis was administered as a subcutaneous injection – 300 mg weekly. For the first 6 weeks of treatment with the new drug, our patient followed a conventional training program for the PWH at the highest level of progression proposed by Mulder (2006) in collaboration with the World Federation of Hemophilia. This training consisted of exercises increasing the range of motion, strength and proprioception of the elbow, knee and ankle joints. The absence of any health problems such as pain, joint dysfunction or loss of range of motion allowed us to decide to introduce the patient to a more demanding and strenuous exercise program. Our training program started after a 2-week break from the last training session. Our goal was to improve lower and upper limb power, dynamic balance and movement patterns, and the quality of life of this patient athlete with hemophilia.

Outcome assessments

All tested variables were measured just before and after the planned training program. Anthropometric variables, i.e. body mass, skeletal muscle mass, and body fat percentage were measured by the body composition analyzer (InBody 270, InBody Co., Seoul, Korea). The range of joint motion was measured by a universal goniometer using standardized anatomical landmarks. Biceps and thigh circumferences were measured using a measuring tape. The following outcome measures were used.

BORG CR10 scale

The Borg CR10 scale was used to assess the exertion of each training unit. This scale is based on a subjective categorical-numerical measure with possible scores from 0 to 10, where 0 means "nothing at all" and 10 means "extremely strong (almost maximum)" (Borg, 1998). The post-workout fatigue assessment also helped to continuously evaluate the safety of the intervention. If a patient reported maximum values (i.e. 9–10 points on the scale) it would require a reduction in training load to avoid overload and potential injury.

Weight-bearing lunge test (WBLT)

Weight-bearing lunge test (WBLT) was used to assess the range of mobility of dorsiflexion of the ankle joint. The participant was asked to place the foot on a line perpendicular to the wall in a standing position so that the knee was in line with the second toe and great toe 10 cm away from the wall. The participant was then to perform a forward lunge, driving the knee toward the wall until contact touched the wall. The foot was moved away or closer from the wall 1 cm at a time until the subject was able to touch the knee at the maximum distance from the wall. Maximal dorsiflexion ROM was assessed using a tape measure as the maximum distance of the toes from the wall during contact between the wall and the knee without lifting the heel off the ground. The subject performer three practice trials and three test trials per limb. The average of the test trials was taken for data analysis

Quality of life index

The Quality of Life Index was used to assess the perceived overall quality of life (QLI – Generic III Version). This version consists of 66 items based on a six-point Likert-type scale. It examines four aspects: health and functioning, socioeconomic, psychological and family (Ferrans and Powers, 1992; Jaracz and Kozubski, 2003).

Table 1. Exercise program training.

Exercise	Series/Reps	Load (kg)
Dumbbell bench press while lying on a Swiss ball	4/12	17.5
Dumbbells curl sitting on bench	4/12	10
Dumbbells tricep extension lying on a horizontal bench	4/12	8
Nordic hamstring curl	4/8	Body weight
Romanian barbell deadlift	4/12	25
Bulgarian squat with a dumbbell	4/12	12.5
Pulling the knees to the abdomen in the support on the forearms with feet on TRX	4/20	Body weight

TRX – Total Body Resistance Exercise, kg – kilograms

Functional movement screen

Functional Movement Screen (FMS) was used to assess the quality of the movement patterns including mobility and stability of peripheral joints and torso. The test consists of 7 functional movement tasks: 1) deep squat; 2) in-line lunge; 3) hurdle step; 4) shoulder mobility; 5) active straight legrRaise; 6) trunk stability push up; and 7) rotary stability. The participant gets 3 points for the correct performance of a movement task, 2 for a compensated movement and 1 for a incorrect movement. If pain occurred during a given test, the participant scored 0 points. The maximum total number of points to be scored was 21 (Teyhen et al., 2012).

Y-balance test

To obtain stability results of the lower limbs, the Y-Balance Test (YBT) Lower Quarter (LQ) and the YBT Upper Quarter (UQ) were used. Both tests use the Y-Balance Test Kit platform (Move2Perform, Evansville, IN, USA). The participant with the free limb moves the block in the anterior (ANT), posterolateral (PL), and posteromedial (PM) directions trying to reach the farthest point possible, and then return to the starting position (Plisky et al., 2009; Shaffer et al., 2013). To perform the UQ-YBT, the patient must support his weight on one upper limb in a supported push-up position, while the opposite limb moves the block as far as possible in the medial (MED), inferolateral (INF) and superolateral (SUP) direction (Gorman, Butler, Plisky, and Kiesel, 2012). Both test protocols were made following Plisky et al. (2009) and Gorman, Butler, Plisky, and Kiesel (2012) suggestions. The analysis uses the maximum reach from 3 trials.

Counter movement jump

Counter Movement Jump (CMJ) was used to assess the height of the jump. The test involves performing a movement task while standing on a force platform (Fusion Sport Smart Jump mat, Fusion Sport, 2 Henley ST, Coopers Plains, QLD, 4108, Australia). The participant was asked to perform a deep squat (minimum of 120°) with hands on hip, and then make a jump upwards with straightening the knees as soon as possible. The best result from 5 trials was registered. The height was calculated as the difference (in cm) of the center of mass of the patient from his standing position to the highest point of the jump. Data from the computer software connected to the platform was used in the analysis (Hilmersson, Edvardsson, and Tornberg, 2015).

Intervention

The intensive strength training program consisted of 18 training sessions. Three sessions per week for 6 weeks. The program consisted of seven selected exercises to strengthen the trunk, lower extremities, and upper extremities. The load of the exercises was the patient's body weight and external resistance using dumbells and barbells. One exercise used the Total Resistance Exercises (TRX) system, which is a suspension training device. The average training unit session lasted 60 minutes. The interval between exercises was of 90 seconds and 60 seconds between series. Before starting the program, the patient underwent strength tests to select the appropriate load for each exercise. The load on each exercise was 80% of the maximum load calculated using the 20 reps method. This method involves the patient performing 20 repetitions of a given exercise at the maximum subjective load. Then, 80% of the number of load obtained is applied to the workout. A detailed description and demonstration of the exercises is shown in Table 1 and Figure 1 and Figure 2.

During the intervention and for 3 months afterward, the patient was under the control of hematologists and physiotherapists. The patient could report any health problem at any time during the training by telephonic contact with a hematologist or during or after a training session to a physiotherapist.

Outcomes

From the moment of taking a new prophylactic treatment through the 6-week strength training program and in the 3-month follow-up, the patient did not report any injuries to the musculoskeletal system or bleeding episodes. Before,

Table 2. Comparison of variables between pre-training and post-training.

		Assesment		
Variables	Measurment	Pre-training	Post-training	Percentage improvement (% change)
Body composition analyses	Body mass (kg)	79.6	83.1	4.40
	Fat percentage (%)	17.60	18.70	6.25
	Skeletal muscle mass (kg)	37.3	38.5	3.22
Circumferences (cm)	Left biceps	34.5	36	4.35
	Right biceps	31.5	32.5	3.17
	Left thigh	46	48.5	5.43
	Right thigh	45.5	48	5.50
Range of motion (degrees)	Flexion right knee	140	140	0.00
	Flexion left knee	140	140	0.00
	Extension right knee	−3	−3	0.00
	Extension left knee	−3	−3	0.00
	Dorsalflexion right ankle	10	10	0.00
	Dorsalflexion left ankle	30	25	−16.67
	Plantarflexion right ankle	25	25	0.00
	Plantarflexion left ankle	25	25	0.00
	Flexion right knee	135	135	0.00
	Flexion left elbow	140	140	0.00
	Extenion right elbow	0	0	0.00
	Extension left elbow	0	0	0.00
WBLT (cm)	Right ankle	3.5	3	−14.3
	Left ankle	9	7	−22.2
BORG CR 10 Scale (pts)	Fatigue (mean of three training units)	1 week – 7.5	6 week – 6	−20.00
Quality of Life Index (QLI) – Generic III Version	Overall	17.98	26.59	47.87
	Health/Function	15.65	29.15	86.24
	Socioeconomic	19.75	21.94	11.08
	Psych/Spiritual	19.75	21.94	11.08
	Family	20.63	26.63	29.09

WBLT- Weight-Bearing Lunge Test, BORG CR 10 – Borg Category-Ratio scale, QLI – Quality of Life Index, kg – kilograms, cm – centimeters

during, and after training, the patient was asked about pain complaints by the attending physiotherapist. At each time point, the patient reported no pain. To evaluate changes in variables, we used the Minimal Detectable Change (MDC) of individual tests in the available test–retest studies. The main data of the assessments carried out in this case report are shown in Tables 2 and Table 3.

After completing the exercise program, the patient gained body mass (i.e. fat and muscle) with a simultaneous increase in the circumference of both thighs (L + 2.5 cm, R + 2.5 cm) and biceps (L + 1.5 cm, R + 1 cm). The Quality of Life Index increased by approximately 48%, where the Health and Function subscale showed an increase of 86%.

The dynamic balance of the upper and lower limbs showed a tendency to improve; however, only the direction of medialis in the UQ Y-Balance test increased from 93.0% to 103.8% over the minimal detectable change (MDC) (Shaffer et al., 2013). Results of the FMS test showed an improvement only in the push-up task from 2 points to 3 points, and the total score increased from 16 to 17. The power of lower limbs has improved by 4.98 cm in jump height. It is worth mentioning that MDC from Hilmersson, Edvardsson, and Tornberg (2015) study was set at 3.9 cm. The ankle dorsiflexion range of movement decreased from 3.5 cm to 3 cm for the right ankle, and from 9 cm to 7 cm for the left ankle in the WBLT.

Discussion

Exercises for hemophilia patients are an important part of their rehabilitation process and overall performance. They improve the joint condition, reduce pain, increase the range of motion, and also have a positive impact on the quality of life of the individual (Schäfer et al., 2016). In addition, appropriate strengthening exercises can improve movement patterns, including lower limb alignment (Wilczyński, Zorena, and Ślęzak, 2020).

However, there is no scientific research on strength training on patients with severe type A hemophilia complicated by an inhibitor. Also, it is worth noting that the new treatment Mastarcimab is still in clinical trials. Thus, it is first case report in which a hemophilia patient undergoes an intensive strengthening program during treatment with a new drug. The patient showed good tolerance to intensive exercises. Particularly noteworthy is the Nordic Hamstring exercise, for which supramaximal contraction is required. This exercise is characterized by high muscle soreness (DOMS) (Cuthbert et al., 2020). Despite the average results of the Borg CR10 scale in the first week of exercise (7.5/10 – described as very heavy) the patient tolerated effort well.

The quality of life index increased significantly, mainly in terms of health and function. Such a change may have occurred due to both the new prophylactic medication and the exercise program. Hemophilic arthropathy harms joint stability and muscle strength in PWH (Luck et al.,

Figure 1. Exercise program training – Upper limbs. 1A/1B – Dumbbell bench press while lying on a Swiss ball, 2A/2B – Dumbbells curl sitting on the bench, 3A/3B – Dumbbells tricep extension lying on a horizontal bench.

2004). Previous studies on the effects of strength training (Hilberg, 2018) and balance exercises (Hill et al., 2010) showed improvement in patients balance results with PWH without inhibitors. The patient showed a trend in improvement in dynamic upper limb balance after the exercise program, but a significant improvement only in the medial direction (i.e. over MDC). In addition, the patient showed improvement in the CMJ test indicating increased lower limb power. Despite restriction of movement in the ankle joint and requiring a minimum of 120 degrees of flexion in the knee joint during the CMJ test the patient performed the jump tests correctly. The CMJ test is a demanding movement task and can potentially increase the risk of bleeding or cause joint pain in people with hemophilia especially those with an inhibitor. We performed five attempts at this test (i.e. three attempts are usual in the literature) which may have increased the mentioned risk. This should be considered when using this test in future studies in patients having joint arthropathy.

Furthermore, the patient showed an increase in the biceps and high circumferences in both left and right limbs. This could have been due to both an increase in muscle mass and fat percentage. Unfortunately, we did not investigate the energy supply from the patient's diet, which may have influenced the above results. Given the nature of this case report, the findings cannot be generalized to other patients and must be carefully considered in conjunction with the affect of the study drug. It would be inappropriate to consider a similar training protocol for other patients with hemophlia and an inhibitor at this time, Randomized clinical trials are needed before drawing conclusions about an exercise program for patients with hemophilia and an inhibitor

Figure 2. Exercise program training – Lower limbs and torso. 4A/4B – Nordic hamstring curl, 5A/5B – Romanian barbell deadlift, 6A/6B – Bulgarian squat with a dumbbell, 7A/7B – Pulling the knees to the abdomen in the support on the forearms with feet on TRX (Total Body Resistance Exercise).

We also must consider that our patient showed decreased range of motion in the ankle joints, mainly in the left ankle joint. The probable cause could be increased ankle muscle tone as a result of training aimed at improving the strength and stability of the limbs and torso. However, we cannot rule out minor bleeding episode, which was not observed by either the patient or the investigators, and which may have affected the decrease in ankle ROM. It should be noted that the reduction in range was not significant (i.e. difference – 0.5 cm right ankle and 2 cm left ankle).

It is worth noting that in this commitment to training, previous physical activity, early detection of the inhibitor, prompt availability of preventive treatment and relatively preserved joint condition allowed for an exercise program based on heavy strength tasks. Patients with an inhibitor showing greater level of joint arthropathy and pain may not be able to perform the program presented in our case.

The main finding of this case report is that the patient did not show any bleeding episodes. There is a high probability that this was due to the new medication. In addition, and related to the above better hemostatic control of the patient's bleeding disorder in addition to the strengthening exercises may have been responsible for the improved quality of life and power of the lower limbs. The new prophylactic treatment can give new

Table 3. Comparison of Y-Balance Test, CMJ, and FMS between pre-training and post-training.

Variables	Measurment	Assesment			
		Pre-training	Post-training	Differences	MDC[a]
LQ Y-Balance Test (LL%)	ANT	52.63	55.52	2.89	8.7
	PL	95.78	103.94	8.16	11.5
	PM	87.89	91.31	3.42	10.3
	COMPOSITE	78.77	83.59	4.82	24.8
UQ Y-Balance Test (LL%)	MED	93.00	103.76	10.76*	8.1
	INF	77.96	81.18	3.22	6.1
	SUP	70.43	72.42	1.99	6.4
	COMPOSITE	80.46	85.79	5.33	-
Counter Movement Jump – CMJ (cm)	Jump height	28.02	33	4.98*	3.9
Functional Movement Screen – FMS (pts)	Total final score (max 21 pts)	16	17	1	2

[a]Minimal Detectable Change (MDC) for the YBT (Shaffer et al., 2013); CMJ (Hilmersson, Edvardsson, and Tornberg, 2015); and FMS (Teyhen et al., 2012); LL % – Lower Limb percentage, LQ – Lower Quarter, UQ – Upper Quarter, ANT – Anterior, PL – Posterolateral, PM – Posteromedial, MED – Medialis, INF – Inferolateral, SUP – Superolateral, YBT – Y-Balance Test, CMJ – Counter Movement Jump, FMS – Functional Movement Screen, cm – centimeters

opportunities to work on an exercise program with hemophilia patients with an inhibitor.

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