

## REVIEW ARTICLE

# Exercise and sport in the treatment of haemophilic patients: a systematic review

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**Summary.** In haemophiliacs, the physical condition, muscular strength, aerobic resistance, anaerobic resistance and proprioception have all diminished. Muscle atrophy and instability, being more vulnerable to stressful motor demands, increase the risk of lesion and establish a vicious circle that is hard to break: pain, immobility, atrophy, articular instability and repeated bleeding episodes. In haemophilia, physical and/or sporting activities were not recommended until the seventies. Nowadays, the overall policy is to recommend certain physical activities, especially swimming, to improve the patient's quality of life, thanks to prophylaxis programmes. The objective of this study is to perform a systematic review of the exercise and sporting activities recommended for haemophiliacs. Experimental and observational studies and clinical assays about rehabilitation for haemophiliacs with exercise and sporting activities have been included. The relevant studies were

identified in Medline, Cinahl, Embase and SportDiscus, and key words were: haemophilia, exercise and sport (with no language restrictions). Works were independently analysed by reviewers and the following were identified: of 3603 studies, 103 were included in this review: 29 (28.15%) were experimental, 27 (26.21%) were observational and 47 (45.63%) were clinical. Physiotherapy, physical activity and sport are basic elements to improve quality of life and the physical condition, increase strength and resistance and to reduce the risk of musculoskeletal lesions and to prevent haemophilic atrophy. In general, professionals in haemophilia believe that regular exercise and rehabilitation with physiotherapy is fundamental, particularly in countries where replacement therapy is not readily available.

**Keywords:** exercise, haemophilia, physiotherapy, rehabilitation, sport

## Introduction

Haemarthrosis, sinovitis and bruising are the most frequent problems for the haemophilic patient, and are frequently accompanied by marked muscle atrophy around the joint, possibly because of a poor use of this muscle [1]. All this inevitably involves a degenerative process of the joint, known as haemophilic arthropathy, where haemorrhaging in joints or intra-articular bleeding represent between 65% and

80% of all bleeding episodes [2], of which 80% are predominantly localized in elbows, ankles and knees [3].

Weakness appearing in joints causes instability and makes patients more vulnerable to stressful situations involving motor reactions that progressively lead to atrophy, which in turn involve a higher degree of instability and the risk of lesions, thus creating a vicious circle that is difficult to solve, one of pain, immobility, atrophy, instability of joints and new bleeding episodes [4], all of which may even result in total invalidity [3]. In addition, haemorrhages usually mean having to keep the affected limb immobile, which also damages the patient's proprioceptive capacity [5].

The basic strategy to fight haemophilia-derived musculoskeletal problems has, until quite recently,

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Accepted after revision 25 July 2008

focused on improving existing pharmacological treatments. In this way, the implementation of prophylaxis programmes by means of two- or three-weekly infusions of factor concentrates has permitted the incidence of haemorrhaging to lower. Consequently, the haemophiliac's quality of life has improved [6]. This major change in the patient's condition has led many experts on the theme to seek complementary solutions which could enhance various aspects of the physical condition. Therefore, opinions in favour of practicing physical activity and performing exercise appeared in the scientific literature as from the seventies [7].

Despite the importance of the theme, certain controversy exists as to the type of activities that these patients should do, bearing in mind their special characteristics. The various methodological approaches used to study this theme are delaying the appearance of clinical evidence which could back the decisions made by the professionals in charge of the patients' welfare.

The main proposal of this review is to analyse the existing literature on the physical exercise and the sports that haemophilic patients perform by presenting the information to facilitate consultations in a brief and structured fashion, and by highlighting the main findings to date. Likewise, attention has been paid to the existing limitations in previous works by proposing new lines of action which may facilitate future actions.

## Methods

### *Inclusion and exclusion criteria*

In order to identify the relevant articles which should be included in this review, those which fulfilled the following criteria were included: (i) those works whose titles and/or abstracts explicitly cited activities aimed at improving the physical condition and whose objectives involved the rehabilitation of musculoskeletal pathologies, or at simply developing the capacities of patients with haemophilia A or B. Likewise, those works which presented descriptive data of the patient's physical condition before and after an intervention were also taken into account; (ii) works presented in the form of articles in journals, abstracts of presentations at WFH Congresses and musculoskeletal committee meetings, other meeting reports; (iii) works whose methodological design was experimental, observational and/or included clinical experiences. Furthermore, those reviews with a section related to exercise, sport and improving the physical condition

were also included; (iv) only those studies whose title and abstract were presented in English were analysed. Nonetheless, no language used in the main text was restricted.

Studies which only focused on rehabilitation treatments that did not imply physical activity as a means of improving the patient's physical condition, exercise and/or sport, were excluded (e.g. surgical interventions). Those articles in which the patients presented other non-haemophilic coagulopathies were also excluded. Finally, books or chapters of books which covered some of the target themes of the study were not analysed although they appear in bibliographic reviews.

### *Search strategy and quality assessment of the methodology*

Relevant studies were identified through searches in the following databases: Medline, Cinahl, Embase and SportDiscus from the time of their origin until April 2007. The key words used in the searches were: haemophilia, haemophilia, exercise and sport. Studies were identified by electronic searches until no further work was found. All works selected by the search strategy were studied independently by reviewers, a process which validated the eligibility of the studies selected. Any ambiguity was solved after all the reviewers had reached a consensus.

The search strategy for all databases was: (i) Haemophilia OR Haemophilia AND Exercise; (ii) Haemophilia OR Haemophilia AND Sport.

### *Data recovery*

Data recovery was first performed by separating works into three clearly different groups: experimental studies, observational studies and clinical trials. Subsequently, the main data were recovered independently (subjects, intervention, results) from the various experimental and observational study types using standard data recovery methods. The more characteristic data of the experimental studies are summarized in chronological order in Tables 1, 2 and 3. Given their variability, the various observational studies are grouped in accordance with the assessment method used (Table 4). With regard to clinical trials, the reviewers proceeded in an equal fashion, i.e. they collected all the conclusions, advice and guidance from the authors' works in relation to rehabilitation through exercise and/or sport from similar subject areas which could be of interest for haemophiliacs. Table 5 shows the subject groupings of clinical trial studies.

**Table 1.** Characteristics of the experimental studies of the year 2006 included in the review.

Authors	Participants	Intervention	Results
Bernades <i>et al.</i> (2006)	25 HP	Football 3 times a week (1 h)	General improvements associated with sport
Bernades <i>et al.</i> (2006)	7 HP (babies)	Swimming twice a week (30 min) 10 months	Positive progress made in motor aspects
Broderick <i>et al.</i> (2006)	70 HP	(a) Series of strength resistance exercises. Three series of 8–12 repetitions 20 min + aerobic training at 60–70% max. 30 min two sessions per week for 1 h for 12 weeks (b) No training	Available in 2008
Calefi <i>et al.</i> (2006a)	13 HP	(a) Postsynovectomy physical exercises (b) No postsurgery exercises	(a) Lower impact of haemarthrosis
Franco <i>et al.</i> (2006b)	HPA 20 years old Case Study	Aquatic exercises using the 'Halliwick method'	Improvements in postural balance
Franco <i>et al.</i> (2006a)	HP after synovectomy	Strength exercises, ROM and proprioception in lower limbs	A decrease of 68% of haemarthrosis in the knee
Harris and Boggio (2006)	13 HP 33 CG	Strength exercises with weights, swimming, cycling, martial arts, golf, walks, basketball, yoga thrice per week, 30' minimum	Improvement in the ROM in most of the 10 joints assessed
Milanovic <i>et al.</i> (2006)	37 HP	General and electrotherapy physical exercises for 3 years	Improvement of force Improvement to the ROM
Querol <i>et al.</i> (2006)	10 HPA severe 10 CG	Force with muscular electrostimulation on quadriceps femoris; 18 sessions at three sessions per week of 30' for 6 weeks	Improved isometric strength: left leg 13.8%; right leg 17.1%; hypertrophic improvement: 24.34%
Schlenkrich <i>et al.</i> (2006)	23 HPA and HPB	Physical exercises done at home for 2 years	Improved overall physical condition
Stolfa <i>et al.</i> (2006)	57 HP (a) 40 with haemorrhages (b) 17 without haemorrhages	(a) Physical therapeutical exercises for 6 months (b) No exercises	(a, b) Optimal conservation of the ROM and daily abilities, 100%

HP, haemophilic patients; CG, control group; HPA, patients with haemophilia A; HPB, patients with haemophilia B; ROM, range of movements.

## Results

The search process identified 3603 studies, of which 103 met the inclusion criteria to be considered in this review. Of these selected studies, 29 (28.15%) were experimental, 27 (26.21%) observational and 47 (45.63%) were clinical trials.

Of the experimental and observational studies analysed, only nine original articles indicated the type of treatment that the patients were following during the experimentation period (specifically, six studies appear in which the patients' treatment appears 'on prophylaxis', and there were three studies with patients on the 'treatment-on-demand'). Surprisingly, a further 13 works did not describe the patients' treatment type. Additionally, there is a high percentage of works included in our review which have been published as abstracts, most of which do

not state the haematological treatment administered. This is most probably because of the limited space that this format entails.

As we have mentioned in the Introduction of this review, the appearance of the replacement factor treatment, in both prophylaxis and on-demand types, has been a huge step forward for the haemophilic patient's quality of life. Thus, the fact that a large number of authors do not state this variable in their works makes the interpretation of their results very difficult. We believe that it is important to indicate the characteristics, type and the dosage per unit/weight and the frequency of the treatment administered. In this way, we could not only compare the results of the various authors more easily, but we could also accurately establish the possible statistical relationships between aspects of the physical condition and forms of treatment.

**Table 2.** Characteristics of the experimental studies for the years 2005 to 2002 included in the review.

Authors	Participants	Intervention	Results
Stephensen <i>et al.</i> (2005)	1 HPA severe after knee synovectomy	(Preoperative) Strength exercises in gyms and swimming pools. Twice a week for 6 months (Postoperative) Isometric force exercises, quadriceps and walking for 6 months	Improvement in muscle strength Improvement in the ROM Rapid recovery of muscle functions
Bispo <i>et al.</i> (2004)	30 HP	Strength exercises for 1 year	Improvement of force or of the ROM Reduced bleedings in joints
Hartl <i>et al.</i> (2004)	10 HP	Exercise programme 'Haemofit Styria' for 4 years	A 58% improvement in joint mobility Significant improvement of force
Kahla <i>et al.</i> (2004)	25 HP	Muscular strengthening and proprioception exercises	Recommended physical therapy
Hilberg <i>et al.</i> (2003)	9 HPA severe (a) 8 CG healthy and active (b) 11 CG HP passive (c)	(a, b) Strength with low resistance on the extensor-flexor knee muscles (20–25 rep) for 6 months Twice a week for 120 min and proprioception training (c) Control situation	Improved isometric strength: (a) Extensor muscles 34%; flexor muscles 29% (b) Extensor muscles 20%; flexor muscles 28% (c) No significant changes (a, b) Improved proprioception
Carvalho <i>et al.</i> (2002)	10 HP	General physical exercises for five sessions a week for 40'; 18 sessions	Non-risk exercise programme for bleeding or traumas; may be done at home
Erban <i>et al.</i> (2002)	130 HP Retrospective	Hydrotherapy and physical exercise; an average of 14–18 days	Positive results for 82.5% Reduction of the average number of target joints
Fondanesche and Schved (2002)	8 HP	Specific physical training for sailing and snowmobiles	Improved physical and psychological condition with complete safety
Mazzariol <i>et al.</i> (2002)	10 HP	General exercises for postural training, coordination and proprioception and kinetics therapy for two sessions a week for 1 h for 3 months	Improvement of the ROM Less functional restrictions
Mulvany <i>et al.</i> (2002)	14 HP	Individual physical exercises for 6 weeks	Improvements in the ROM (joints), force and walking
Orr (2002)	10 HP Randomly selected in two groups (a, b)	(a) Hydrotherapy (b) Physical exercises at home for 8 weeks	Less bleeding episodes in joints and less pain
Terezinha and Guilherme (2002)	20 HP	Electrostimulation on the quadriceps femoris; three sessions a week for 30' for 1 year	Improvements in synovitis and functional rehabilitation
Tiktinsky <i>et al.</i> (2002)	Patients 1 and 2: prospective (a) patients 3, 4 and 5: retrospective (b)	(a) Strength with low resistance in wrists and ankles for 1–2 years (b) Same exercise programme for 11–21 years (a, b) Three times a week for 45–60'	(a, b) Increase of muscle force (a) Less frequent bleeding episodes from 2–3 to 1–2 a week (b) Less frequent bleeding episodes up to 2–4 a month

HP, haemophilic patients; CG, control group; HPA, patients with haemophilia A; HPB, patients with haemophilia B; ROM, range of movements.

Prior to the detailed analysis of the various articles, it is essential to indicate two general aspects that we have observed at a methodological level: (i) there are very few experimental works which use a control group in their design (see Tables 1, 2 and 3); (ii) the measurement methods and techniques used vary greatly (see Table 4). Obviously, the low prevalence of the disease and the wide geographic distribution of the patients make both aspects more difficult. Nonetheless, encouraging multi-centre projects could

evidently help improve this situation and would lead to a better control of the experiments. We now go on to present the main findings of this review which we arrange in two blocks and whose common link is haemophilia: works related to exercise and sport.

#### *Exercise and haemophilia*

The analysis performed in our review of the articles reveals that there is a large number of works which

**Table 3.** Characteristics of the experimental studies for the years 1999 to 1982 included in the review.

Authors	Participants	Intervention	Results
Heijnen and De Kleijn (1999)	20 HP severe	Strength exercises for the extensor and flexor muscles, abductors and the hip extensor muscles. Walking and postural training for 4 weeks	Improved joint contractions
Greenan-Fowler <i>et al.</i> (1987)	10 HP severe	Strength exercises for the extensor and flexor muscles in elbows, knees and ankles. Cycling, swimming and running for 1 year; thrice a week minimum	Of those who wished to practice exercise for 12 months, 94% did so for the first 3 months, and 84–60% did so for the entire year
Pelletier <i>et al.</i> (1987)	1 HPA severe	Isometric strength at 60% of the maximum voluntary contraction on the quadriceps femoris for 3 weeks	Improved isometric strength: 40–70%
Greene and Strickler (1983)	32 HP severe	Isokinetic strength of the flexor and extensor muscles in the knee	Improved isokinetic strength
Koch <i>et al.</i> (1982)	2 HP severe	Intensive dynamic strength training programme	Improved strength and ROM as well as decreased bleeding frequency

HP, haemophilic patients; CG, control group; HPA, patients with haemophilia A; HPB, patients with haemophilia B; ROM, range of movements.

**Table 4.** Instruments used to assess the observational studies included in the review.

Assessment instrument used	References
Questionnaire	Fiala <i>et al.</i> (2003), Heijnen <i>et al.</i> (2000), Van Genderen <i>et al.</i> (2002), Dourado (2004), Fribaud <i>et al.</i> (2002), Nazzaro (2002), Von Mackensen <i>et al.</i> (2006), Heijnen <i>et al.</i> (2004), Egan <i>et al.</i> (2006), Schved <i>et al.</i> (2006)
Combination of questionnaire and maximum oxygen volume	Van der Net <i>et al.</i> (2006)
Combination of questionnaire and isokinetic strength	Ohmine <i>et al.</i> (2006)
Isometric or isokinetic strength	González <i>et al.</i> (2007), Falk <i>et al.</i> (2005), Pietri <i>et al.</i> (1992)
Combination of force and proprioception	Hilberg <i>et al.</i> (2001), Krishnamurthy <i>et al.</i> (2004), Tiktinsky <i>et al.</i> (2006)
Combination of force and ROM	Schoenmakers <i>et al.</i> (2001), Ohno <i>et al.</i> (2002), Kwon and Kim (2006)
Combination of force and resistance	Falk <i>et al.</i> (2000), Koch <i>et al.</i> (1984), Seuser (2004)
Combination of muscle force and muscular trophism	Querol <i>et al.</i> (2004)
Bone mineral density	Barnes <i>et al.</i> (2004)
Body composition and daily energy used	Wittmeier and Kriellaars (2006)

ROM, range of movements.

associate the patients' degenerative joint processes with lack of physical activity and exercise. It is usual to find works which relate a worse physical condition (e.g. less capacity to produce force, muscular inabilities, etc.) with a higher risk of muscle and joint lesions [8–11].

More often than not, the physical problems that these patients face make it difficult for them to perform everyday activities normally [12,13]. These problems are intensified in elderly haemophiliacs who also suffer arthritis and are at risk of falls with all the obvious consequences involved. Therefore, there is a need to devise health programmes to reduce the physical and mental impact of ageing in such patients [14].

#### *Preventing haemophilic arthropathy through exercise*

Some authors believe that it is essential to teach young people the importance of physical exercise to prevent musculoskeletal problems [15]. With this practice, it is possible for young haemophiliacs, with healthy joints and whose everyday activities are not restricted, to enjoy a similar physical condition and aerobic capacity, and lifestyles that are as active as those of their healthy counterparts [16–18].

Young haemophiliacs are good at keeping to certain exercises [19]. Despite this, studies have appeared when this physical intervention is not

**Table 5.** Groups of clinical experiments included in the review, arranged according to subject matters.

Subject matters	References
General recommendations about doing physical exercises	Buzzard (1998), Querol <i>et al.</i> (2001), Street <i>et al.</i> (2006), Ribbons and Rees (1999), Buzzard (1999), Beeton <i>et al.</i> (1998), De Kleijn <i>et al.</i> (2004), Buzzard and Jones (1988), Gilbert and Radomslis (1999), Gilbert (2000), Miller <i>et al.</i> (1997), Heijnen (2002), Cassis (2004), Stephensen <i>et al.</i> (2004), Dovc (2004), Buzzard (2002), Tlacuilo-Parra <i>et al.</i> (2006), Lobet <i>et al.</i> (2006), Buzzard (2006), Dalzell (2006)
General recommendations about participating in physical and/or sporting activities	Mulder <i>et al.</i> (2004), Fiala <i>et al.</i> (2002), McLain and Heldrich (1990), De Toni <i>et al.</i> (1985), Querol <i>et al.</i> (2002), Aydogdu <i>et al.</i> (2001), Beeton (2001), Danusantoso and Heijnen (2001), Fondanesche and Schved (2006), Battistella (1998), Weigel and Carlson (1975), Federici and Mari (1980), Buzzard (1996), Dalzell and Poulsen (2004), Schved (2004), Narayan (2006), Kalnins (2006), Calefi <i>et al.</i> (2006b)
Recommendations for developing countries	Jilson (2002), Narayan (2004), Bolton-Maggs (2004), Chen <i>et al.</i> (2006)
Experiences of physical activities during sport camps	Tiktinsky <i>et al.</i> (2004), Sohail <i>et al.</i> (2002), Borel-Derlon <i>et al.</i> (2002), Garrido (2002), Guillon <i>et al.</i> (2004)

performed at early ages, which demonstrate smaller muscle strength and anaerobic resistance in comparison with healthy subjects [4,20,21].

In addition, it should be pointed out that young haemophiliacs who have not participated in physical education at an early age have low bone mineral density (BMD), which is a risk factor for osteoporosis later on in life [22]. In order to avoid this situation, weight-bearing exercises in youth are suggested which ensure these patients have the adequate BMD to avoid subsequent additional problems [22,23]. Furthermore, another additional problem is the increasing obesity phenomenon which complicates the problem because of its influence on the mechanics of these patients' joints [24].

Finally, we wish to point out that young people who present a neuromuscular disorder in their limbs previously experience radiological alterations, and early diagnosis could lead to strength training programmes which presumably would avoid problems [25].

The physical condition of adult haemophilic patients has been seen to be significantly inferior to that of the healthy subjects as far as muscle strength, proprioception and aerobic or anaerobic resistance are concerned [8,10,20,25–27]. This loss of strength and proprioception in haemophilic patients may be related to the frequency of bleeding [28].

Consequently, it is worth increasing the patients' physical capacities so that their quality of life may improve [29,30]. For this reason, some authors in the last decade have started recommending training to improve proprioception [26,27] and muscle strength as a therapeutic method [9,31–35].

Proprioception seems to be clearly compromised by the severe arthropathies in the joints of these patients [36]. Therefore, specialized training for global proprioception would be helpful to compensate for existing proprioceptive deficits as the proprioceptive performance of such patients is significantly damaged in relation to healthy subjects [26]. If to this we add a loss of proprioceptive responses with age, and the fact that most bleeding in joints occurs in the lower limbs, it seems to be vitally important for these patients to receive a suitable, early proprioceptive therapy as a measure to prevent haemarthrosis. Nonetheless, no specific proposals are found in the literature on such training. Indeed, only one author proposed controlled training for improvement purposes [27]. It is necessary that future works cover this matter in detail.

#### *Strength training in the haemophilic patient*

Many authors have pointed out the potential benefits of strength training [1,15,25,37]. Evidently, strength training may be performed in various ways. Some authors have undertaken studies, which are based on resistance training with light loads, and achieved increased forces among patients [1,7], especially in the muscle groups which surround the target joint. Besides, a reduction of the frequency and severity of haemorrhages and their associated pains has occasionally been verified [1]. Conversely, other authors have opted for strength training for hypertrophy with medium to heavy weights, and they obtained similar results for strength improvement [32,35], which is

occasionally accompanied with greater muscle mass [35].

The authors of a very limited number of notable works already featured in the literature on this theme submitted their patients to various types of training. They may be summarized as follows:

- Koch *et al.* [38]; intensive dynamic strength training programme;
- Greene and Strickler [31]; isokinetic strength training programme;
- Pelletier *et al.* [32]; isometric strength training;
- Tiktinsky *et al.* [1]; prospective and retroprospective pilot study on patients who underwent strength training with light weights;
- Hilberg *et al.* [7]; isometric strength training and proprioceptive training;
- Querol *et al.* [35]; strength training with muscle electrostimulation.

As we can see, in recent years we have gone from a mere description of interventions to a more in-depth description of the means and methods used in the experiments [39]. Despite the tremendous efforts made however, these present significant limitations as to the type of dependent variables selected. Our viewpoint is that, out of necessity, the works about strength have to collect data on the changes brought about on the nervous system activity and muscle trophism which have been brought about through strength training. These variables are essential to make progress in the understanding of the effects of strength training on the organism of haemophilic patients. Although there has been the odd work which has attempted to include both parameters in the same study [35], the reduced sample size of the experiment does not allow drawing definite conclusions. Therefore, it is fundamental that neurophysiological and image techniques are used in all the studies which seek to significantly increase strength.

Another important drawback in the works analysed on this subject is the lack of clarity when describing training protocols. In this sense, many works exist in which a specific type of training cannot be separated. In such cases, training based on regular, varied physical exercise is mentioned which might comprise of strength and resistance exercises, and sports. Some of them mention improvements of force and/or range in joints [40–50], while others refer to a reduction in either the frequency or severity of bleeding episodes [51–56]. Therefore, regular, varied exercise may help as joint disability is reduced through the strengthening of the muscles and tendons surrounding the joint, thus protecting the joint from haemarthrosis [44,57].

Evidently, and as Falk *et al.* [20] pointed out in 2000, it is necessary to perform future in-depth research work into the effects of strength training. This idea is backed by Hilberg *et al.* [7] in 2003, but they go one step further and tentatively suggest that reducing the substitution prophylaxis with either factor VIII or IX is possible under conditions that improve proprioception and muscle strength, although they also point out that future works in this direction would be useful [7].

*Physical exercise for the treatment of synovitis and postsurgical rehabilitation.* Of course there are also some works which recommend exercise to improve chronic synovitis, and which stress the importance of functional recovery with isometric exercises in the early stages of a lesion, and then progressing towards more dynamical exercises with greater loads throughout the treatment [58–60].

The variety of orthotic aids which are now available may help to improve the quality of life of these patients through the prevention of musculoskeletal lesions [61]. On occasion however, joint damage is so bad that one of the few remaining possibilities is to perform either a synoviorthesis or synovectomy. It is in such cases that different authors recommend physical exercise as a means to prevent future lesions [34,62,63]. In 2001, Querol *et al.* [34] suggested a protocol for postsynoviorthesis rehabilitation in which only isometric contractions of the flexor and extensor muscles involved were performed various times a day during the first 72 h. On the other hand, Stephensen *et al.* [63] in 2005 went beyond the use of postoperative treatment and described a preoperative exercise programme, and a postoperative exercise programme with positive results, which improved the range of movement and muscle strength of the joints where joint function was quickly recovered and the risk of bleeding diminished.

For patients undergoing total joint replacement or arthroplasty, some authors have recently been using flexor–extensor exercises to guarantee the range of movement, and to restore balance through proprioception. All this was alternated with open kinetic chain training to strengthen muscles [64–66].

Nonetheless, and despite the aforementioned exercise programmes, specific protocols of action are not described in the literature should any bleeding of the joints appear after these physical treatments [67].

### *Sport and haemophilia*

A dispute about the sport activity to be practiced by haemophiliacs continued until the seventies [68].

Even today the importance of a sport-specific therapy as an integral element of haemophilia treatment is still not openly recognized [69]. To date however, we can see how a large majority of authors, whose works cover sport among haemophiliacs, focus on which sport activities should be recommended and which should be ruled out, arguments which are probably fuelled by the fear of a risk of haemorrhaging.

*Sports classification for haemophilic patients.* One of the most widely used classifications to prescribe sport in haemophilic patients is that of the American Pediatric Society, which is known as the APS list. It establishes three groups of sports depending on the risk involved for the patients, classified as high, medium and low (e.g. rugby, skiing and archery).

Therefore in 1990, authors like McLain *et al.* [70] who followed the APS classification, traditionally suggested that haemophiliacs did not practice any 'contact sport' but always considered the severity of the disease. They suggested that these patients could practice 'non-contact' sports, or at the most, those termed as 'restricted contact' sports, such as cross-country skiing, of which some referenced works exist [71]. However, there are references about haemophiliacs who practice contact sports for years, which the APS classify as high-risk sports [72,73], and even adventure sports [41,74], with no bleeding problems.

In recent years however, modifications of various types emerge in this classification, and there are even new classifications. The authors who have dealt with this matter suggest increasing categories and adopting new criteria [68,75–77]. One first important point that these works propose is that the decision-making for each patient becomes more individual in terms of their clinical status. Besides, more complex parameters are beginning to be used to classify sports whose classification is based on biomechanical aspects, the level of difficulty in learning the sport and on the risk of lesions. Therefore, it is necessary to not only study the risk the sport involves but to accomplish a link between the patient in question and their sporting activity in accordance with both the biomechanical demands of that sport and the patient's physical capacity [75,78].

This new approach to classify sports is particularly important for children as it is long-term prevention work which will allow the formation of motor patterns and will help to improve the young patients' self-image [79,80]. For this to come about however, it is important to minimize the risk of lesions through education with both the individual and the commu-

nity so that haemophiliacs may practice sport with optimum safety [81].

*The physical, psychological and social benefits derived from practicing sport.* Evidently, we should bear in mind that practically no sport exists which is 100% safe as they all have their advantages and disadvantages [82]. If we start from this premise, it is important to highlight that haemophilic sports people have every right to participate in sports which cannot be denied them, providing their physical condition permits this possibility. This point is extremely valid because of the physical, psychological and social benefits a team sport offers them [72]. However, if we take the study by Fiala *et al.* [83] as an example, the number of coaches in the first American football division who allowed haemophiliacs to participate decreased as they followed the APS classification, and soccer is a high-risk sport in this list. Presently haemophiliacs participate, but special care plans and prevention measures need to be applied.

Even so, haemophilic patients feel highly attracted to the most popular sports regardless of their level of danger. There is every possibility that, the fact that they feel part of socially relevant activities, they take serious risks [84]. It is necessary to reach a decision among all the parties involved when it comes to recommending patients about which sport to practice. The doctor will have to consider attractive alternatives which should, in turn, be considered carefully on the patients', parents' and trainers' parts to seek less media-related approaches. Having successfully considered the alternatives, less standard sports may even be an option (e.g. Tai-Chi), but which still provide the patient with many potential benefits [85,86].

Presently, haemophilic patients are well counselled to participate in sports, and to a great extent, this is possible thanks to the sporting activities made available to such patients, even to those who cannot run [87]. This idea is further confirmed with the growing trend of sport camps organized for haemophiliacs. These camps organize a wide range of sports, of which the following are highlighted: swimming and water sports, team sports, badminton, canoe kayak, trekking and beach games [88–92]. One very important aspect of such sport camps is to adapt the activities to the patient's age group [93].

Indeed, it is known that haemophilic patients who practice sport do not easily abandon this activity despite the difficulties they encounter as a result of their pathology [94–96]. In accordance to what has been presented herein, it is clear that sport has



become a positive option for patients, and swimming and water sports are the most cited activities in the literature, and which this group of patients accepts more readily, probably because of the low impact that these activities have on the organism [70,87,97–99]. Nonetheless, and just as McLain *et al.* [70] pointed out in 1990, it is worth mentioning that although swimming is highly recommendable, not only because of the aforementioned benefits, but also because it is an excellent aerobic exercise, it is necessary to stress that it may entail some risk for patients with severe haemophilia who have haemarthrosis in their elbows and shoulders. The author only recommends such sports for patients with mild haemophilia.

The present day uncertainty as to whether water therapies should be applied or not to haemophiliacs is backed by the limited number of experimental and/or observational works in this type of literature. Furthermore, we may also add that none of these works numerically specify the results obtained. Some of the works we found [100–103] only provide a vague description of the programmes used and stress the possible benefits as a result of them. It seems obvious that the methodological treatment of most of the works which have appeared to date do not provide conclusive clinical evidence.

#### *Sport in countries with limited access to pharmacologic treatments*

Last, but not least importantly, we conclude this review by mentioning a group of clinical trials based on the role that exercise and sport play in haemophilia in developing countries. Authors like Bolton *et al.* [104] question the efficiency of physiotherapy in areas where treatment for haemophiliacs is limited, or is simply not available. In this sense, a consensus among the authors apparently exists and they all stress that regular exercise and rehabilitation through physiotherapy are fundamental, particularly in developing countries where factor replacement therapies are not available, as they improve muscle strength and coordination, and they prevent or reduce muscular contractions and pain [59,78,93,105].

Therefore, the alternative is to make rehabilitation as economic as possible so that it is based on exercises which may either be performed at home with educational materials, books or CD demonstrations handed out to haemophilic patients, and which explain the appropriate exercises to strengthen muscles and improve the range of movements in joints [106] or on resources available at a community level

may be used, such as swimming in canals, strength exercises using filled sacks, etc. [107].

## Conclusions

The analysis undertaken in this review reveals a broad spectrum of opinions in favour of haemophilic patients practicing sport and performing physical exercise. Likewise, the large numbers of works on the subject present a whole variety of intervention proposals. The potential benefits cover a very wide range of aspects, such as the physical condition, prevention of musculoskeletal pathologies and even their rehabilitation.

However, serious methodological drawbacks exist in the way these studies have been dealt with, which, in the vast majority of cases, are the result of clinical experiences of doctors in charge of the patients' welfare and not the result of a thorough research process. Even in those works which include an excellent experimental design limitations are encountered in terms of the number of subjects included in the experiments, the use of the material employed and the way the training protocols are described.

## Disclosures

The authors stated that they had no interests which might be perceived as posing a conflict or bias.

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