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# The effect of resistance training on the frequency of bleeding in haemophilia patients: a pilot study

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**Summary.** The benefits and feasibility of progressive resistance training on muscle strength and bleeding profile were studied prospectively in two patients with severe haemophilia. Additionally, retrospective data were collected from three patients who had been training for 11–21 years (one patient for 21 years and two patients for 11 years). Muscle strength increased, especially in muscle groups surrounding the target joints (elbow and knee). Bleeding frequency decreased from 2–3 times per week to 1–2 times per week. Patients who had been training for > 11 years reported bleeding episodes of 2–4 times per month prior to training, but after > 11 years of progressive training a marked decrease in bleeding occurred, as well as a decrease in severity. These data support the importance of resistance training for haemophilia patients, not only for increasing muscle strength, but also for decreasing the frequency and severity of bleeding episodes and the associated pain. A controlled study, with a greater number of patients, is needed to confirm the suggested benefits of resistance training in haemophilia patients.

**Keywords**: exercise, haemophilia, physical activity.

## Introduction

Haemophilic arthropathy is accompanied by marked atrophy of the muscles surrounding the affected joint, due to muscle disuse. Muscle weakness sets in, the joint becomes less stable and more vulnerable to stress, and thus, a vicious circle perpetuates: recurrent haemarthroses lead to worsening of synovitis, which further increases the frequency and severity of haemarthroses, which accelerate arthropathy, reducing muscle development among other effects, and can result in crippling and disability. Prophylactic replacement therapy may prevent this vicious circle, but once a joint is damaged by haemarthroses, deterioration will continue and prophylactic therapy can only slow down, but not prevent, this process. Restriction of physical activity to reduce the risk of trauma and bleeding is common among patients. Moreover, parents of children with haemophilia frequently limit their children's participation in normal activities. Unfortunately, this restriction accelerates muscle atrophy, which leads to lower stability and greater vulnerability of the joints, as well as repeated haemarthroses [1].

A more constructive approach to prevent or delay arthropathy is to strengthen the muscles surrounding the involved joint. Indeed, many physicians recommend physical activity to their haemophilia patients, although the data supporting this recommendation are scarce [2,3]. The most commonly recommended activity is swimming, which is considered to be less traumatic than other sport activities. Swimming, a nonimpact activity, can benefit the cardiovascular system, as well as provide high resistance for the muscles [4–7]. Resistance training is also recommended for muscle strengthening and is claimed to increase joint stability, thereby reducing the risk of spontaneous bleeding episodes [3,10]. In fact, a limited number of early studies have investigated the effect of resistance or isometric exercises in young haemophilia patients [1,11,12]. These studies reported encouraging results of increased muscle strength with no change in, or even reduced frequency of, bleeding episodes. However, the effect on the

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frequency of bleeding in these studies was reported only as the subjective impression of the authors, with no quantitative data.

We have been following up three patients with severe haemophilia who have been training for 11–21 years (one patient for 21 years of resistance training, one patient for 11 years of resistance training, karate and running, and one patient for 11 years of swimming and resistance training). These patients reported bleeding episodes of 2–4 times per month prior to training. Gradually, bleeding episodes became increasingly less frequent, so that at present, after 11–21 years of training, the patients report no spontaneous bleeding. All started training at a young age (9–17 years) and continue to do so into adulthood. One subject became the National Karate Champion at the age of 21 and served in the army as a self-defence instructor and trainer. All subjects report that when they discontinue their training for a period longer than 2 weeks, the bleeding increases in frequency and severity. In view of these promising reports, we prospectively followed up the bleeding profile of two severe haemophilia patients who began progressive resistance training.

## Subjects and methods

### *Prospective study*

Subjects included two cousins with haemophilia (patients 1 and 2), aged 26 and 37 years, with severe haemophilia A (factor VIII [FVIII] < 1%). Both were receiving on-demand replacement therapy (15–20 IU $kg^{-1}$) despite frequent bleeding episodes (2–3 times $week^{-1}$ before training) due to their refusal to receive regular and adequate prophylactic treatment. After obtaining their informed consent to participate in the study, the subjects began resistance training and were followed up prospectively. Patient 1 had not undergone any surgical orthopaedic procedure prior to initiation of training. Patient 2 had undergone a bilateral knee synovectomy when he was 12 years old and a Judet procedure (release of the quadriceps muscle from its origin) at the left knee 6 years later. The subjects' personal characteristics and basic information about their training and follow-up appear in Table 1.

Both patients participated in progressive resistance training 3 times $week^{-1}$, 45–60 min at each session, patient 1 for 2 years, and patient 2 for 1 year. Each session consisted of warm-up and stretching (10–15 min), resistance exercises (30–45 min), and a cool-down period (5–10 min). Warm-up consisted of stretching exercises for the calf muscles, hamstrings, hip adductors, low back muscles, neck rotators and upper arm musculature. Resistance exercises were individualized and included lifting of free weights (small sandbags secured to the wrist or ankle with Velcro bands) added in increments of 0.5 or 1.0 kg, i.e. the programme was designed to be ahigh-repetition–low-resistance programme. Cool-down consisted of stretching exercises, especially for muscles surrounding the target joint.

With patient 1, the most affected joint was the right elbow. Thus, emphasis was placed on strengthening elbow flexors and extensors. Additional exercises included pronation and supination of the forearm, as well as external and internal rotation of the shoulder. The most affected joint in patient 2 was the right knee. Therefore, an emphasis was placed on exercises for the lower extremities. He performed knee flexion and extension in an effort to strengthen the muscles around the knee in particular. He also performed hip flexion, extension, abduction and adduction, as well as plantar flexion and dorsiflexion. All exercises were performed in antigravity positions while the patient was in a seated, or reclined position on the treatment table.

Subjects began the training programme by performing the maximal number of unloaded repetitions. The number of repetitions was increased in each session by up to 10%, until the subjects were able to perform 150 unloaded repetitions. At this stage, resistance of 0.5 or 1.0 kg in free weights was added and the number of repetitions dropped to whatever the subject could perform, usually beginning at 20 repetitions. Subsequently, the progression was as described above, although the number of repetitions was allowed to increase to 250 before adding resistance.

**Table 1.** Subject's physical characteristics and training programme and follow-up.

| Subject no. | Adult height (cm) | Adult weight (kg) | Target joint | Age at start of training (years) | Duration of training (years) | Type of training |
|---|---|---|---|---|---|---|
| 1 | 170 | 65 | Elbow | 26 | 2 | Resistance |
| 2 | 165 | 70 | Knee | 37 | 1 | Resistance |

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*Retrospective study*

Training programmes for the three subjects studied retrospectively span over a period of 11–21 years and are described briefly. Patient 3 began training at the age of 9 years, and has been training for 21 years. He has been engaged in resistance training 3 times week$^{-1}$, although at certain periods he trained 4–5 times week$^{-1}$, for 45 min at each session. Exercise was performed in three sets of 15 repetitions or three sets of eight, six and four repetitions. Exercises were for the upper (e.g. bench press), as well as the lower (e.g. leg press) body.

Patient 4 began training at the age of 12 years, and has been training for 11 years. He has engaged in resistance training for the upper and lower body (3 times week$^{-1}$, 12–15 repetitions of each exercise), as well as running (up to 10 km), interval training, and especially karate training.

Patient 5 began training at the age of 17 years and has been training for 11 years. He swims 3 times week$^{-1}$ for 45 min, and in the past 18 months has also added resistance training (3 times week$^{-1}$) for the upper and lower body.

*Bleeding history*

The patients completed a 'bleeding diary', which included the site of the bleed, its cause (trauma, spontaneous), degree of pain (scored on a 0–5 scale where 0 = no pain and 5 = excruciating pain), limitation in range of motion (extreme, moderate, minimal or none) and the replacement treatment given. The diary was started 6 months before initiation of the training programme and continued throughout the training period.

## Results

Table 2 provides the results of the evaluation of range of motion and manual muscle strength before and after training for patients 1 and 2. In general, the range of motion remained unchanged while muscle strength, as evaluated manually, clearly improved.

A quantitative description of muscle strength performance before and following the training programme in patients 1 and 2 appears in Table 3. The increase in strength is clearly apparent in both subjects.

The bleeding profile before and after training appears in Table 4. At present, the patients continue training and receive 15–20 IU kg$^{-1}$ FVIII concentrate once in 7 (patient 1) or 10 days (patient 2), even in the absence of bleeding, as a partial prophylactic treatment (patients' own decision). Table 3 also includes retrospective data for the subjects who had started training in their youth. Their data were reconstructed from the charts and from memory. It is apparent that frequency of bleeding episodes, as well as pain associated with each episode, decreased by the end of the training programme. It is noteworthy that all subjects reported that cessation of training for a period longer than 2 weeks is associated with increased frequency and severity of bleeding.

## Discussion

Based on evaluation of retrospective data of three patients (Table 4) and a survey of the literature, the present pilot study was designed to evaluate the effect of progressive resistance training on bleeding frequency and severity among adult haemophilia patients.

**Table 2.** Evaluation of Range of Motion (ROM) and Manual Muscle Strength (MMS).

| | | ROM (degrees of motion) | | | MMS* | |
|---|---|---|---|---|---|---|
| Subject no. | Joint | Movement | Pre | Post | Pre | Post |
| 1 | Shoulder | Flexion | 0–170 | 0–180 | Good | Normal |
| | | Abduction | 0–170 | 0–180 | Good | Normal |
| | Elbow | Flexion | 20–135 | 5–145 | Good | Normal |
| | | Extension | −20 | −5 | Good | Normal |
| 2 | Hip | Flexion | 15–110 | 5–115 | Good | Normal |
| | | Extension | −15 | −5 | Good | Normal |
| | | Abduction | 0–30 | 0–35 | Good | Normal |
| | | Adduction | 0–20 | 0–25 | Good | Normal |
| | Knee | Flexion | 15–80 | 10–95 | Good | Normal |
| | | Extension | −15 | −10 | Good | Normal |
| | Ankle | Dorsiflexion | 0–5 | 0–15 | Fair | Good |
| | | Plantarflexion | 0–30 | 0–50 | Good | Normal |

*According to [13].

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**Table 3.** Muscle strength before and following training.

| Subject no. | Exercise | Pretraining | | Post-training | |
|---|---|---|---|---|---|
| | | Repetitions | Resistance (kg) | Repetitions | Resistance (kg) |
| 1 | Shoulder | | | | |
| | Flexion | 20 | 0 | 20 | 3 |
| | Abduction | 20 | 0 | 20 | 3 |
| | Elbow | | | | |
| | Flexion | 30 | 0 | 40 | 0 |
| | Extension | 30 | 0 | 40 | 0 |
| 2 | Hip | | | | |
| | Flexion | 30 | 0 | 100 | 5.5 |
| | Extension | 60 | 0 | 220 | 4 |
| | Abduction | 50 | 0 | 250 | 4 |
| | Adduction | 40 | 0 | 200 | 3 |
| | Knee | | | | |
| | Flexion | 20 | 0 | 100 | 3 |
| | Extension | 30 | 0 | 150 | 3.5 |
| | Ankle | | | | |
| | Plantarflexion | 20 | 0 | 100 | * |
| | Dorsiflexion | 20 | 0 | 100 | 2 |

*Body weight was used as resistance.

**Table 4.** Bleeding profile at the start and end of training.

| Subject no. | Bleeding frequency (times per week) | | Pain* | | ROM limitation | | |
|---|---|---|---|---|---|---|---|
| | Start | End | Start | End | Start | End | Site |
| **Prospective data** | | | | | | | |
| 1 | 2–3 | 1 | 5 | 3 | Extreme | Minimal | Elbow |
| 2 | 2–3 | 1–2 | 5 | 2 | Extreme | Moderate | Knee |
| **Retrospective data** | | | | | | | |
| 3 | 1 | nsb | 4 | 0 | Moderate | None | Knee |
| 4 | 0.5 | nsb | 4 | 0 | Moderate | None | Knee/Ankle |
| 5 | 1–2 | nsb | 4–5 | 1–2 | Moderate–extreme | None | Knee |

*Pain was recorded on a scale of 0–5, with 0 = no pain and 5 = maximal pain. Duration was average length of each bleeding episode. ROM, range of motion; nsb, no spontaneous bleeds.

Two haemophilia patients were studied prospectively before and during 1–2 years of progressive resistance training. The results from these cases suggest beneficial effects of resistance training not only on muscle strength and range of motion, but perhaps more importantly, on the frequency and severity of bleeding episodes, as well as on the pain associated with bleeding. In view of the many beneficial effects associated with physical training in general (e.g. elevated endorphin levels in response to aerobic exercise [1,5]), and resistance training in particular (e.g. elevated levels of anabolic hormones [16]), it is tempting to speculate that some of these effects may be related to the apparent reduction in pain. Despite the fact that only two patients were studied prospectively, this is the first prospective account that suggests a decrease in the frequency and severity of bleeding among haemophilia patients following resistance training. Retrospective data were collected from three other patients who trained over a period of 11–21 years and achieved a state whereby they reported only traumatic bleeding episodes with no spontaneous bleeding. It should be noted that the patients reported in this study began training at different ages (adolescence to adulthood), and that all demonstrated consistent positive outcomes.

Many investigators and physicians recommend exercise in general, and strength development in particular for their young haemophilia patients [2,3]. Haemophilia patients are often characterized by reduced muscle strength and muscle power [14]. The reduced muscle strength can lead to joint instability, which in turn results in decreased ability to withstand stress [17–19]. As muscles become weaker, the joint is less protected, leading to an increase in the frequency and severity of haemarthroses, which lead to chronic synovitis and contribute

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to joint arthropathy. The rationale behind the recommendation for strength development is that an enhancement in muscle strength will increase joint stability, provide protection for the joint, and eventually result in reduced frequency and severity of spontaneous bleeds, thus delaying arthropathy.

Several studies can be found in the literature to support this rationale. Koch *et al.* [11] reported an increase in muscle strength in two haemophilia patients, aged 12 and 14 years, who participated in an aggressive strength-training programme. Precise details of the training programme were not provided, although the authors stated that subjects trained twice daily, isometrically as well as isotonically. The authors noted that strength gains and increased range of motion were accompanied by reduced bleeding frequency, although no data were provided. Additionally, it was noted that when one of the subjects discontinued his training, recurrent haemarthroses were evident. In spite of the small number of subjects, and the fact that only limited details were provided regarding the programme or the bleeding frequency, this was the first report to suggest that resistance training may be beneficial for young haemophilia patients in reducing the frequency of spontaneous bleeds and the joint damage that accompanies these bleeds.

About the same time, Greene and Strickler [1] reported an enhancement in muscle strength among 32 haemophilia patients, ranging from 7 to 51 years of age, after 6 months of strength training. The patients performed isokinetic strengthening exercises for the knee extensors and flexors daily 15 min. No prophylactic treatment was reported. The authors noted that there was no increase in the frequency of haemarthroses during the training period. Rather, patients averaged 3.4 bleeding episodes during the 6 months prior to training and 2.1 episodes during the 6 months of training, again suggesting a decrease in the frequency of bleeding. However, the study was conducted over a relatively short period of time.

Pelletier *et al.* [12] reported a 40–70% strength enhancement in a 12-year-old haemophilia patient suffering from chronic knee arthroses who trained for 3 weeks, performing 10 isometric repetitions of 10 s once daily. The authors noted that there was no increase in haemarthroses or muscle bleeds throughout the training period, although no data were provided.

The present report supports the previous studies and extends them in several ways:

1. The cases described in this report were followed for a relatively long period (6 months of baseline and 1–2 years of training). The retrospective reports also encompass a long period of time (up to 21 years), longer than any previous study investigating the effect of resistance training in haemophilia patients.
2. Precise bleeding histories and diaries were recorded, which quantitatively demonstrate a clear reduction in bleeding frequency during the training period. The reduction in bleeding was accompanied by a clear, recorded reduction in the associated pain. This quantitative report extends previous subjective impressions of no increase, or reduced bleeding frequency with resistance training [1,11,12]. The reduction in pain sensation was associated with the bleeds themselves, but also with other daily activities. An additional factor that may have contributed to the reduced pain is the increased range of motion that accompanied the strength gain. Alternatively, the general decrease in pain raises the possibility that exercise may have a systemic effect on pain reduction.
3. Finally, in extension of previous studies that dealt with target joints in the lower extremity only (which are the most common target joints among haemophilia patients), the present study demonstrates that resistance training can also be beneficial for upper extremity joints. Indeed, if a systemic effect is considered, emphasis on strengthening one region can have overall beneficial effects. The cases described in this report do not clearly indicate such an effect, although the reported reduction in overall pain and discomfort should prompt a more methodical study of a possible systemic effect.

The apparent decrease in the frequency of bleeding episodes is not explained by changes in the circulating levels of FVIII, as these did not change in the patients included in this study. Among healthy individuals, a single bout of exercise results in an increase in circulating FVIII levels. The increase is dependent upon exercise intensity and continues into recovery [20]. However, Koch *et al.* [19] demonstrated no increase in FVIII levels after exercise among severe haemophilia patients.

It is suggested that, as previously mentioned, the increase in strength of the muscles around the involved joints, as well as the increased range of motion, provided enhanced joint stability, and protected the structures and soft tissues contained within and around the joint. The enhanced muscle strength allowed the joint to withstand greater forces acting upon it. This is of great importance in weight-bearing joints, as well as in rotating movements in nonweight-

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bearing joints. Furthermore, we cannot rule out the possibility of hormonal or biochemical changes that occurred during training, and reduced the likelihood of spontaneous bleeding. This possibility is suggested from the reported lag phase between cessation of training to recurrence of haemarthroses; during the first 2 weeks of cessation of training, there is no marked decrease in muscle strength. Therefore, the recurrent haemarthroses may be due to a reduction in hormonal or biochemical substances that are elevated during exercise. However, further studies are required to evaluate this hypothesis.

In view of the findings of this study, it is recommended that haemophilia patients engage in progressive resistance exercises routinely in order to strengthen muscles around target joints, as well as for general body conditioning. The use of a low-resistance–high repetition programme was shown to be a potentially successful method for reducing haemarthroses and associated complications.

Previous reports have demonstrated that reduced physical activity among young haemophilia patients is associated with reduced muscular strength and power [13] and reduced aerobic power [21]. Thus, it is suggested that patients who manage to reduce substantially the severity of spontaneous bleeds also engage in other physical activities. These, in turn, could serve to develop additional components of physical fitness, such as cardiovascular endurance, as well as to possibly enhance self-esteem and self-confidence. Further study on a larger scale, comparing an exercising group to a nonexercising group, is required in order to determine the effect of exercise in general, and resistance training in particular, on the bleeding profile and associated pain in haemophilia patients. Additionally, there is a need for a study of the biochemical or hormonal effects that such exercise has in haemophilia patients.

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