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Quality of life and sports activities in patients with haemophilia

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Summary. Quality of life (QoL) is a multidimensional construct pertaining to an individual's physical, emotional, mental, social and behavioural components of well-being and functioning. QoL can be assessed using both generic and disease-specific instruments. QoL assessment in haemophilia is a relatively new area of study; the first data were published in 1990 using generic QoL questionnaires. Only recently have haemophilia-specific questionnaires been developed, first for children and then for adults. Because sports activities include not only physical and functional aspects, but also have an impact on an individual's self-esteem and social interactions, they should play an essential role in QoL assessment. Until the 1970s, persons with haemophilia were advised to avoid any kind of physical activity because of the risk of bleeds. Nowadays, however, the attitude towards sports for patients with haemophilia has changed, and the World Federation of Haemophilia has formulated recommendations concerning physical activities for patients with haemophilia. Although sports activities are recommended, their importance as an integral element in haemophilia management has not yet been widely recognized. Awareness of the importance of sports activities for this patient group has increased, and several sports projects are ongoing. For example, a twice-annual sports camp for adult patients has been held in Germany for the past 2 years; and physical improvements, QoL and subjective training effects are being measured. Another study is investigating the prevalence of sports activities in Italian children with haemophilia and their attitudes towards sports and is also assessing QoL. In Israel, a study is planned in which haemophilic children will participate in a karate training programme, and a QoL evaluation will be added to the physical assessment. In conclusion, sports activities can improve not only physical well-being, but also the emotional and social well-being of persons with haemophilia and, therefore, should become part of the global approach to haemophilia management.

Keywords: haemophilia, health-related quality of life, sport activities

Introduction

Quality of life (QoL) assessment is considered an important outcome measure in medicine that describes patients' well-being and function, evaluates the effects of treatment intervention and assesses the quality and efficacy of care [1]. The World Health Organization (WHO) definition of *health* (1948) states that 'health is not merely the absence of a disease or infirmity, but the state of the complete physical, mental, and social well-being of a person'. The term *health-related quality of life* (HRQoL) was defined based on the WHO definition as a multidimensional construct pertaining to the physical, emotional, mental, social and behavioural components of well-being and function as perceived by patients and observers [2]. HRQoL is influenced not only by a disease and its treatment, but also by personal characteristics, such as coping or locus of control, by living conditions and by socioeconomic status. According to the literature on various diseases, psychosocial factors that influence QoL include perceived stress and satisfaction with social activities for women with heart disease [3]; social support and health behaviour for patients with

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active tuberculosis [4]; and emotional distress, coping, self-esteem and support from friends for schizophrenic patients [5]. Similar determinants were found for patients with haemophilia: age [6,7]; life satisfaction and social support [8]; and self-esteem, anxiety, social desirability and depression [9].

Since the mid-1980s, instruments that are completed by patients for the assessment of QoL have been developed [10]. These questionnaires give patients an opportunity to report their own experience of functioning and well-being [11]. QoL is measured in terms of generic and disease-specific assessments using validated instruments. Whereas generic instruments can be used irrespective of a specific disease, in patients with different conditions or in the general population, disease-specific measures are developed for patients with specific health conditions, thus providing a clear pattern of their symptoms or impairments [12]. Because the patient's perspective is important, the use of self-rated measures is recommended [13]. In contrast, other rated measures, or 'proxy' measures, are used in young children or in patients unable to answer for themselves. Different instruments are available for children and adults, taking into consideration age and developmental status [14].

Quality of life assessment in haemophilia

Bleeding disorders such as haemophilia and their treatment impact patients' QoL and can affect the everyday life of both patients and their families [6,15]. Although assessment of QoL in patients with haemophilia is important, this field is a relatively new research area in which data were first published in 1990 [16]. Validated generic and haemophilia-specific instruments are necessary for adequate assessment of QoL in this patient population. Since 1990, QoL has been measured using mainly generic questionnaires such as the SF-36 and EQ-5D [17,18]. Only recently have haemophilia-specific questionnaires been developed for children (such as the Haemo-QoL and CHO-KLAT) and for adults (such as the Medtap questionnaire and Haem-A-QoL). Table 1 shows an overview of generic and disease-specific questionnaires that can be used for the assessment of QoL in adult and paediatric patients with haemophilia.

Sport activities in patients with haemophilia

Until the 1970s, it was common practice to discourage persons with haemophilia from any kind of sports because of the risk of bleeds [38]. Today, the attitude towards participation in sports by haemophilic individuals has changed, and physical activity is considered beneficial for this group of patients [39]. The World Federation of Hemophilia (WFH) has formulated recommendations for participation in sports activities by patients with haemophilia [40]. Although sports and physical activities are recommended for haemophilic patients [41], the importance of sports therapy as an integral element in haemophilia treatment has not yet been widely recognized [42]. No true guidelines are available, but the type and severity of haemophilia should be taken into consideration when choosing a sport [43]. Moreover, Buzzard [39] emphasizes that the choice of sport should be made on an individual basis. In other words, patients should not be forced to participate in any kind of sport they do not like. Patients should choose an enjoyable sport so that the physical activity will be most beneficial [44].

The aim of this article was to examine the non-physical benefits of sports activities for patients with haemophilia and how sports can influence their QoL.

Table 1. Overview of QoL instruments for use in patients with haemophilia.

Type of instrument	Target age group	
	Children	Adults
Generic	KINDL [19]	Nottingham Health Profile (NHP) [20]
	CHQ [21]	Quality of Well-being Index (QWB) [22]
	TACQOL [23]	Sickness Impact Profile (SIP) [24]
	PedsQL [25]	SF-36 Health Survey (SF-36) [26]
	–	EQ-5D [27]
	–	WHOQoL Questionnaire [28]
Haemophilia-specific*	Haemo-QoL [29]	Medtap [30]
	CHO-KLAT [31]	Haem-A-QoL [32]
	HemDux [33]	Hemofilia-QoL [34]
	QoL for young children [35]	Hemolatin-QoL [36]
	QUAL-Hemo [37]	QUAL-Hemo [37]

*Only two of the haemophilia-specific quality of life (QoL) instruments include a dimension to assess sports activities: the paediatric instrument Haemo-QoL and the adult instrument Haem-A-QoL.

Role of sport activities for persons with haemophilia

Several studies have been carried out with the aim of understanding the role of physical exercise and sports for persons with haemophilia. Effects were found mainly in physical aspects, such as muscle strength,

but few studies have investigated the impact of sport activities on the psychological aspects of health (see Table 2).

One of the most important non-physical benefits of sports activities related to social aspects (i.e. *socialization*) in terms of the patient feeling that he or she belongs to a group, is socially accepted, is doing something with friends and is not alone. Another important non-physical benefit of sport is *self-esteem*. Being able to participate in sports activities provides the feeling of being like others, including healthy people, and leads to a positive self-assessment that makes the person more self-confident. Body image and body control are also positively influenced by participation in sports activities, and problems related to the health condition can be managed better. Only recently, however, have studies been conducted that investigate QoL in patients with haemophilia that include assessments of the effects of sports activities.

European Haemo-QoL study

The European Haemo-QoL study comprised 339 paediatric patients with severe haemophilia who had no evidence of inhibitors or infection [29]. The mean age of patients was 10 years (range: 4–16). Two-thirds of the patients (n = 212) were receiving prophylaxis treatment; of these, 19.8% were receiving primary prophylaxis. The average number of bleeds per month was 1.

Patients' QoL was assessed using the validated Haemo-QoL questionnaire, which is available in three age-group-specific versions (4–7, 8–12 and 13–16 years). The Haemo-QoL consists of 8–12 domains, depending on the age group version used. All versions include a dimension related to sports and school. The following five statements relate to sports: (i) 'because of haemophilia I had to refrain from sports that I like'; (ii) 'I had to do indoor activities more than other kids because of my haemophilia'; (iii) 'I had to refrain from sports like roller-blading or soccer'; (iv) 'I participated in sports just as much as any other kid'; (v) 'I participated in sports classes at school despite my haemophilia'.

Adolescents were impaired mostly in the dimensions 'friends' and 'perceived support'. Concerning sport, 19.7% always or often had to refrain from sports, and 28.2% always or often had to refrain from roller-blading or soccer. In contrast, 56.5% of participants did as much sporting activity as other kids, and 70.1% always or often participated in sports classes at school.

Italian COCHE study

The Italian COCHE study included 232 adult patients with haemophilia with no evidence of inhibitors [55]. Of these, 201 patients (86.6%) had haemophilia A, 72.4% were affected by severe haemophilia and 15.9% were infected with human immunodeficiency virus (HIV). The mean age was 37 years. One-third of the patients (n = 78) received prophylactic treatment. Patients averaged two bleeds per month.

Quality of life was assessed using the validated Haem-A-QoL questionnaire, which consists of 46 items pertaining to 10 dimensions, including a

Table 2. Studies investigating the effect of sport activities on patients with haemophilia.

Impact	Reference	Study conclusions
Physical	Greene and Strickler [45]	An isokinetic muscle-strengthening programme led to increased muscle strength but not to an increased number of haemorrhagic episodes
	Koch *et al.* [46]	Physical fitness is poor in haemophilic children
	Koch *et al.* [47]	Controlled exercise led to modest improvement in several coagulation parameters in patients with mild or moderate haemophilia
	Pietri [48]	Haemophilic children with unilateral haemarthrosis of the knee have reduced muscle function
	Falk *et al.* [49]	Haemophilic children have poor anaerobic power and lower muscle strength
	Hilberg *et al.* [50]	Muscle strength and proprioception are impaired in adult patients with haemophilia
	Schoenmakers *et al.* [51]	Motor performance and ADLs are comparable in Dutch paediatric patients with haemophilia compared with non-haemophilic peers, but their disease has an impact on pain and can place restrictions on sports activity
	Tiktinsky *et al.* [52]	Resistance training in haemophilic patients increases muscle strength, decreases the frequency and severity of bleeding episodes and reduces pain
	Hilberg [42]	Maximum isometric muscle strength in the legs is increased in haemophilic adults after training
Psychological	Salvini *et al.* [53]	Sports participation increases the level of self-esteem, social adaptation, body image and normal self-assessment and attitude
	de Mondenard [54]	Sport activities influence psychological development and help to showcase abilities, overcome a handicap and master health problems

ADL, activities of daily living.

dimension related to sports and leisure. The following three statements relate to sports: (i) 'I had to refrain from sports that I like'; (ii) 'I had to refrain from soccer' and (iii) 'I did just as much sports as others'.

Compared with children, adult patients were impaired mainly in the dimension 'sport and leisure time'. This study showed that 56.7% of the adult patients always or often had to refrain from sports, and 75.5% always or often had to refrain from soccer. Only 15.4% of the adult patients participated in sports as much as their peers. Compared with children, adults were significantly more impaired in their physical activities ($P < 0.001$).

Hemophilia and Exercise Project (HEP)

As part of the Hemophilia and Exercise Project (HEP), 23 adult haemophilic patients participated in a sport training camp in Germany (http://www.haemophilia-exercise.de). Among these patients, 87% had severe haemophilia, 52.5% were receiving prophylactic treatment and 21.7% were infected with HIV. The mean age was 43 years (range: 26–64). Patients had an average of 1.5 bleeds per month.

In addition to the measurement of objective training effects, such as isometric muscle strength and proprioception, QoL was assessed using the generic SF-36, which comprises 36 items representing eight dimensions (physical functioning, role limitation because of physical health problems, bodily pain, general health perception, vitality, role limitation because of emotional problems, social functioning and general mental health). The SF-36 results can be consolidated into two summary scales: the Physical Component Summary (PCS) and the Mental Component Summary (MCS). Subjective training effects were also measured using a newly developed sport-specific questionnaire (HEP-Test) [56], which comprises 23 items pertaining to five domains (mobility, coordination, endurance, perception, general) and a total score (HEP-Total).

The results show that patients were impaired mainly in the dimension 'general aspects' and 'coordination'. To evaluate the extent to which physical activity impacts patients' QoL, regression analysis was performed considering the HEP-Total as an indicator of physical activity. The variance of QoL could be explained by the HEP-Test as follows: 52.8% of the variance in the PCS of the SF-36 could be due to sports activity (beta: 0.727, $T = 4.850$, $P < 0.001$), whereas 18.4% of the MCS could be attributed to sport activity (beta: 0.429, $T = 2.174$, $P < 0.041$).

Conclusions and outlook

Quality of life in patients with haemophilia is influenced not only by clinical aspects, such as treatment regimen [6] and bleeding frequency [57], but also by psychosocial variables, such as social support [8] and self-esteem [9]. Sports activities benefit patients in physical aspects such as muscle strength [42] and fitness [46], and they result in the protection of joints [39], prevention of bleeds [58] and reduced disability. However, sports activities also have an important effect on psychological aspects, such as self-esteem and social adaptation [53], as well as on the psychological development of haemophilic children [54]. Therefore, the measurement of sport activities should be combined with QoL assessment. Studies have demonstrated that QoL in persons with haemophilia is impaired by limitations in sports activities.

Awareness of the importance of sport activities for the physical and emotional well-being of people with haemophilia has increased, and several sports projects are ongoing. In the German HEP, adult patients with haemophilia attend a sports training camp three times a year for 2 years; their muscle strength and proprioception are measured, their QoL is assessed (using the SF-36 and Haem-A-QoL) and subjective training effects are evaluated (using the HEP-Test). In the Emo-Sport Study, the prevalence of sport activities in Italian haemophilic children is being investigated. The attitudes of children and their parents towards sports are being evaluated, and QoL is being assessed (using the Haemo-QoL). In Israel, an interventional study is planned in which adolescent patients with severe haemophilia will undergo karate training. Patients will also undergo isokinetic and anaerobic testing, and their QoL will be assessed (using the Haemo-QoL). A study has been planned in Australia in which the effects of a regular exercise programme on aerobic fitness, strength and QoL of haemophilic children will be evaluated [59].

Because sports activities can improve not only physical well-being, but also the emotional and social well-being of a person, physical exercise and sport activities should be advocated in the global approach to haemophilia management. Decisions on the most suitable sport should be made in cooperation between the patient, his or her doctor, and the physiotherapist and should take into account the patient's physical status, interests and social needs.

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