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Professional functioning of young adults with congenital coagulation disorders in the Netherlands

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Introduction and Aim: Suboptimal health-related quality of life and lowered employment rates found in a previous study in young adults (YA) with congenital coagulation disorders (CCD) in the Netherlands underline the need for more insight into professional functioning of YA with CCD and into determinants of professional functioning.

Methods: Young adults (18-30 years) with CCD participated in a cross-sectional study. Professional functioning was assessed with the Work Productivity and Activity Impairment questionnaire (WPAI). Potential determinants were assessed with the Course of Life Questionnaire (CoLQ), Pediatric Quality of Life Inventory Young Adult version (PedsQL_YA), Illness Cognition Questionnaire (ICQ) and Haemophilia Activities List (HAL). Logistic regression analyses were performed in the complete sample of YA with CCD, and in YA men with haemophilia separately, to examine determinants of WPAI outcomes.

Results: Ninety-four YA (77 men; mean age 24.1 years, SD 3.5 and 17 women; mean age 24.5 years, SD 3.8) with CCD (74% haemophilia A/B) participated. 74.5% of YA were paid employed for on average 30 hours per week. Of these, more than a quarter reported work impairment. Older age and a non-severe type of haemophilia (in the sample of YA men with haemophilia) were associated with successful (paid) employment. No variables were associated with professional functioning (expressed as Presenteeism and Overall work impairment) in patients with CCD or haemophilia.

Conclusion: Three-quarters of YA with CCD were successful in finding paid employment. Though absenteeism was low, YA with paid employment needs attention as a considerable part experienced work impairment.

KEYWORDS

haemophilia, impairment, professional functioning, work, young adults

1 | INTRODUCTION

Haemophilia A and B and von Willebrand disease (VWD) are the most common congenital coagulation disorders (CCD) identified.[1] Congenital coagulation disorders are a result of a deficiency of the blood clotting system, in which bleeding is prolonged and excessive, and may occur spontaneous or posttraumatic.[2] Haemophilia is X-linked; women are carriers and men are affected. Other congenital coagulation disorders, such as VWD, have an autosomal inheritance pattern and affect both men and women. For women with coagulation disorders, heavy menstruation and postpartum bleeding are common.[3]

Since the '80s, the use of plasma concentrates of coagulation factors became common. This enabled to start prophylactic treatment in patients with severe haemophilia (<1% clotting factor VIII or IX present in the blood) in order to prevent bleeding and joint damage, ultimately allowing patients to maintain a near normal lifestyle with little risk for arthropathy.[4] Still, children and (young) adults growing up with coagulation disorders may experience impairments in daily life, such as restrictions in physical activity, travelling and school participation.[5-7] Concerns exist about the long-term impact of the disease and its treatment on adaptation and functioning in adult life. Most studies focus on physical complications (eg, prevention of joint damage and inhibitor development),[8,9] but important measures of successful treatment in childhood should also include the level of achieved independency, and professional and social participation in adulthood.[10-12] Finding an education or job that fits with some of the physical restrictions that YA with CCD encounter is important to strengthen work-related psychosocial skills.[13-15]

In two previous studies among the same patients as in the present study, we have found that, despite growing up in the current era and in a resource rich country with adequate treatment available, young adults (YA) with CCD in the Netherlands still experience obstacles and issues in daily life with regard to their treatment and condition.[5,6] For example, lower health-related quality of life (HRQOL), lower self-esteem and a lower rate of paid jobs were found in YA men with CCD compared to healthy peers,[6] which has also been found in the US.[16,17] Research conducted in 2001 in the Netherlands also demonstrated that adult men with severe haemophilia participated less in full-time work compared with the general population,[18] a finding which is common in chronically ill populations.[19,20] Unemployment among patients with CCD is an issue of concern from a societal perspective.

Impairment in professional functioning among people with chronic illness often seems to be related to pain, fatigue and physical limitations.[21] However, a considerable discrepancy between the level of functioning experienced by patients and the underlying pathology of the disease is often found. In patients with the same severity of disease, physical, mental and social problems experienced can vary from patient to patient and has therefore led to hypotheses about the contribution of psychosocial factors to health outcomes.[19,22] So far, literature on the role of psychosocial factors in relation to professional functioning in patients, and especially young adults, with CCD is scarce.[19,23] One study has shown that feelings about social stigma can hinder patients with CCD in finding employment.[14] Other literature has shown that many haemophilia patients select their job or training taking into account their limitations related to haemophilia (eg, jobs with not too much physical activity or without a risk of trauma) and that some patients believe that they have lost a job in the past because of their haemophilia.[24,25]

Suboptimal HRQOL and low employment rates found in a previous study[6] have led us to conceive the current study, in which we aim to gain more in-depth insight into professional functioning (paid employment, impairment in working) of YA with CCD and to assess whether sociodemographics, disease characteristics, functional disabilities, physical HRQOL and illness cognitions are associated with professional functioning.

2 | MATERIAL AND METHODS

2.1 | Participants

Young adult men and women aged 18-30 years with haemophilia A/B, VWD type 2 or 3 or other congenital coagulation disorders were eligible for participation if they were able to complete Dutch online questionnaires. Patients under treatment in one of the three participating haemophilia treatment centres (HTC's) in the Netherlands (Amsterdam UMC, Erasmus University Medical Center Rotterdam and Leiden University Medical Center) or YA who are a member of the Dutch Hemophilia Patient Society Young Adult committee (DHPSYA) were included (Table 1).

2.2 | Procedure

Invitational letters, including login codes for online questionnaires, were sent out between May and October 2015 to the eligible patients of the HTC's and to members of the DHPSYA after approval by the Medical Ethics Committees of the Amsterdam UMC, and Leiden University Medical Center. To recruit additional respondents, the DHPSYA posted an online call for participation on their website and Facebook page. Informed consent was obtained from all participants prior to completing questionnaires.

2.3 | Measures

2.3.1 | Sociodemographic and disease characteristics

Sociodemographic information of the participants was collected with questions from the Course of Life Questionnaire (CoLQ),[26] regarding age, gender, country of birth, education and marital status. Education was divided into three categories according to the classification of Statistics Netherlands (www.cbs.nl); low (primary education, lower vocational education, lower and middle general secondary education), middle (middle vocational education, higher secondary education, pre university education) and high (higher vocational education, university). Additionally, respondents were asked medical questions regarding the type of disease, severity

TABLE 1 Characteristics of young adults with coagulation disorders (CCD) by gender

	Men with CCD		Women with CCD	
	N	M (SD)	N	M (SD)
Sociodemographic characteristics				
Age	77	24.14 (3.53)	17	24.59 (3.79)
	N	%	N	%
Country of birth (Netherlands)	68	88.3	16	94.1
Education[a]				
High	19	24.7	7	41.2
Middle	43	55.8	6	35.3
Low	15	19.5	4	23.5
Marital status (married/living together)	21	27.3	6	35.3
CCD characteristics				
Type of coagulation disorder				
Haemophilia A	55	71.4	1	5.9
Haemophilia B	14	18.2	0	0.0
Von Willebrand type 2/3	5	7.8	10	58.8
Other congenital coagulation disorder	2	2.6	6	35.3
Type of treatment coagulation disorder				
Prophylaxis	37	48.1	1	5.9
On demand - in case of bleed	40	51.9	16	94.1
Severity of haemophilia				
Non-severe (>1%)	35	50.7	1	5.9
Severe (<1%)	34	49.3	0	0.0
	Median (range)	Median (range)		
Number of bleeds past 6 mo requiring treatment	1.00 (0-20)	0.00 (0-20)		
Functional disabilities (HAL scores)[b,c]	**N**	**M (SD)**		
Upper extremity activities	69	92.82 (18.56)		
Basic lower extremity activities	69	86.43 (22.87)		
Complex lower extremity activities	69	85.51 (21.86)		
Sum score	48	90.99 (20.75)		

M, mean; SD, standard deviation; HAL, Haemophilia Activities List, scoring 0-100 with higher scores representing better functioning.
[a]high = higher vocational education, university; middle = middle vocational education, higher secondary education, pre university education; low = primary education, lower vocational education, lower and middle general secondary education
[b]Only assessed in men with haemophilia.
[c]21 respondents had a missing value "not applicable" on one or more items.

(severe haemophilia: <1% clotting factor VIII or IX present in blood; non-severe haemophilia: 1%-40% clotting factor VIII or IX), treatment and number of bleeds that occurred over the past 6 months.

2.3.2 | Professional functioning: Work Productivity and Activity Impairment questionnaire (WPAI)

The Work Productivity and Activity Impairment (WPAI) questionnaire is a validated self-report questionnaire that assesses the impact of disease on work and other daily activities.[27] For the present study about professional functioning, the Dutch WPAI items about work were used: paid employment (yes/no), if yes: average working hours per week; number of missed hours at work the last 7 days due to the disease; degree of disease influence on work productivity the last 7 days (0-10, where 0 indicates no influence and 10 indicates that the disease completely prevented to do the job). Three outcomes are derived from the WPAI work-related items as follows: (a) Absenteeism (% worktime missed: missed hours at work in the last 7 days due to the disease divided by the average

TABLE 2 Professional functioning of young adults with coagulation disorders (CCD) by gender

	Men with CCD		Women with CCD	
	N	%	N	%
Paid employment (yes)	58	75.3	12	70.6
Absenteeism: >0% working hours missed due to CCD	3	5.2	0	0.0
Presenteeism: ≥10% impairment while working due to CCD	14	24.1	2	16.7
Overall work impairment: ≥10% due to CCD	14	24.1	2	16.7

working hours per week multiplied by 100); (b) Presenteeism [% impairment while working due to the disease, that is, the degree (0-10) the disease influenced work productivity divided by 10 and multiplied by 100]; (c) Overall work impairment, which is a combination of absenteeism and presenteeism: absenteeism + ([100-absenteeism] × presenteeism).

For all three outcomes, a higher percentage indicates more work impairment. For use in the present analyses, the three outcomes were dichotomized into whether or not (0% vs >0%) Absenteeism, Presenteeism or Overall work impairment was present. For Presenteeism and Overall work impairment, there were no scores between 0% and 10%, so for the concerning dichotomous variables, we used "<10% vs ≥10%".

2.3.3 | Haemophilia Activities List (HAL)

The Haemophilia Activities List (HAL) was used in YA men with haemophilia only (N = 69), to assess self-perceived limitations in activities (functional disabilities).[28] The HAL is a disease-specific questionnaire with 42 items in seven domains of disabilities, which are based on the International Classification of Functioning, Disability and Health (ICF)[29]: lying down/sitting/kneeling/standing (8 items), functions of the legs (9 items), functions of the arms (4 items), use of transportation (3 items), self-care (5 items), household tasks (6 items), leisure activities, and sports and other activities (7 items). Answers are given on a six-point Likert scale, ranging from "impossible" (1), "always problems with" (2) to "never problems with" (6). The items in the domains of transportation, household tasks and leisure activities have the additional answer option "not applicable". Consequently, these scores were recorded as missing value. Three component scores "upper extremity activities" (range 9-54), "basic lower extremity activities" (range 6-36) and "complex lower extremity activities" (range 9-54) are calculated, as well as a sum score of all items (range 42-252). Answers are rescaled and normalized to a 0-100 scale, with higher scores representing better functional status. The validity and reliability of the HAL are good in adult patients with haemophilia.[28,30] Cronbach's alpha in the present study ranged from 0.93 (function of arms) to 0.98 (self-care) for the domain scores and from 0.95 (complex lower extremity activities) to 0.99 (sum score) for the component scores.

2.3.4 | Pediatric Quality of Life Inventory (PedsQL 4.0) generic core scales young adult version

The physical health scale (8 items) of the Dutch version of the Pediatric Quality of Life Inventory generic core scales young adult version (PedsQL_YA; 18-30 years) was used.[31,32] The PedsQL_YA is a generic HRQOL instrument and consists of 23 items in total. Each item states a problem, for example, "it is hard for me to run". On a 5-point Likert scale, ranging from "never" (0) to "almost always" (4), the patient indicates whether he/she had problems with that item. Each answer is reversed scored and rescaled to a 0-100 scale so that higher scores indicate better reported physical functioning. The validity and reliability of the PedsQL_YA scales are good.[32] Cronbach's alpha of the physical health scale in our sample was 0.86.

2.3.5 | Illness Cognitions Questionnaire (ICQ)

To assess illness cognitions, the Dutch Illness Cognition Questionnaire (ICQ) was used. The ICQ is developed for adults which measures illness beliefs across various chronic conditions.[33] It contains 18 items in three scales, related to cognitive ways patients ascribe meaning to chronic illness: helplessness (6 items), acceptance (6 items) and perceived disease benefits (6 items). Answers are given on a four-point Likert scale ranging from "not at all" (1) to "completely" (4). Scale scores are calculated by summing up the item scores. Higher scores indicate stronger presence of the illness cognition. The validity and reliability of the ICQ scales are good .[33,34] Cronbach's alpha in the present study ranged from 0.79 (acceptation) to 0.86 (helplessness and disease benefits).

2.4 | Data analysis

Descriptive analyses were performed to characterize the YA with CCD and to gain insight in their professional functioning.

Next, logistic regression analyses were performed to identify potential determinants of professional functioning (WPAI outcomes: paid employment status, Presenteeism and Overall work impairment): sociodemographics and disease characteristics (see Table 1), functional disabilities, physical HRQOL and illness cognitions. Due to low variance, Absenteeism (see Table 2) could not be used as an outcome in the logistic regression analyses. Logistic regression analyses were performed for all CCD and separately for men with haemophilia.

Because of the large number of variables in relation to the sample size, preselection was necessary. Sociodemographic (country of birth, education and marital status) and disease characteristics (type of coagulation disorder, type of treatment, severity of haemophilia and number of bleeds), physical HRQOL (PedsQL_YA), illness cognitions (helplessness, acceptance, disease benefits; ICQ) and, in haemophilia only, functional disabilities (HAL component scores: upper extremity activities, basic lower extremity activities, complex lower extremities and sum score) were included in the logistic regression

TABLE 3 Logistic regression model of work impairment (WPAI outcomes) for young adults with CCD (N = 70)

	Impairment while working due to CCD ≥10%		Overall work impairment due to CCD ≥10%	
	Odds Ratio (CI 95%)	*P*-value	Odds Ratio (CI 95%)	*P*-value
Bleeds (≥1 past 6 mo)	1.76 (0.34-9.01)	0.50	1.76 (0.34-9.01)	0.50
Physical HRQOL	0.94 (0.89-1.01)	0.07	0.94 (0.89-1.01)	0.07
Helplessness	1.37 (0.92-2.03)	0.12	1.37 (0.92-2.03)	0.12
Acceptation	0.91 (0.68-1.22)	0.54	0.91 (0.68-1.22)	0.54

HRQOL, health-related quality of life, as measured with PedsQL_YA.

models if significantly ($P < 0.05$) associated with the WPAI outcomes in univariate analyses (according to Pearson correlation or χ^2 tests). All logistic regression analyses were controlled for age. Gender was not included in the models because it was strongly associated with characteristics of the disease. The results of the logistic regression analyses are expressed as Odds Ratios (OR) with 95% confidence intervals (CI).

3 | RESULTS

3.1 | Characteristics of YA with CCD

In total, 94 YA completed the online questionnaires (52% overall response rate): 34 YA (46% response rate) from the Amsterdam UMC, 37 YA (46% response rate) from the Erasmus University Medical Center Rotterdam, 10 YA (33% response rate) from the Leiden University Medical Center and 10 YA from the DHPSYA (83% response rate). An online call yielded another 3 YA. The mean age of the 77 men (81.9%) was 24.1 years (SD 3.5) and of the 17 women (18.1%) 24.6 years (SD 3.8). Table 1 presents the sociodemographic and disease characteristics of the study population and functional disabilities (HAL) of the subgroup of YA men with haemophilia.

3.2 | Professional functioning

Table 2 presents professional functioning according to the WPAI outcomes per gender. Out of 94 participating YA, 70 (74.5%) YA were paid employed. These 70 YA worked on average 29.8 (SD 14.0) hours per week. In the age group 18-25 years, 64% of YA were paid employed, while in the age group 26-30 years, 92% were paid employed. Absenteeism of 3%-25% of their average working hours was present in 4.3% (N = 3) of the YA with paid employment. Of the 70 YA with paid employment, 22.9% (N = 16) were ≥10% impaired while working due to CCD (Presenteeism), and 22.9% (N = 16) had ≥10% Overall work impairment due to CCD.

3.3 | Determinants of professional functioning

3.3.1 | Total cohort of YA with CCD

Older YA with CCD were more likely to have paid employment (OR 1.30 95% CI 1.1-1.5; data not shown). The results of the other logistic regression analyses are shown in Table 3. Presenteeism and Overall impairment due to CCD while working was not associated with sociodemographic and disease characteristics, physical HRQOL and illness cognitions.

3.3.2 | YA men with haemophilia

In the group of YA men with haemophilia, older patients (OR 1.21 95% CI 1.1-1.4) and patients with non-severe haemophilia (OR 7.82 95% CI 2.8-21.8; data not shown) were more likely to have paid employment. The results of the other logistic regression analyses are shown in Table 4. None of the variables included in the logistic regression models were significantly associated with Presenteeism or Overall impairment due to CCD while working were not associated with sociodemographic and disease characteristics, functional disabilities, physical HRQOL and illness cognitions.

4 | DISCUSSION AND CONCLUSION

This study examined professional functioning in young adult (YA) patients with congenital coagulation disorders (CCD). The results showed that three-quarters of YA with CCD were paid employed for on average 30 hours per week, which does not seem bad taking into account that the YA who were student cannot be expected to work full time (36-40 hours per week). Non-severe type of haemophilia was associated with a greater likelihood of being paid employed and the older the YA the more likely that the YA was paid employed. In the age group 26-30 years, when most YA have completed their education, paid employment was 92%. Of those with a paid job, one-quarter reported work impairment of ≥10%. Among those with a paid job, no sociodemographic, disease or other variables (functional disabilities, physical HRQOL and illness cognitions) were found to be associated with work impairment.

Compared to adults with other chronic illnesses, YA with CCD seemed to have adjusted well to a job once they have found one. In our sample, rates of missed working time and rates of work impairment were low in comparison to WPAI outcomes in adults with other chronic illnesses.[35,36] However, this comparison is hampered by the fact that our sample consisted of YA who have grown up with a congenital disease while the other studies concerned patients who became ill in adulthood. As a result, the latter patients may not have been able to take their current illness into account when choosing their career. Another explanation for the favourable outcomes could be that YA with worse employment outcomes did not participate in our study. Nevertheless, the low absenteeism we found in the YA with CCD indicates that patients with CCD in the Netherlands have

TABLE 4 Logistic regression model of work impairment (WPAI outcomes) in young adult men with haemophilia (N = 52)

	Impairment while working due to haemophilia ≥10%		Overall work impairment due to haemophilia ≥10%	
	Odds Ratio (CI 95%)	*P*-value	Odds Ratio (CI 95%)	*P*-value
Physical HRQOL	0.94 (0.86-1.02)	0.15	0.94 (0.86-1.02)	0.15
Helplessness	1.07 (0.60-1.90)	0.82	1.07 (0.60-1.90)	0.82
Acceptation	0.71 (0.48-1.06)	0.10	0.71 (0.48-1.06)	0.10
Disease benefits	1.28 (0.95-1.72)	0.11	1.28 (0.95-1.72)	0.11
Complex lower extremity activities[a]	0.99 (0.93-1.07)	0.85	0.99 (0.93-1.07)	0.85

HRQOL, health-related quality of life, as measured with PedsQL_YA.
[a]HAL basic lower extremity activities and HAL sum score were excluded from the model due to high multicollinearity with HAL complex lower extremity activities ($r > 0.80$).
Note: Number of bleeds was excluded from the model due to too high predictability of the outcomes.

adjusted well to their chronic disease status and are able to find a job that fits with their impairments. This could be due to the fact that half of the sample consisted of YA with non-severe CCD and that the patients with severe CCD used prophylaxis, which is common in the Netherlands. Prophylaxis limits joint damage which in turn could help prevent work disabilities. The fact that older patients did not score worse on the Haemophilia Activity List than younger patients (data not shown) is in line with this explanation because without prophylaxis, older YA with CCD would be more likely to suffer from joint damage than younger YA with CCD. That the patients are doing well, is probably also a spinoff of the enormous improvement in the care for haemophilia patients in the Netherlands in the last two decades. Treatment and care are concentrated in a small number of Haemophilia treatment centres, where multidisciplinary teams including social workers and/or paediatric psychologists are aware of psychosocial problems.

However, the percentage of YA with CCD that reported work impairment due to their disease (22.3% and 32.9%) is not negligible. In daily practice, it is important to monitor whether the requirements of the job match the possibilities of the YA with CCD not only when YA are in search of a job but also once they have found a job. It is recommended to screen whether YA need additional support in finding or doing their job by asking them to complete Patient-Reported Outcomes Measures (PROMs) before an outpatient visit, for example by using the KLIK portal.[37] Healthcare providers could refer the YA's to social workers or a job agency specialized in helping adolescents and young adults with chronic illness. Such an agency is "Emma at Work", a job mediation agency for adolescents with a somatic disease.[38] Founded by the Emma Children's Hospital Amsterdam UMC, it is now also located in other hospitals in the Netherlands and is open for all young people with a chronic condition. Moreover, it is important that employers are encouraged to adjust the workload and tasks to fit the abilities of patients, especially for those on prophylactic treatment.[5,16,17]

Previous studies among patients with haemophilia showed that physical limitations and pain did interfere with daily activity and work[39,40] and that professional functioning was associated with pain, fatigue and other physical limitations,[16,17,23] findings which we could not confirm in our study. Unfortunately, we did not assess pain and fatigue in our study, which would be interesting to include in the future, since it is known that pain is also negatively associated with physical and mental HRQOL.[39] Apart from age and severity of the CCD, we found no associations between professional functioning and sociodemographics (eg, educational level and marital status), disease characteristics (eg, number of bleeds and type of treatment), functional disabilities, physical HRQOL and illness cognitions. We do not have information on the type of work that YA perform, which would have been interesting to relate to the degree of work impairment experienced.

This study has a few limitations to take into account. First, though the response rates of the patients from the HTC's and the DHPSYA were satisfactory, we do not know whether the results are representative for YA with coagulation disorders in the Netherlands since sociodemographic and medical information of many non-respondents was unknown. Second, we were not able to compare the WPAI outcome "paid employment" to a healthy norm group because this information was not available. However, from a previous study among the same patients as in the present study,[6] it has already been known that the rate of paid employment was significantly lower in men with CCD than in male peers from the general population. Third, although the sample size was reasonably large, the power to detect determinants of the professional outcomes was still limited.

In conclusion, three-quarters of YA with CCD in the Netherlands were successful in finding paid employment. Though absenteeism was low among the YA with paid employment, employed YA with CCD need attention as more than one quarter of them experienced impairment in professional functioning.

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AUTHOR CONTRIBUTIONS

PL carried out the literature study, organized data collection, led data analysis and drafted the manuscript. LH supervised data collection and data analysis, and critically revised the manuscript. HMS supervised data collection and data analysis, and critically revised the manuscript. MP conceived the study, supervised data collection and critically revised the manuscript. MG conceived the study, supervised data collection and critically revised the manuscript for intellectual content. MC, MK and JE critically revised the manuscript for intellectual content. All authors read and approved the final manuscript.

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REFERENCES

1. Barlow JH, Stapley J, Ellard DR. Living with haemophilia and von Willebrand's: a descriptive qualitative study. *Patient Educ Couns*. 2007;68(3):235-242.
2. Mannucci PM, Tuddenham E. The hemophilias: from royal genes to gene therapy. *N Engl J Med*. 2001;344(23):1773-1779.
3. Khair K, Holland M, Pollard D. The experience of girls and young women with inherited bleeding disorders. *Haemophilia*. 2013;19(5):e276-281.
4. Bauer KA. Current challenges in the management of hemophilia. *Am J Manag Care*. 2015;21:S112-S122 111p.
5. Limperg P, Peters M, Gibbons E, et al. Themes in daily life of adolescents and young adults with congenital bleeding disorders: a qualitative study. *Haemophilia*. 2016;22(4):e330-e333.
6. Limperg PF, Haverman L, Maurice-Stam H, et al. Health-related quality of life, developmental milestones, and self-esteem in young adults with bleeding disorders. *Qual Life Res*. 2017;27(1):12.
7. Soucie JM, Grosse SD, Siddiqi A, et al. The effects of joint disease, inhibitors and other complications on health-related quality of life among males with severe haemophilia A in the United States. *Haemophilia*. 2017;23(4):e287-e293.
8. Franchini M, Mannucci PM. Inhibitors of propagation of coagulation (factors VIII, IX and XI): a review of current therapeutic practice. *Br J Clin Pharmacol*. 2011;72(4):553-562.
9. Manco-Johnson MJ, Abshire TC, Shapiro AD, et al. Prophylaxis versus episodic treatment to prevent joint disease in boys with severe hemophilia. *N Engl J Med*. 2007;357(6):535-544.
10. Tjaden LA, Maurice-Stam H, Grootenhuis MA, Jager KJ, Groothoff JW. Impact of renal replacement therapy in childhood on long-term socioprofessional outcomes: a 30-year follow-up study. *J Pediatr*. 2016;171:189-195.
11. Schrijvers L, Beijlevelt-Van der Zande M, Peters M, et al. Achieving self-management of prophylactic treatment in adolescents: the case of haemophilia. *Patient Educ Couns*. 2016;99(7):1179-1183.
12. Sattoe JN, Hilberink SR, van Staa A, Bal R. Lagging behind or not? Four distinctive social participation patterns among young adults with chronic conditions. *J Adolesc Health*. 2014;54(4):397-403.
13. Young G. Transitioning issues in adolescent to young adult hemophilia patients with inhibitors: an approach for a growing population. *Blood Coagul Fibrinolysis*. 2010;21:848-857.
14. Cassis F. Psychosocial care for people with hemophilia. *Treatment Hemophilia*. 2007;44:1-12.
15. Cassis F, Querol F, Iorio A, Forsyth A. Psychosocial aspects of haemophilia: a systematic review of methodologies and findings. *Haemophilia*. 2012;18:101-114.
16. Witkop M, Guelcher C, Forsyth A, et al. Treatment outcomes, quality of life, and impact of hemophilia on young adults (aged 18–30 years) with hemophilia. *Am J Hematol*. 2015;90(suppl 2):S3-10.
17. Curtis R, Baker J, Riske B, et al. Young adults with hemophilia in the U.S.: demographics, comorbidities, and health status. *Am J Hematol*. 2015;90(suppl 2):S11-16.
18. Plug I, Peters M, Mauser-Bunschoten EP, et al. Social participation of patients with hemophilia in the Netherlands. *Blood*. 2008;111(4):1811-1815.
19. Boot C, Heijmans M, van der Gulden J, Rijken M. The role of illness perceptions in labor participation of the chronically ill. *Int Arch Occup Environ Health*. 2008;82(1):13-20.
20. Hoving JL, van der Meer M, Volkova AY, Frings-Dresen M. Illness perceptions and work participation: a systematic review. *Int ArchOccup Environ Health*. 2010;83(6):595-605.
21. Baanders AN, Rijken PM, Peters L. Labour participation of the chronically ill. A profile sketch. *Eur J Public Health*. 2002;12(2):124-130.
22. Engel GL. The need for a new medical model: a challenge for biomedicine. *Science*. 1977;196(4286):129-136.
23. Chen SL. Economic costs of hemophilia and the impact of prophylactic treatment on patient management. *Am J Manag Care*. 2016;22(5 suppl):s126-133.
24. Forsyth AL, Gregory M, Nugent D, et al. Haemophilia Experiences, Results and Opportunities (HERO) Study: survey methodology and population demographics. *Haemophilia*. 2014;20(1):44-51.
25. Talaulikar D, Shadbolt B, McDonald A, Pidcock M. Health-related quality of life in chronic coagulation disorders. *Haemophilia*. 2006;12(6):633-642.
26. Grootenhuis MA, Stam H, Destree-Vonk A, Heymans HS, Last BF. Levensloop vragenlijst voor jong-volwassenen. [Course of life questionnaire for young adults]. *Gedrag en Gezondheid*. 2003;31(1):336-350.
27. Reilly MC, Zbrozek AS, Dukes EM. The validity and reproducibility of a work productivity and activity impairment instrument. *Pharmacoeconomics*. 1993;4(5):353-365.
28. van Genderen FR, van Meeteren NL, van der Bom JG, et al. Functional consequences of haemophilia in adults: the development of the Haemophilia Activities List. *Haemophilia*. 2004;10(5):565-571.
29. *ICF: International Classification of Functioning, Disability and Health*. Geneva: World Health Organization; 2001.
30. van Genderen FR, Westers P, Heijnen L, et al. Measuring patients' perceptions on their functional abilities: validation of the Haemophilia Activities List. *Haemophilia*. 2006;12(1):36-46.
31. Varni JW, Limbers CA. The PedsQL 4.0 Generic Core Scales Young Adult Version: feasibility, reliability and validity in a university student population. *J Health Psychol*. 2009;14(4):611-622.
32. Limperg PF, Haverman L, van Oers HA, Van Rossum MA, Maurice-Stam H, Grootenhuis MA. Health related quality of life in Dutch young adults: psychometric properties of the PedsQL generic core scales young adult version. *Health Qual Life Outcomes*. 2014;12(1):9.
33. Evers AW, Kraaimaat FW, van Lankveld W, Jongen PJ, Jacobs JW, Bijlsma JW. Beyond unfavorable thinking: the illness cognition questionnaire for chronic diseases. *J Consult Clin Psychol*. 2001;69(6):1026-1036.

34. Lauwerier E, Crombez G, Van Damme S, Goubert L, Vogelaers D, Evers AW. The construct validity of the illness cognition questionnaire: the robustness of the three-factor structure across patients with chronic pain and chronic fatigue. *Int J Behav Med*. 2010;17(2):90-96.
35. de Hooge M, Ramonda R, Lorenzin M, et al. Work productivity is associated with disease activity and functional ability in Italian patients with early axial spondyloarthritis: an observational study from the SPACE cohort. *Arthritis Res Ther*. 2016;18(1):265.
36. Maksymowych WP, Gooch KL, Wong RL, Kupper H, van der Heijde D. Impact of age, sex, physical function, health-related quality of life, and treatment with adalimumab on work status and work productivity of patients with ankylosing spondylitis. *J Rheumatol*. 2010;37(2):385-392.
37. Haverman L, van Rossum MA, van Veenendaal M, et al. Effectiveness of a web-based application to monitor health-related quality of life. *Pediatrics*. 2013;131(2):e533-e543.
38. Brouwer P, Giesen FB. *De meerwaarde van Emma at work [The added value of Emma at Work]*. Hoofddorp, the Netherlands: TNO Kwaliteit van Leven; 2009.
39. McLaughlin JM, Munn JE, Anderson TL, Lambing A, Tortella B, Witkop ML. Predictors of quality of life among adolescents and young adults with a bleeding disorder. *Health Qual Life Outcomes*. 2017;15(1):9.
40. Witkop ML, McLaughlin JM, Anderson TL, Munn JE, Lambing A, Tortella B. Predictors of non-adherence to prescribed prophylactic clotting-factor treatment regimens among adolescent and young adults with a bleeding disorder. *Haemophilia*. 2016;22(4):e245-e250.

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