Assessment of self-/parent-reported quality of life in Japanese children with hemophilia using the Japanese version of KIDSCREEN-52

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Abstract

***Introduction*:** Assessing health-related quality of life (HRQOL) is critical for providing comprehensive clinical care to patients with hemophilia. HRQOL in individuals with similar cultural backgrounds should be compare using internationally standardized, generic questionnaires. ***Aim*:** To evaluate self-/parent-assessed HRQOL in Japanese children and adolescents with hemophilia A or B. ***Methods*:** Children and adolescents aged 8-18 years were enrolled. The hemophilia group comprised families with hemophilia and the control group comprised those without chronic illness. HRQOL was assessed using the self-/parent-reported questionnaire KIDSCREEN-52, Japanese version. The Oslo 3-Item Social Support Scale was investigated. ***Results*:** The questionnaire was completed by 36 families in the hemophilia group and 160 parents and children in the control group. Hemophilia children aged 8-12 years had lower scores for 'moods and emotions' than control children; the parents had lower scores in the hemophilia group than in the control group for 'moods and emotions', 'social support and peers', and 'school environment'. No significant differences in HRQOL were observed between both groups of adolescents aged 13-18 years or their parents. Neck-shoulder pain was associated with a low psychological state, including 'self-perception', but other joint pains did not affect the outcomes of the HRQOL indices. Social support weaknesses were associated with low physical and psychological states, whereas unexpected hospital visits identified low values for 'self-perception', 'autonomy', and 'school environment'. ***Conclusion*:** Proactive mental and clinical care in hemophilia families, especially with young children, will foster a better environment for patients and their parents and ease concerns about progress in hemophilia.

Keywords: hemophilia, health-related quality of life, self-assessment, children, survey

Introduction

Hemophilia A and B are hereditary, X-linked recessive bleeding disorders caused by deficiencies of coagulation factors VIII and IX, with a prevalence of 1 per 5,000 and 25,000 men, respectively [1,2]. Clinical hemorrhagic symptoms in the affected individuals are, for the most part, represented by spontaneous bleeding or hemorrhage resulting from traumatic or surgical procedures. Repeated bleeding episodes lead to long-term musculoskeletal complications, including synovitis, degenerative arthropathy, and articular deformities. The bleeding symptoms and the bleeding-nonrelated symptoms in these patients, together with the need for intensive hemostatic therapy, interfere with their lifestyles. To prevent the repetitive bleeding and resultant joint damage, regular prophylaxis treatment is widely performed [3,4]. In Japan, approximately 90% of patients with severe hemophilia receive prophylaxis treatment [5]. More recently, the introduction of extended half-life recombinant factor VIII or IX products provides reduced frequency of infusion [6]. Despite the development of hemostatic management, the real status of quality of life (QOL) for patients with hemophilia remains incompletely understood. The assessment with focus of annual bleeding rates alone in current clinical trials may be not enough to understand the patients' QOL. Therefore, health-related QOL (HRQOL) is believed to play an important part in comprehensive medical care. Several HRQOL protocols have been proposed for the assessment of hemophilia children using disease-specific questionnaires [7-14], but few studies have investigated self-reported assessment using an internationally standardized, generic questionnaire [8,10]. In this context, a modification of the widely accepted HRQOL questionnaire (KIDSCREEN-52) has been established and validated in Japanese children with hemophilia (J-KIDSCREEN-52), which is assessed by patients and their parents [15].

Previous studies have demonstrated that several factors influenced HRQOL in children and adolescents with hemophilia [16,17]. In particular, the development of an inhibitor alloantibody against factor VIII or IX and articular deformities markedly affected HRQOL. Unexpected bleeding, frequent hemarthroses, school absences, and unplanned hospital visits were also reported as significant risk factors for low HRQOL [18]. In circumstances in which hemophilia treatment now aims to improve the 'normal activity of life', comparing the age-matched hemophiliacs with the non-hemophiliacs is important. Furthermore, reliable indices of QOL could depend, however, on comparisons between hemophiliacs and non-hemophiliacs with

similar cultural backgrounds. The present study was therefore designed to survey self- and parent-reported HRQOL in Japanese children and adolescents with hemophilia, compared with those with no history of chronic illness, utilizing the J-KIDSCREEN-52 recommendations.

Materials and Methods

Participants - In total, 60 patients with hemophilia were recruited by individuals who had been previously diagnosed at one university hospital (Nara Medical University) and two general hospitals (Higashiosaka City Medical Centre and Hoshigaoka Medical Centre) in Japan. Children and adolescents aged 8-18 years with any type of hemophilia who could read and understand the questionnaire were enrolled in the study. The children were defined as those aged 8-12 years and adolescents as those aged 13-18 years. The control group consisted of 260 male patients from six outpatient clinics and one general hospital who were aged 8-18 years with no history of chronic illness such as severe asthma, atopic dermatitis, and epilepsy and with no condition of acute illness such as pharyngitis/laryngitis, pneumonia, and allergic rhinitis. The questionnaires were given to each family after ethical considerations and detailed explanation of the study objectives were made. The receipt of the completed questionnaire from the patients and their parents in both the hemophilia and control groups was accepted as consent for participation in the study. The study protocol was approved by the Ethical Committees of each of the participating institutions.

QOL survey - Self- and parent-reported HRQOL was assessed using the J-KIDSCREEN-52, which was originally described as an appropriate tool for measuring HRQOL in children and adolescents living with cancer, diabetes, and juvenile idiopathic arthritis [19]. The protocol defines ten dimensions of HRQOL: 'physical well-being' (physical and motor activity, cheerfully definition); 'psychological well-being' (satisfaction with life and positive thinking); 'moods and emotions' (sense of helplessness, loneliness, and depressive feelings); 'self-perception' (body and inner self-image and satisfaction with appearance); 'autonomy' (freedom of choice and self-sufficiency); 'parent relations and home life' (relationship with parents, feeling of love by parents, and atmosphere at home); 'financial resources' (having enough money for social activities); 'social support and peers' (relationship with peers and quality of communication with friends); 'school environment' (satisfaction with school life, teachers, and academic results); and 'social acceptance' (bullying, feelings of rejection by peers,

and anxiety among one's social group).

Behavioural patterns, including feeling or intensity of an attitude, were assessed using a 5-point Likert response scale. The scores were coded from 1 to 5, negatively formulated items were recoded, and item scores for each respective dimension were added. Higher scores indicated better HRQOL in each dimension. A specific example of the question was: 'Have you felt fit and well?' and the answer was: 'Not at all, slightly, moderately, very, extremely'. This question assessed the intensity of attitudes towards certain questions. Another example of the question was: 'Have you been happy with way you are?' and the answer was: 'Never, seldom, sometimes, often, always'. This assesses the frequency of certain behaviours and feelings towards certain questions [22]. Within the standardization subjective, the mean *t*-score for each dimension was 50 points with 10 points of standard deviation [20-23]. The reliability and the validity of the J-KIDSCREEN-52 have been confirmed using children/adolescent and parent/proxy questionnaires [15].

Other variables - The parents were requested to confirm the presence or the absence of chronic health conditions within their families, the number of family members, and parental marital status. The Oslo 3-Item Social Support (OSS-3) scale [24] was adopted to assess the levels of social support. The original OSS-3 questionnaire was translated and arranged according to the Japanese life style by the current authors. The scale identifies: (i) how many people are available to provide a sense of security and support to the child (1 item) and (ii) the level of emotional and instrumental support provided by those people (2 items). The total score was calculated by the sum of these three indices. The scores ranged from 3 to 14 points, with the higher scores indicating the presence of stronger social support.

Clinical data - Clinical data were extracted from the patients' medical records. These included previous medical history, family history, age at onset of the disorder, type and severity of hemophilia, type of home treatment, frequency of injections, presence of target joints, status of inhibitor development, and surgical history. The profiles of each participant are presented in Table 1.

Statistical analysis - The mean values and the differences between the hemophilia group and the control group were compared using descriptive statistics and the chi-squared test, respectively. The Student's *t*-test was used to compare the mean HRQOL scores in both groups, and the Mann-Whitney test was utilized to compare the median HRQOL scores in the hemophilia group. A two-tailed $p< 0.05$ was considered to be statistically significant. All of the statistical analyses were performed using SPSS version 24 (IBM SPSS Statistics, Chicago, IL, USA).

Results

Profiles of participants in the hemophilia group and the control group - The completed questionnaires were returned by 160 of 260 potential male children and adolescents in the control group (response rate, 61.5%). Thirty-six of 60 potential subjects in the hemophilia group returned the completed questionnaires (response rate, 60.0%), indicating similar response rates in both groups. However, there were no significant differences in the average QOL scores in these healthy subjects at a similar age to those recorded in the J-KIDSCREEN-52 literature [15], and the application of selection bias was considered to be unnecessary (data not shown). No significant differences in age distribution, extent of social support, and rate of response or family structure were apparent between the patient and the control groups, indicating a similar cultural background within these families (Table 1A).

Comparison of the J-KIDSCREEN-52 scores between the hemophilia and the control groups - A significant difference was observed between children (8-12 years of age) in the hemophilia group compared with those in the control group in one dimension of the self-assessed HRQOL. In this analysis, the hemophilia group showed a significantly lower score for 'moods and emotions' than the control group (mean, 48.2 and 52.4, respectively; $p = 0.023$) (Figure 1A). In contrast, no significant differences in any of the self-assessed HRQOL dimensions were observed between these groups in the adolescent category (13-18 years of ages) (Figure 2A). Alternatively, analyses of parent-assessed HRQOL demonstrated significant differences between patients and controls in several indices. In particular, in the lower age category (8-12 years of age), the hemophilia parents' group demonstrated significantly lower scores than the parents in the control group in 'moods and emotions' (43.4 and 49.6, respectively; $p = 0.042$), 'social support and peers' (47.6 and 54.0; $p = 0.041$), 'self-perception' (45.1 and 50.3; $p = 0.026$), and 'school environment' (49.0 and 54.6; $p = 0.048$) (Figure 1B). However, no significant differences in any of these dimensions were noted in the records of parents of

adolescents (Figure 2B).

Influence of specific features on J-KIDSCREEN-52 dimensions in the hemophilia group - The clinical information on children and adolescents in the hemophilia group is summarized in Table 1B. Based on this information, further detailed analyses were performed on three features (social support weakness, target joint, and unexpected hospital visits) that have been previously reported to be associated with a low QOL [16-18]. Comparisons of the median scores of patients' HRQOL related to J-KIDSCREEN-52 dimensions are summarized in Table 2. A weakness in social support was associated with a low physical state and a low psychological state and appeared to negatively influence family relationships and school environment. The identification of a target joint resulted in a low physical state and a low psychological state. This clinical complication also appeared to affect the responses of social support and peers. In addition, unexpected hospital visits resulted in low QOL parameters for self-perception, autonomy, family relations, and school environment. The treatment period of regular prophylaxis between patients with target joint(s) and those without target joint(s) was also compared, but no significant difference in both groups was observed (data not shown), may supporting the reconsideration of prophylaxis regimen for the patients with target joints, because it is known that patients on long-term prophylaxis have fewer or no target joints.

The relationship between body pain [17] and QOL was also examined. Neck and shoulder pain appeared to have a significant negative influence on the patient's psychological state, self-perception, and school environment. However, other episodic joint pain, including elbows, hands, knees, and ankles seemed to have little effect on these determinants of QOL (data not shown).

Discussion

To the best of our knowledge, this report is the first to compare self-/parent-assessed HRQOL in children with hemophilia and those without hemophilia, among individuals from a similar cultural background in the same country. An internationally standardized, generic questionnaire was utilized, which demonstrated significant differences between the hemophilia group and the control group in self-reported QOL and parent-assessed QOL.

Self-assessment in the lower-aged group of patients with hemophilia (8-12 years) identified some concerns about their QOL, especially related to mood and emotions. These anxieties appeared to have eased in the higher-aged group, suggesting a progressive understanding of the disorder. The parents' assessment of the younger children reflected more negative images of emotions, social support and friends, and school environment, but again these apprehensions were not reported by the parents of adolescents. Similar conclusions have been reported in other studies [25]. In recent years, advances in hemophilia treatment, especially prophylaxis treatment and development of products, have resulted in a markedly improved QOL to near-normal levels of QOL for the majority of children and adolescents [26,27], and in the present survey, the self- and parent-assessment HRQOL scores in children aged 13-18 years receiving prophylaxis were not significantly different from those of the control group, strongly confirming the effectiveness of prophylactic therapy.

In our patients with hemophilia, the presence of target joints led to a low HRQOL related to both psychological and physical states. This finding was consistent with previous reports on negative factors affecting the social life of children [16-18]. Therefore, it seemed possible that a greater awareness of target joints during routine comprehensive hemophilia care could improve QOL in these individuals. Moreover, the development of a FVIII inhibitor, one of the current unmet needs of hemophilia care, appears to be a key event affecting QOL. The number of patients with inhibitors in the current study was small (n = 3) and comparisons with non-inhibitor patients were not totally reliable. Nevertheless, the presence of an inhibitor tended to reduce HRQOL (presence vs absence of inhibitor; social support and peers, 13.6 vs 48.4; $p < 0.05$), suggesting that modern protocols intended to prevent antibody development or treat inhibitor patients could lead to improvements in QOL [28]. In this context, an activated factor VIII (VIIIa)-mimicking bispecific antibody (emicizumab) recognizing factor IXa and factor X is now available [29] and can markedly reduce bleeding events in patients with hemophilia A with or without inhibitors [30-33]. It seems likely that a wider use of emicizumab could improve HRQOL, particularly in inhibitor-positive patients.

Furthermore, our data demonstrated that unplanned hospital visits interfered with school events and tended to limit meetings with friends. These social contacts provide the opportunity to enhance HRQOL [34]. Neck and shoulder pain appeared to be a particular indicator of a low

psychological state, although it remained unclear if the symptoms were due to hemorrhage. In contrast, bleeding into the knee, ankle, and elbow joints, which is known to be a major feature in severe hemophilia, was not significantly associated with any HRQOL dimension, including physical and psychological states (data not shown). The findings were consistent with the hypothesis that unexplained pain accentuates the anxiety about bleeding in children and adolescents with hemophilia [35].

Few studies have compared HRQOL in children with and without hemophilia. Earlier research has indicated that adults with hemophilia, aged 16-70 years, had a significantly lower QOL (than the normal control group) with respect to physical, but not psychological, aspects [36]. However, our investigation focused on children and adolescents only up to the age of 18 years, and it seemed likely that the improvements that were evident in the current survey reflected, at least in part, the quality of medication now available for patients from an early age. Nevertheless, our findings demonstrated that parents of hemophilia children had potentially serious fears related to the school environment of their children, especially those aged 8-12 years. It is also conceivable that adolescents with hemophilia may have a closer relationship with their parents because of the need for long-term commitments as compared with the control group. These results suggested that families would benefit from a careful explanation of the clinical consequences of hemophilia at the time of diagnosis and at important milestones in their lives, for example, kindergarten, primary, junior high, and high school. In addition, continuing education and updated information on hemophilia, including ongoing treatment plans and future prospects, could prevent parents from developing negative attitudes towards this inherited hemorrhagic disorder [37]. Therefore, the evidence indicates that professional support for patients and their parents could be vital for sustaining social growth in children and adolescents with hemophilia [38].

Strengths and Limitations

This report describes the first comparison of self-assessed QOL in a control group and a hemophilia group in Japan using a standardized generic questionnaire. The hemophilia group of subjects was enrolled from local hospitals and a major hemophilia treatment centre, and closely represented generic Japanese patients with hemophilia. In addition, the study surveyed the QOL of children assessed by their parents and utilized OSS-3 to examine social support weaknesses.

However, there are several limitations to this study. First, there is a likelihood of selection bias because the collection rates were not high (approximately 60%) for the control group and hemophilia group. In the control group, it may be expected that patients with more complaints were inclined to participate in the present study. Therefore, the difference of QOL in both groups may be underestimated. Second, 36 patients with hemophilia were recruited, and the sample size of each age group was limited. Therefore, the statistical distributions were restricted to a univariate analysis. A further study enrolling a larger number of subjects appears to be warranted to strengthen our present data. In addition, there was one patient classed as mild hemophilia, and the findings were therefore focused on the severe/moderate phenotype. Discrepancies in HRQOL scores based on J-KIDSCREEN-52 may be recorded between severe/moderate patients treated with prophylaxis and the mild type of patients treated on demand. A similar survey targeting patients with mild hemophilia is warranted. Nevertheless, the present study provided an analysable sample size of children with hemophilia living in the community and clearly demonstrated the impact of several related factors on self- and parent-assessed HRQOL.

Conclusion

Self-assessment by children demonstrated potential anxieties in the group of patients with hemophilia aged 8-12 years. Similarly, the parents of these younger individuals with hemophilia reported psychological concerns, particularly related to the school environment of their children. The results suggested that children and adolescents with hemophilia and their parents could benefit from psychosocial intervention to reduce the risk of a poor QOL, especially in the lower-aged group. The study also demonstrated that children with hemophilia, especially those with recurrent neck or shoulder pain identified as a target joint, developed a low HRQOL that could be improved with early intervention. The survey indicated that continuing education and professional support would foster a better environment for patients and their families to ease their concerns about progress in this difficult hereditary disorder. In addition, our results would contribute towards assessing the usefulness of novel therapy for hemophilia treatment in the future.

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Authorship

Contribution: Y.F. performed the experiments, designed the research, analyzed and interpreted the data, created the figures, and wrote the paper. K.N. performed the experiments, designed the research, interpreted the data, wrote the paper, edited the manuscript, and approved the final version of the manuscript for publication. K.Y. performed the experiments and designed the research. S.N., K.O., K.S., N.K., and M.S. designed and supervised the research. M.N. and S.K. managed the clinic.

Conflicts of Interest

K.O. and K.S. received research funding from YKK AP Inc., Ushio Inc., Tokyo Electric Power Company, EnviroLife Research Institute Co., Ltd., Sekisui Chemical Co., Ltd., LIXIL Corp., and Kyocera Corporation. The other authors declare that they have no potential conflicts of interest to disclose.

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Figure Legends

Figure 1. Comparison of the 10 dimension scores in the hemophilia and control groups aged 8-12 years (children) in the J-KIDSCREEN-52 analyses. (*Pane*l **A**) **Patient's side**. *Differences in the dimension of 'moods and emotions' were statistically significant. (*Panel* **B**) **Parent's side**. *Differences in the dimensions of 'moods and emotions', 'self-perception', 'social support and peers', and 'school environment' were statistically significant. The axis represents the calculated scores for each dimension obtained from the questionnaires. The mean and standard deviation values are shown. A two-tailed $p < 0.05$ was considered to be statistically significant.

Figure 2. Comparison of the 10 dimension scores in the hemophilia and control groups aged 13-18 years (adolescents) in the J-KIDSCREEN-52 analyses. (*Pane*l **A**) **Patient's side**. There were no significant differences. (*Panel* **B**) **Parent's side**. There were no significant differences. The axis represents the calculated scores for each dimension obtained from the questionnaires. The mean and standard deviation values are shown. A two-tailed $p < 0.05$ was considered to be statistically significant.

Table 1. Demographic characteristics of participants

(A) Control and hemophilia groups

		Control (n = 160)	Hemophilia (n = 36)
Age	Years	11.3 ± 2.7	12.7 ± 3.3
8-12 years	n (%)	87 (54.4)	17 (47.2)
13-18 years	n (%)	73 (45.6)	19 (52.8)
Social support - weakness †	n (%)	49 (30.6)	14 (38.9)

†Oslo 3-Item Social Support scale was used.

(B) Clinical data in hemophilia group

Clinical Data		N	(%)
Type	Hemophilia A	31	(86.1)
	Hemophilia B	5	(13.9)
Severity	Severe	28	(77.8)
	Moderate	7	(19.4)
	Mild	1	(2.8)
Sibling (hemophilia)	Present	10	(27.8)
	None	26	(72.2)
Inhibitor	Positive	3	(8.3)
	Negative	33	(91.7)
Injection frequency	3 times/week	25	(69.4)
	2 times/week	7	(19.4)
	1 time/week	2	(5.6)
Home treatment	Prophylaxis	34†	(94.4)
	On demand	2	(5.6)
Unexpected hospital visit	Present	9	(25.0)
	None	27	(75.0)
Target joint	Present	14	(38.9)
	None	22	(61.1)
Orthopedic surgery	Present	3	(8.3)
	None	33	(91.7)

†All of the patients showed almost good adherence from the patients' record.

Table 2. Comparison of median for the J-KIDSCREEN-52 scores by responders

J-KIDSCREEN-52 Dimensions	Social Support		*p*	Target Joint		*p*	Unexpected Hospital Visit		*p*	Neck and Shoulder Pain		*p*
	Weak (n=14)	Moderate and Strong (n=22)		Present (n=14)	None (n=22)		Present (n=9)	None (n=27)		Present (n=5)	None (n=30)	
Physical well-being	42.5	55.6	**0.012**	42.5	59.4	**0.002**	42.5	55.6	0.079	42.5	55.6	0.207
Psychological well-being	42.5	51.8	**0.013**	45.1	51.8	**0.006**	41.5	51.8	0.067	39.9	50.5	**0.025**
Moods and emotions	43.9	50.2	0.141	43.9	50.2	0.240	43.9	51.3	0.117	43.9	50.2	0.237
Self-perception	44.6	44.6	0.775	43.2	46.1	0.121	41.8	46.1	**0.025**	40.5	46.1	**0.013**
Autonomy	49.7	48.7	0.665	48.7	50.8	0.170	42.1	50.8	**0.008**	42.1	49.7	0.567
Parent relations and home life	44.6	58.5	**0.022**	45.7	56.6	0.062	45.7	56.6	**0.046**	42.6	54.7	0.099
Financial resources	41.9	41.9	0.240	41.9	44.3	0.665	39.7	46.6	0.180	32.5	44.3	0.054
Social support and peers	40.9	56.5	**0.004**	40.9	56.5	**0.006**	48.4	48.4	0.450	38.2	49.3	0.321
School environment	47.9	54.2	**0.013**	47.9	54.2	0.170	43.8	52.2	**0.047**	40.9	53.2	**0.001**
Social acceptance (bullying)	53.5	50.5	0.451	58.9	42.2	0.327	42.2	58.9	0.330	38.4	58.9	0.202

Each group was compared using Mann–Whitney U test ($p < 0.05$). All of the values show the median values, and the number in parenthesis shows the number of persons. Bold text indicates significant difference.

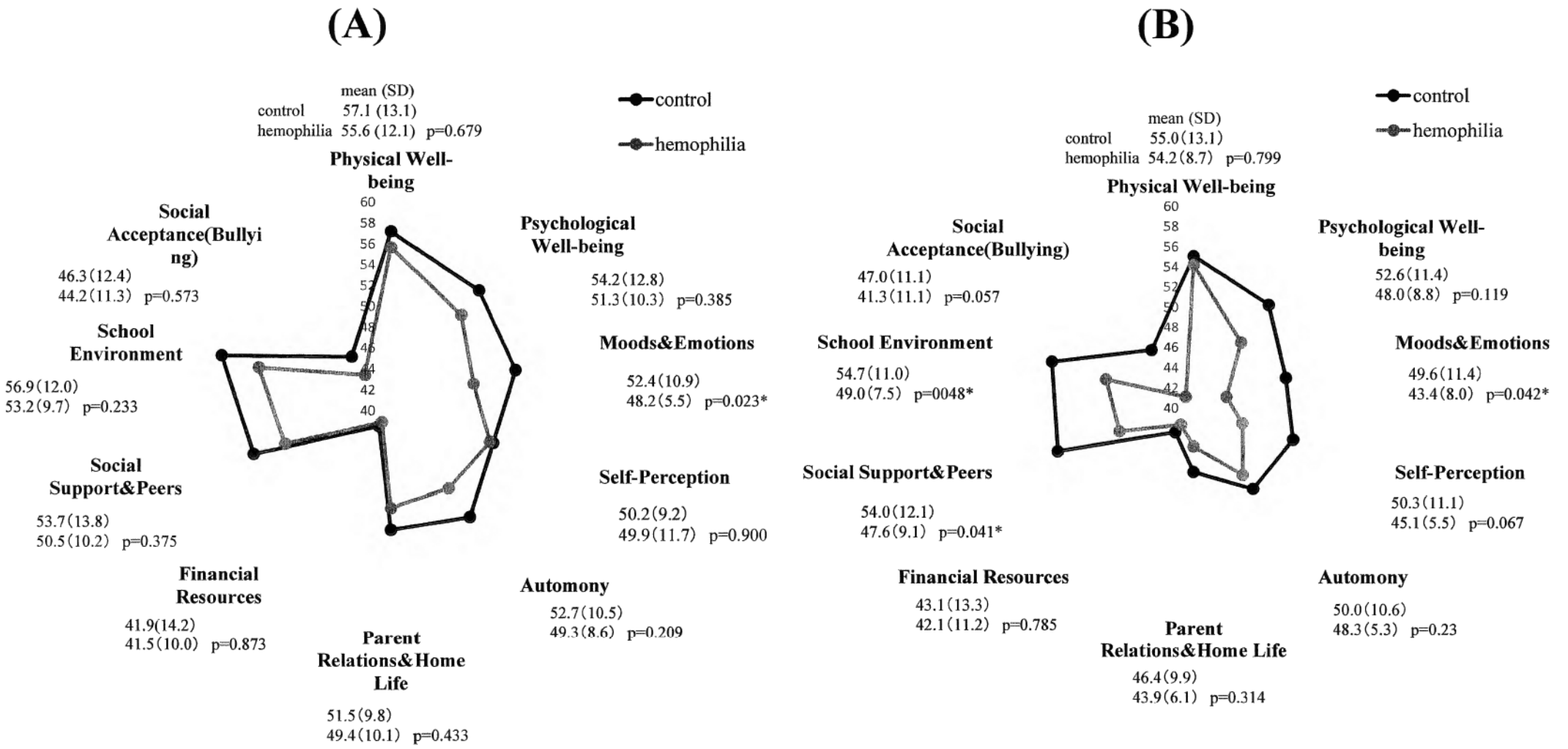
(A)
mean (SD)
control 57.1 (13.1)
hemophilia 55.6 (12.1) p=0.679
control
hemophilia
Physical Well-being
Psychological Well-being
54.2(12.8)
51.3(10.3) p=0.385
Moods&Emotions
52.4(10.9)
48.2(5.5) p=0.023*
Self-Perception
50.2(9.2)
49.9(11.7) p=0.900
Automony
52.7(10.5)
49.3(8.6) p=0.209
Parent Relations&Home Life
51.5(9.8)
49.4(10.1) p=0.433
Financial Resources
41.9(14.2)
41.5(10.0) p=0.873
Social Support&Peers
53.7(13.8)
50.5(10.2) p=0.375
School Environment
56.9(12.0)
53.2(9.7) p=0.233
Social Acceptance(Bullying)
46.3(12.4)
44.2(11.3) p=0.573
60
58
56
54
52
50
48
46
44
42
40
(B)
mean (SD)
control 55.0(13.1)
hemophilia 54.2(8.7) p=0.799
control
hemophilia
Physical Well-being
Psychological Well-being
52.6(11.4)
48.0(8.8) p=0.119
Moods&Emotions
49.6(11.4)
43.4(8.0) p=0.042*
Self-Perception
50.3(11.1)
45.1(5.5) p=0.067
Automony
50.0(10.6)
48.3(5.3) p=0.23
Parent Relations&Home Life
46.4(9.9)
43.9(6.1) p=0.314
Financial Resources
43.1(13.3)
42.1(11.2) p=0.785
Social Support&Peers
54.0(12.1)
47.6(9.1) p=0.041*
School Environment
54.7(11.0)
49.0(7.5) p=0048*
Social Acceptance(Bullying)
47.0(11.1)
41.3(11.1) p=0.057
60
58
56
54
52
50
48
46
44
42
40

Figure 1

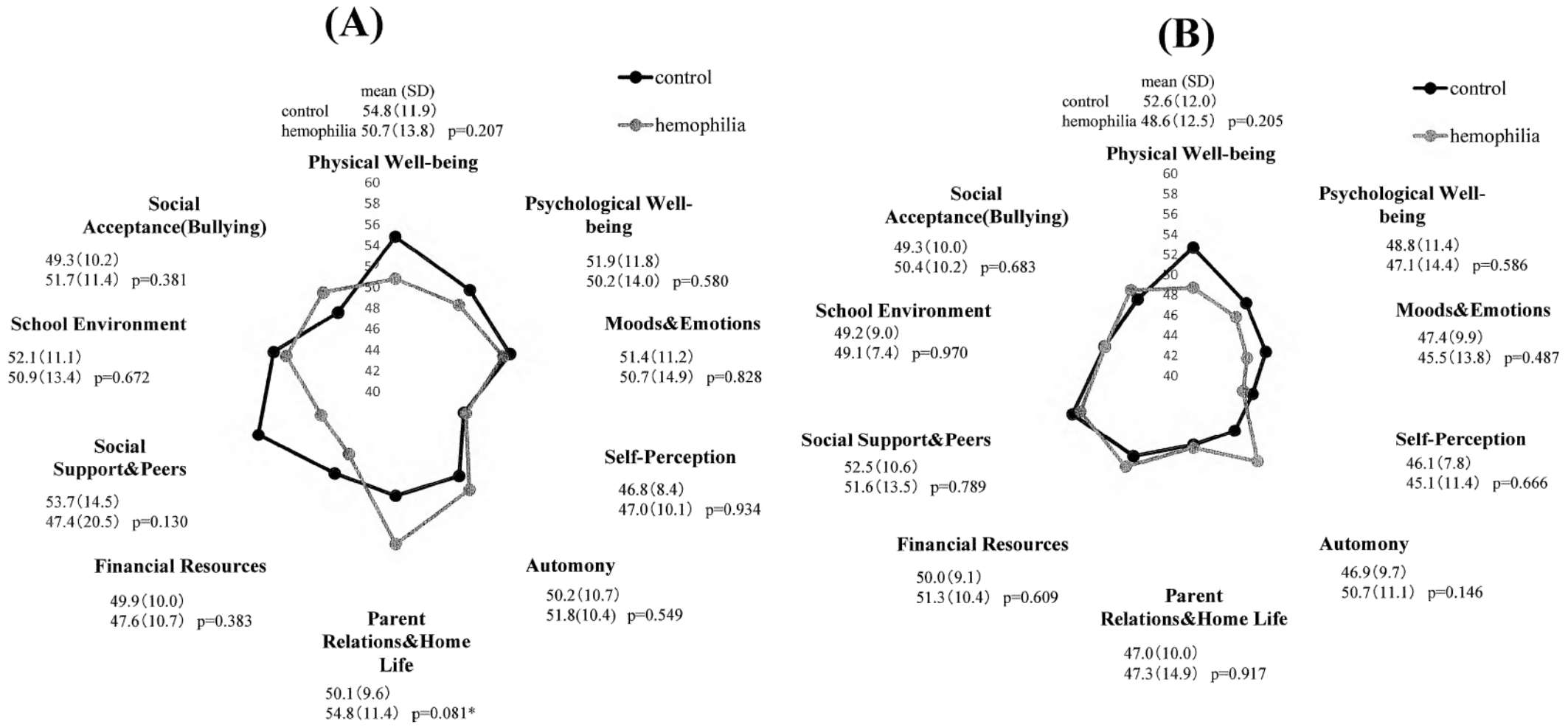
(A)
mean (SD)
control 54.8(11.9)
hemophilia 50.7(13.8) p=0.207
control
hemophilia
Physical Well-being
Social Acceptance(Bullying)
49.3(10.2)
51.7(11.4) p=0.381
Psychological Well-being
51.9(11.8)
50.2(14.0) p=0.580
School Environment
52.1(11.1)
50.9(13.4) p=0.672
Moods&Emotions
51.4(11.2)
50.7(14.9) p=0.828
Social Support&Peers
53.7(14.5)
47.4(20.5) p=0.130
Self-Perception
46.8(8.4)
47.0(10.1) p=0.934
Financial Resources
49.9(10.0)
47.6(10.7) p=0.383
Automony
50.2(10.7)
51.8(10.4) p=0.549
Parent Relations&Home Life
50.1(9.6)
54.8(11.4) p=0.081*
60
58
56
54
50
48
46
44
42
40
(B)
mean (SD)
control 52.6(12.0)
hemophilia 48.6(12.5) p=0.205
control
hemophilia
Physical Well-being
Social Acceptance(Bullying)
49.3(10.0)
50.4(10.2) p=0.683
Psychological Well-being
48.8(11.4)
47.1(14.4) p=0.586
School Environment
49.2(9.0)
49.1(7.4) p=0.970
Moods&Emotions
47.4(9.9)
45.5(13.8) p=0.487
Social Support&Peers
52.5(10.6)
51.6(13.5) p=0.789
Self-Perception
46.1(7.8)
45.1(11.4) p=0.666
Financial Resources
50.0(9.1)
51.3(10.4) p=0.609
Automony
46.9(9.7)
50.7(11.1) p=0.146
Parent Relations&Home Life
47.0(10.0)
47.3(14.9) p=0.917
60
58
56
54
52
50
48
46
44
42
40

Figure 2