

Predictors of Quality of Life among Adolescent and Young Adults with a Bleeding Disorder, 2012

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Background

• Little is known about what factors predict better quality of life (QoL) among persons with a bleeding disorder, especially among adolescents and young adults (AYAs).

Objective

• We describe factors related to QoL among AYAs diagnosed with Hemophilia A or B or von Willebrand disease (VWD).

Materials and Methods

Study Population

• Cross-sectional, online survey from a convenience sample of AYA persons with hemophilia (PWH) or VWD.

• Participants had to i) be aged 13-25 years, ii) read, write, and speak English, and iii) have Hemophilia A, B, or VWD.

• Recruitment occurred at major US hemophilia meetings (e.g., Inhibitor Summits and NHF meetings), US hemophilia treatment centers (HTC), and through a Facebook™ page dedicated to the study from April through December 2012.

Measuring Quality of Life (self-reported SF-36)

• QoL was measured using the 36-Item Short Form Health Survey (SF-36) mental (MCS) and physical composite summary (PCS) scores [1].

Measuring Chronic Pain (self-reported FPS-R)

• Chronic pain was measured using the revised Faces Pain Scale (FPS-R). The FPS-R is a visual scale composed of 6 faces illustrating an increasing level of pain intensity [2].

• Chronic pain was defined as: 'pain that you have every day or almost every day, and that always or almost always seems to be there even when you are not having a bleed at that moment.'

• For purpose of analysis, chronic pain was dichotomized as high for those who reported their pain as 'moderate,' 'severe,' 'very severe,' or 'worst pain possible' (i.e., FPS-R ≥4) and low for 'mild pain' or 'no pain' (i.e., FPS-R <4).

Measuring Adherence (self-reported VERITAS-Pro)

• Adherence was assessed using the Validated Hemophilia Regimen Treatment Adherence Scale (VERITAS)-Pro [3] and VERITAS-PRN [4] for prophylactic and on-demand (i.e., episodic) participants, respectively.

• VERITAS scores range from 24 (most adherent) to 120 (least adherent).

• As an experimental measure, we also combined VERITAS-Pro and VERITAS-PRN responses into one category [5].

Other Self-reported Data Collected

• Age, gender, race/ethnicity, health insurance status & type, and educational level of participant's parents.

• Data were also collected about bleeding disorder type (Hemophilia A or B, or VWD), whether or not the participant ever developed an inhibitor to treatment, and bleeding disorder severity.

Statistical Analysis

• Statistical association with SF-36 scores was assessed using the non-parametric Wilcoxon rank sum test or Kruskal Wallis test.

• Because the primary outcome variables of interest (SF-36 PCS and MCS scores) were largely skewed, multivariable, quantile regression models were used to assess factors associated with PCS and MCS scores.

• All statistical analyses were performed using SAS 9.2 (Cary, NC) and STATA 12 (College Station, TX).

• All p-values were calculated using two-sided tests.

Results

• Overall, 108 AYAs participated. Of which, 85 (79%), 5 (7%), and 15 (14%) had Haemophilia A, B, and VWD, respectively.

• Those with mild, moderate, and severe disease made up 20%, 8%, and 71% of the population, respectively.

• Most (65%) had never developed an inhibitor and treated prophylactically (68%). Half were aged 13-17 years, most were white (80%), non-Hispanic (89%), and never married (94%). The majority (94%) had some type of health insurance.

• Chronic pain was reported as moderate-to-severe (FPS-R ≥4) for 31% of respondents.

• Median PCS and MCS were 81.3 (IQR: 61.1–93.1; range: 12.9–100) and 75.5 (IQR: 60.0–84.3; range: 27.1–100), respectively (Figures 1 & 2).

• Mean values for PCS, MCS, and the eight multi-item subscales were generally lower than the median (except VT and SF) due to low outlying values (Figure 3).

Figure 1 Distribution of SF-36 Physical Composite Summary (PCS) Scores (n=108)

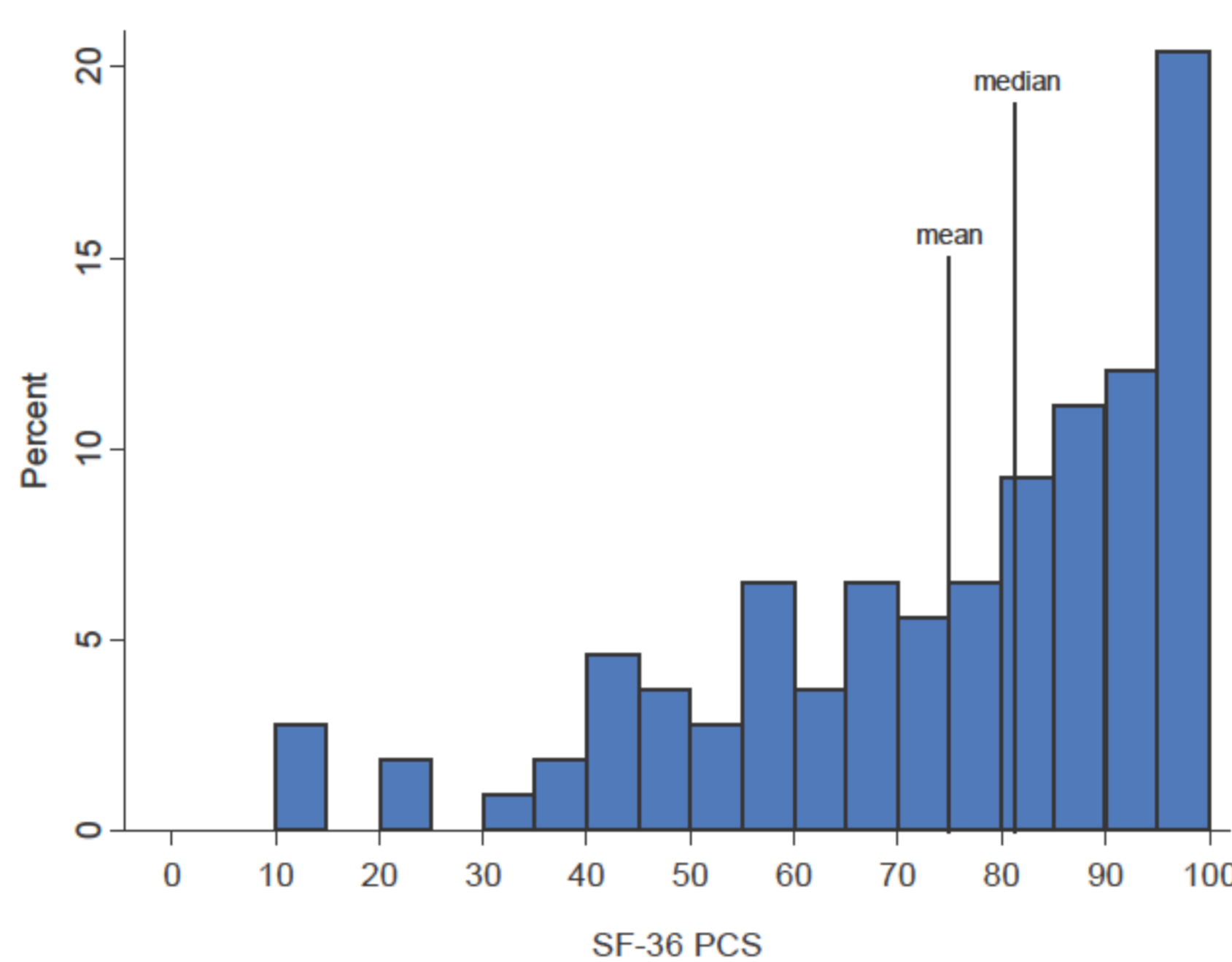


Figure 2 Distribution of SF-36 Mental Composite Summary (MCS) Scores (n=108)

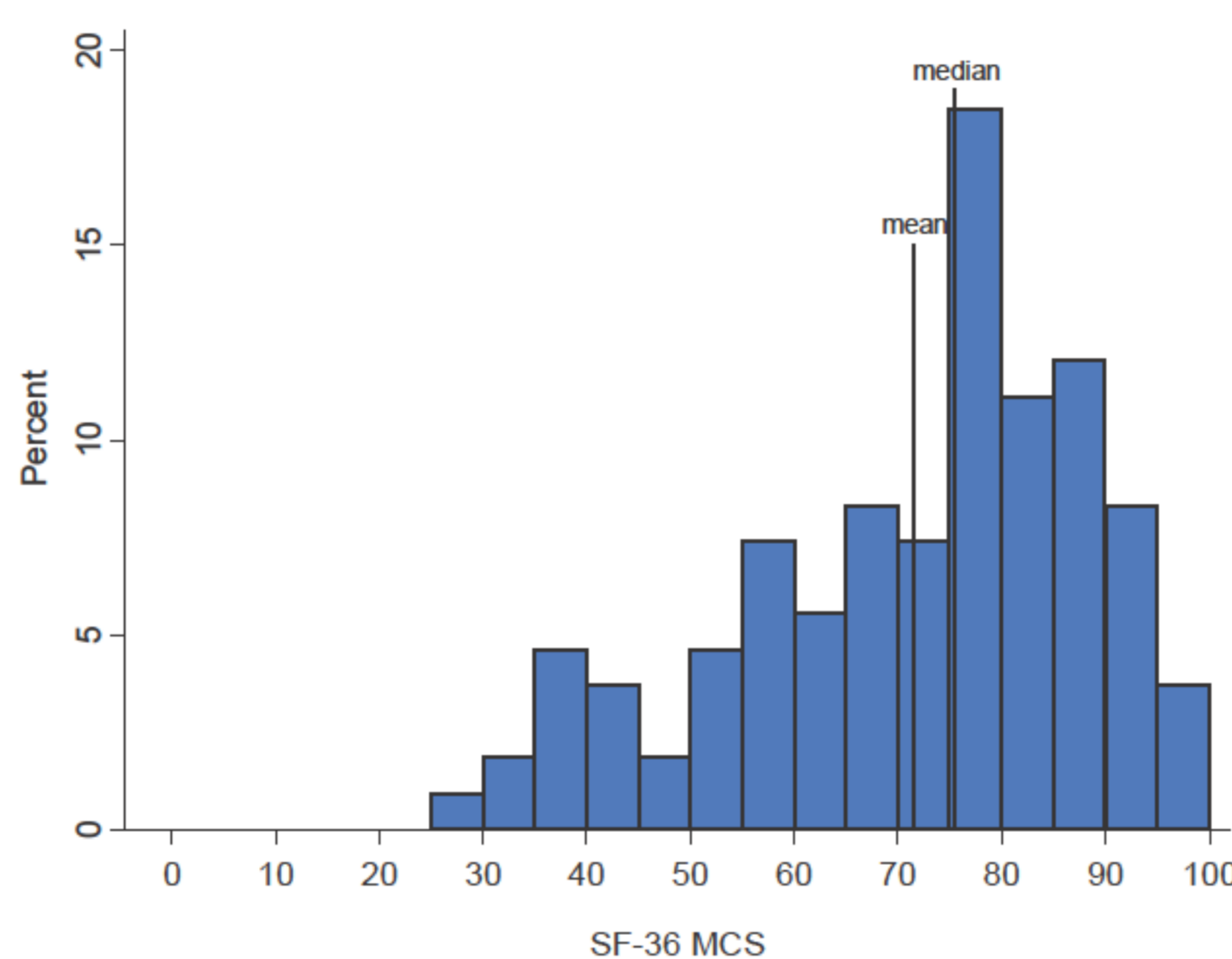
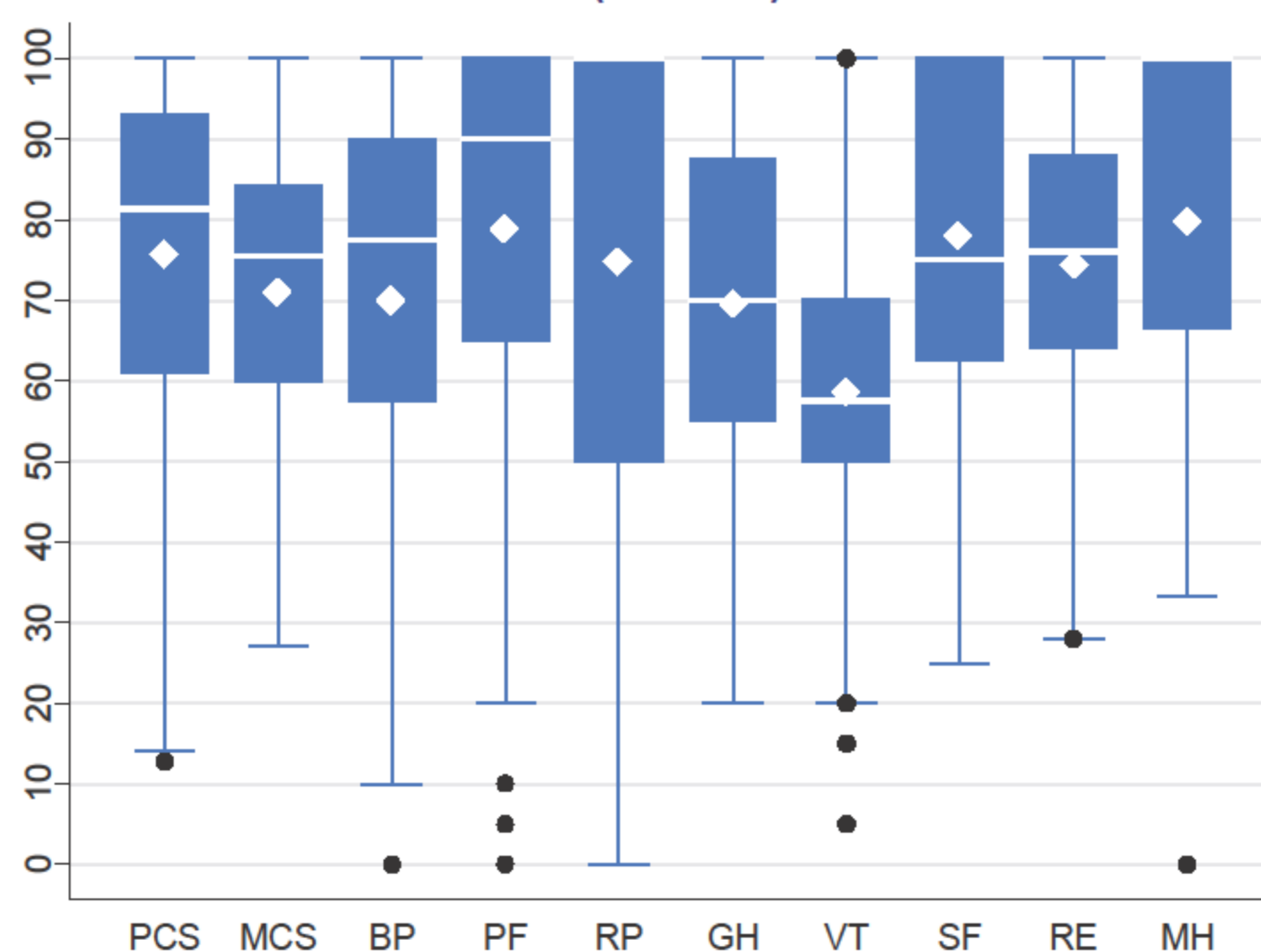


Figure 3 Box Plots for SF-36 Composite and Subscale Scores (n=108)



The SF-36 is composed of 8 multi-item scales assessing i) bodily pain (BP, 2 items), ii) physical function (PF, 10 items), iii) role limitations due to physical health problems (RP, 4 items), iv) general health (GH, 5 items), v) vitality (VT, 4 items), vi) social functioning (SF, 2 items), vii) role limitations due to emotional problems (RE, 3 items) and emotional well-being / mental health (MH, 5 items).

• At the univariate level, adolescents (vs. young adults), non-whites, those who reported ever developing an inhibitor, and those who reported moderate to severe (vs. none to mild) chronic pain had statistically significantly lower PCS scores (Figure 4).

• Adolescents (vs. young adults), those who reported moderate to severe (vs. none to mild) chronic pain, and those who were non-adherent to prescribed clotting-factor treatment regimens had statistically significantly lower MCS scores (Figure 4).

Figure 4 Median SF-36 Composite and Subscale Scores by Respondent Characteristic

*significant at p<.05 **significant at p<.01 ***significant at p<.001



• Final quantile regression modeling results for PCS and MCS scores are below (Tables 1 & 2).

Table 1 Quantile Regression Model Estimating Median SF-36 PCS Scores (n=108)

Characteristic	Coef. (95%CI)	p-value
Gender		
Female	-13.1 (-23.8, -2.4)	.02
Male	reference	
Inhibitor Development		
Ever	-13.1 (-21.5, -4.7)	<.01
Never	reference	
Chronic Pain^a		
Moderate to Severe	-25.5 (-33.8, -17.2)	<.001
None to Mild	reference	

^aChronic Pain was measured using the revised Faces Pain Scale (FPS-R) and was dichotomized as FPS-R<4 (i.e., 'mild' or 'no pain') and FPS-R≥4 (i.e., 'moderate' to 'worst pain possible').

Table 2 Quantile Regression Model Estimating Median SF-36 MCS Scores (n=108)^a

Characteristic	Coef. (95%CI)	p-value
Treatment Regimen		
Prophylaxis	10.0 (0.7, 19.3)	.04
On-demand	reference	
Chronic Pain^b		
Moderate to Severe	-10.0 (-19.2, -0.8)	.03
None to Mild	reference	

^aEthnicity (Hispanic vs. non-Hispanic) and history of inhibitor development (ever vs. never) were also included in the model because they increased the precision of the estimates.

^bChronic Pain was measured using the revised Faces Pain Scale (FPS-R) and was dichotomized as FPS-R<4 (i.e., 'mild' or 'no pain') and FPS-R≥4 (i.e., 'moderate' to 'worst pain possible').

Conclusions

• Efforts should be made to prevent and manage chronic pain, which was strongly related to both physical and mental QoL, in the AYA PWH and VWD populations.

• Previous research in this population has suggested that better adherence to prescribed treatment regimens is associated with less chronic pain [5].

• Future research should explore i) why women had lower physical QoL scores—even after adjustment for other sociodemographic and clinical factors, and ii) how prophylaxis may improve overall mental/emotional QoL in the AYA PWH and VWD populations.

[1] Ware JE, Jr, Sherbourne CD. *Med Care*. 1992; 30: 473-83.

[2] Hicks CL, et al. *Pain*. 2001; 93: 173-83.

[3] Duncan N, et al. *Haemophilia*. Mar 2010; 16(2):247-255.

[4] Duncan NA, et al. *Haemophilia*. 2010; 16: 47-53.

[5] McLaughlin, J.M., Witkop, M.L., et al., Better Adherence to Prescribed Treatment Regimen is Related to Less Chronic Pain among Adolescents and Young Adults with Moderate or Severe Haemophilia. *Haemophilia*. published online ahead of print Feb 11, 2014.

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