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Development of a tool to describe overall health, social independence and activity limitation of adolescents and young adults with disability

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Abstract

There is a need for research that focuses on the correlation between self-perceived quality of life (QoL) and the health outcomes of adolescents with disability transitioning to adulthood. To better understand the transition experience of adolescents and young adults with disability, we developed a questionnaire to assess the impact of disability on QoL. We recruited 174 participants who were 15–24 years old and diagnosed with Fragile X syndrome (FXS), spina bifida (SB) or muscular dystrophy (MD) and conducted an exploratory factor analysis to identify factors that characterize QoL. Five factors emerged: emotional health, physical health, independence, activity limitation, and community participation. To validate the tool, we linked medical claims and other administrative data records and examined the association of the factor scores with health care utilization and found the questionnaire can be utilized among diverse groups of young people with disability.

Keywords

Survey instrument; Quality of life; Rare conditions; Factor analysis; Fragile X syndrome; Muscular dystrophy; Spina bifida

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1. Introduction

There is a limited amount of research that focuses on the correlation between self-perceived quality of life (QoL), including emotional health, physical health, independence, activity limitation, and community participation, and the health outcomes of adolescents with disability transitioning to adulthood. The literature has summarized four tasks adolescents are expected to complete as they enter adulthood: establishing identity, forming relationships outside of the family unit, achieving independence from family, and finding a job (White, 1997). Adolescents and young adults may face challenges in completing these tasks depending on the nature and severity of their disability. While there is a growing body of literature surrounding the transition experience of people with disability, there is a need for an accessible and easily understood instrument that measures QoL of adolescents and young adults with disability.

In order to better understand the transition experience of adolescents and young adults with disability, we developed a questionnaire to assess the impact of disability on QoL. We administered the survey to people with three substantially different disabilities: fragile X syndrome (FXS), spina bifida (SB), or muscular dystrophy (MD). All three conditions are rare with prevalence below one per 10,000 people (Dicianno, Gaines, Collins, & Lee, 2009; Garber, Visootsak, & Warren, 2008; Hartley et al., 2011) The conditions cause some level of disability beginning in childhood, which can contribute to difficulties in the transition from adolescence to adulthood.

FXS is the leading cause of inherited intellectual disability (ID) and primarily impacts males. People with FXS do not generally have physical limitations that would require special accommodations needed by people with other types of ID (CDC, 2012a). SB is a congenital neural tube defect that frequently causes neurologic deficits below the level of the lesion, which may include paralysis. It is sometimes accompanied by hydrocephalus, which can result in neurodevelopmental complications (CDC, 2011). MD is a group of neuromuscular disabilities that include both childhood and adult onset. MD may involve progressive physical disability and declining mobility, cardiac and respiratory function (CDC, 2012b).

QoL questionnaires usually target either the general population or people with specific disorders. To be appropriate for large groups of people, generic QoL questionnaires do not include questions that would be of concern for persons with disability; this makes generic surveys inadequate for assessing the QoL of people with disability. In contrast, condition-specific surveys are sensitive to concerns of a particular population, but are difficult to use across populations (Dijkers, 1999; Guyatt et al., 1997; Liu et al., 2010; Rosenbaum & Saigal, 1996). The purpose of this study is to present the results of a validated QoL questionnaire that is general enough to be applied across all disability groups, but specific enough to address QoL concerns of individual disability groups.

2. Methods

The QoL survey for people with disability (specifically FXS MD, or SB) was developed in four phases: tool development, study recruitment, tool reduction, and tool validation.

2.1. Phase 1: tool development

The survey questions were selected from four validated and reliable surveys: the American Community Survey (ACS), the National Longitudinal Transition Study-2 (NLTS2), and the RAND-36 Measure of Health-Related Quality of Life (RAND-36).

The 14 demographic questions in our survey came from the ACS, which is part of the U.S. Census. Our survey also incorporated eleven questions from the RAND-36 designed to measure eight health concepts: physical functioning, role limitations caused by physical health problems, role limitations caused by emotional problems, social functioning, emotional well-being, energy/fatigue, pain, and general health problems. Nine questions that addressed mobility, medical care, condition type, use of help during the completion of the survey, and identification number to track responses were added by our research team. The remaining 120 questions included in our survey were taken from the NLTS2 in the sections identified as: social and leisure time activities, high school experiences, personal interests and activities, personal health, household, leaving high school, 2-year junior or community college, 4-year college or university, and jobs during last 2 years.

2.2. Phase 2: study recruitment

The same survey was administered to two different populations: US residents outside of SC and SC residents. To reach a national audience, we announced the survey through social media and a number of advocacy organizations. We enlisted a wide array of national organizations to post announcements in their newsletters and on their websites. The postings asked US residents who were 15–24 years of age, and diagnosed with SB, MD, or FXS to respond to an online questionnaire about their self-perceived health, social life, education, work experience and community participation. If needed, the participant was allowed to have help answering the questions. This was a convenience sample with no personal identifiers, but participants reported their age and state of residence. Only data collected on participants aged 15–24 was used in this analysis.

We conducted recruitment of South Carolina residents in a similar fashion, but also included medical providers in the recruitment process. We asked residents of South Carolina who had FXS, SB, or MD and were 15–24 years old to sign an informed consent form giving us permission to link their survey answers to data compiled as part of a larger study investigating the transition from adolescent to adult services for people with rare health conditions in South Carolina; details of which can be found in a methods paper (Royer et al., 2014). The data for this larger project including Medicaid and State Health Plan medical claims data is housed at the South Carolina Revenue and Fiscal Affairs Office, Health and Demographics (H&D). Data linkages and analyses were performed by H&D staff. South Carolina participants were compensated $50 for completing the survey. Seventy-seven people participated from South Carolina and we linked 64 participants to their medical claims, giving us an 83% match rate.

H&D created a dataset of people who were 15–24 years old between the years 2000–2010. In order to establish generalizability of our findings to the entire state, we asked H&D to begin with the 1038 people in the cohort that were 15–24 years old during the 2000–2010 study period. Because we only wanted to look at people who would qualify to participate in the survey during our recruitment period from 2012 to present, we eliminated people in the cohort who were born before 1988 and those who died before the recruitment period began; leaving 455 potential participants for the analysis. The 64/77 people who we could match to their medical claims were then compared to the remainder of the cohort.

There were no significant differences found between the recruited sample and the cohort of potential participants in the following areas: sex, race, county type, SES, work experience, education level, and visit counts. The only difference found was in the ‘‘conditions’’ category. We found that people with FXS (23/67 or 34.3%) participated more than people with MD (23/90 or 25.6%) than people with SB (31/298 or 10.4%), which is different from South Carolina’s distribution of the conditions (SB > FXS > MD).

2.3. Phase 3: tool reduction

Out of the 154 questions, 92 questions about high school, college, and work were excluded from the analyses due to the limited number of responses created by skip patterns. For example, if a person enrolled in college took the survey, the person did not answer questions about high school. Out of the remaining 62 questions, 30 questions were excluded since the questions addressed demographic information about the participant. Two questions were excluded because there was no variability in the responses among respondents. In total, 30 out of the 154 questions in the survey were included in the factor analysis. The maximum number of questions that a participant with specific characteristics could have answered is shown in Table 1.

We performed an exploratory factor analysis to understand the constructs describing the experience of people with three target conditions. The goal was to reduce the number of survey questions by finding correlated items and questions that explain most of the variation from the larger survey. In this way, we identified related questions that make up the various influences describing the transition experience of people with disability.

To identify the factors that contribute to QoL, we initially performed the principal factor analysis method with the selection of maximum priors option (‘priors = max’), and all factors were retained with eigenvalues greater than or equal to one (‘mineigen’ = 1). These criteria admitted five factors for which the absolute value of the loading value was greater than 0.45 indicating a fair loading. A strong factor is defined as one that has at least three questions loading on the factor. After rerunning the analysis including only those questions for which the absolute value of the loading factor was greater than 0.45 and those with communality estimates greater than 0.45, five factors were identified. Utilizing a promax rotation produced five factors with the same questions but with a slightly different factor composition. The promax rotation does not assume that the factors are independent, rather it allows correlation between the factors. This rotation method causes high loadings to become slightly smaller and lower loadings to disappear to nearly zero to simplify the structure for easy interpretation. We named the five factors emotional health (EH), physical health (PH), independence (IND), activity limitations (AL), and community participation (CP). There was very little cross-loading between the five factors.

After the factors were identified, we calculated and compared overall factor scores of the three disability groups (FXS, SB, and MD) to explore differences between groups. The questions within a factor were allowed to contribute equally to the factor score and then summed.

This created an overall factor score for each survey respondent. A factor score represents a subject's actual standing on an underlying factor. In our survey, a higher score reflects better QoL while a lower score reflects a poorer QoL. The smaller a score is, the more negatively the participant answered all the questions within a factor.

All of the statistical analyses were computed using SAS 9.4 statistical software. The survey questions were analyzed using Pearson correlation coefficients and exploratory factor analysis used the 'proc factor' command. To compare responses between disability groups, we estimated a one-way ANOVA model. To compare the mean scores for each pair of factors, we used Tukey's HSD adjustment to allow for an overall error rate of .05.

2.4. Phase 4: tool validation

To validate the study, we linked South Carolina participants with their medical claim records and other administrative data housed at H&D and examined the association of their factor scores with related information pertinent to each factor. All diagnoses and visits were averaged over number of years enrolled. We examined the association between the EH factor score and severe mental health diagnosis, any mental health diagnosis and number of behavioral health visits. We hypothesized that as mental health diagnoses or behavioral health visits increased, the EH factor score would decrease. We examined the association between the PH score and emergency room visits, number of inpatient hospitalizations, and number of medical care visits. Our hypothesis was that the higher the number of inpatient, emergency room or medical care, the lower the PH factor score. We examined the association between the IND factor score and whether or not the participant started 12th grade and whether the person was ever employed. For the IND factor, we believed that as the level of education increased for a person (calculated by the code for entry into 12th grade), the IND factor score would increase for all groups. We examined the association between the AL factor score and start of 12th grade, employment status, and durable medical equipment (DME) utilization. We believed that for all groups except MD as the number of DME prescribed increased, the AL score would decrease. We examined the association with the CP factor score and Community Long-Term Care (CLTC) services (such as assistance in bathing, dressing, and toileting that help individuals remain at home and avoid unnecessary nursing home placement) and home health services. The CP factor score was hypothesized to decrease as more home health services were utilized and increase as more CLTC services were utilized. Spearman and Pearson correlations were calculated. Significance levels were defined at a p-value of <0.05.

3. Results

The demographic characteristics of the survey participants are in Table 2. We obtained a similar distribution of the three disability groups. There are more male participants, as expected, since both FXS and MD primarily affect males. About 79% of our participants were white.

Out of the 30 questions included in the factor analysis, 20 questions had loadings above the threshold and loaded onto five factors. After examining the questions within each factor, we named the factors as follows: factor 1: emotional health (EH), factor 2: physical health (PH), factor 3: independence (IND), factor 4: activity limitation (AL) and factor 5: community participation (CP). The 10 questions whose loadings were below the threshold are not highly related to the five factors; however, each may represent an important independent variable that can be used in further analysis. The 20 question tool is included in Appendix A, and Appendix B contains the score key.

A list of the questions that comprise each factor, their factor loading values, and communality estimates are shown in Table 3. A loading value is the correlation between a question and the underlying factor (a higher correlation implies the question is more strongly related to the factor). Loading values for all variables were greater than 0.52. A communality estimate is the variance of an observed variable that is accounted for by the common factors. All of the communality values were 0.46 or higher.

A graphical representation of the differences in overall factor scores among the three disability groups is in Fig. 1. The FXS group reported the highest score on PH and the MD group reported the lowest score on PH. These differences among the three group factor score means were statistically significant. The SB group reported a significantly higher score on IND than the FXS group while the FXS and MD groups reported a statistically similar score. There were no statistical differences in factor scores for EH, AL, or CP among the three disability groups.

We also examined differences in mean scores on individual questions within factors among disability groups which is shown in Table 4. The percent variance associated with each factor is also listed in Table 4. These five factors accounted for 79.4% of the total variation in the survey. PH and EH each accounted for 20.1% and 20.5% of the total variation while IND accounted for 11.8%, AL accounted for 17.9%, and CP accounted for 9.0%.

Out of the four questions in the EH factor, the three groups differed significantly in one question. When asked about emotions/mood (happy, sad, nervous, worn out, etc.), the FXS group responded more positively than those with SB or MD; people with SB or MD responded similarly.

Group differences were found in all five questions of the questions in the PH factor. These differences were consistent with the characteristics of the conditions. For example, the FXS group reported better overall health and less pain than those with either MD or SB. When comparing their health to other people, participants with FXS responded positively while people with SB or MD responded negatively. It is expected for people with MD and SB to respond negatively to physical health questions since they have physical limitations related to their condition.

Within the four questions in the IND factor, the three groups differ in two questions. People with FXS reported less frequent interaction with friends by phone during the past year than participants with SB or MD. When asked about driver's licenses, allowances, credit cards, savings and checking account ownership, people with SB reported having more of these items than people with either MD or FXS, and people with MD or FXS reported a similar number of these items.

The three groups were found to be significantly different in two out of the four questions within the AL factor. Participants with FXS reported having less problems with their work or daily activities due to their physical health, and people with MD or SB reported similar problems. The MD group reported the most interference with social activities due to their physical health or emotional problems while the FXS and SB group reported slight interference. There were no differences found between the three groups in the CP factor.

To validate the self-reporting survey, we examined associations between factor score and service utilization for all three groups combined and each group separately. Using the linked administrative data, we validated four out of the five factors. Table 5 contains the correlations between the factor scores and services.

In regard to EH factor, the hypothesis was confirmed in all three of the populations for average severe mental health diagnoses. The EH factor was not confirmed for average any mental health diagnoses or total behavioral health visits for any of the populations. For average number of inpatient visits, our PH factor hypothesis was confirmed for those with SB. For average emergency room visits, the PH factor hypothesis was not confirmed in any of the populations. The association between the IND factor score and start of 12th grade was confirmed in the group with SB. There was no association found for those in the MD or FXS groups. The association between AL and DME utilization was confirmed for all populations. We were unable to confirm either of the hypotheses for the CP factor.

4. Discussion

We designed a survey to capture the transition experience of adolescents and young adults 15–24 years old with FXS, MD, or SB. We used exploratory factor analysis to identify key underlying constructs in the QoL for adolescents and young adults with disability. Our approach allowed us to explore the structure of each question without imposing any preconceived constraints on the responses. We explained 79.4% of the variation among all the questions in the factor analysis with just five factors based on 20 questions.

In comparing scores across disability types, we found that PH scores were the highest in participants with FXS, second highest in participants with SB, and the lowest in participants with MD. IND scores were higher for people with SB than for the other two groups. Additional research is warranted to identify the factors that contribute to the differences in health and social status across the different conditions and to develop strategies to address those underlying factors to improve the lives of young people with these disabilities.

A number of limitations to this study need to be considered. Our survey was not population-based. The respondents were volunteers who agreed to complete an online survey, thus we cannot comment on the representativeness of the sample. Second, exploratory factor analysis has some limitations including reliability of the measurement tool, sample size, and the sample selection. By using questions from reliable and validated sources, we minimized potential problems with reliability and validity. However, sample sizes for some questions on high school, college, work, and services and necessary accommodations were too small to be included in the exploratory factor analysis.

The survey tool and exploratory factor analysis add to our understanding of the experience of adolescents and young adults with the three disabilities we studied. Our analysis showed substantial differences between these diverse groups, revealing that the tool effectively assesses the QoL of adolescents and young adults with diverse disability. Therefore, it is expected that the examined parts of the survey tool would measure the self-reported EH, PH, CP, IND, and AL for young people with other disabilities.

Acknowledgements

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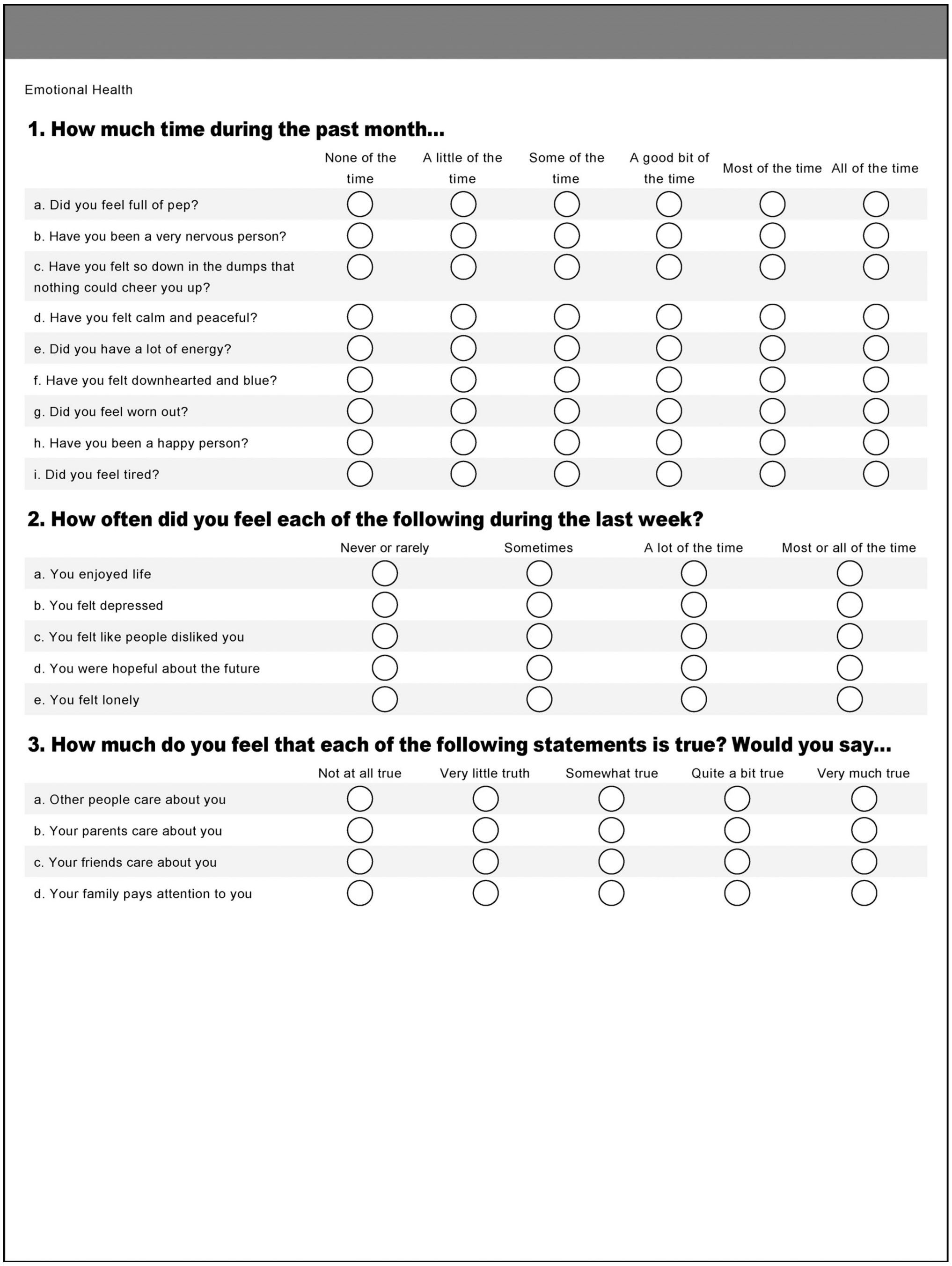
Disclosure

The findings and conclusions in this report are those of the authors and do not necessarily represent the views of the Centers for Disease Control and Prevention, the South Carolina Revenue and Fiscal Affairs Office, the South Carolina Department of Health and Human Services, the South Carolina Public Employee Benefit Authority, the South Carolina Department of Education or the South Carolina Department of Social Services.

Appendix A



Emotional Health

1. How much time during the past month...

	None of the time	A little of the time	Some of the time	A good bit of the time	Most of the time	All of the time
a. Did you feel full of pep?	○	○	○	○	○	○
b. Have you been a very nervous person?	○	○	○	○	○	○
c. Have you felt so down in the dumps that nothing could cheer you up?	○	○	○	○	○	○
d. Have you felt calm and peaceful?	○	○	○	○	○	○
e. Did you have a lot of energy?	○	○	○	○	○	○
f. Have you felt downhearted and blue?	○	○	○	○	○	○
g. Did you feel worn out?	○	○	○	○	○	○
h. Have you been a happy person?	○	○	○	○	○	○
i. Did you feel tired?	○	○	○	○	○	○

2. How often did you feel each of the following during the last week?

	Never or rarely	Sometimes	A lot of the time	Most or all of the time
a. You enjoyed life	○	○	○	○
b. You felt depressed	○	○	○	○
c. You felt like people disliked you	○	○	○	○
d. You were hopeful about the future	○	○	○	○
e. You felt lonely	○	○	○	○

3. How much do you feel that each of the following statements is true? Would you say...

	Not at all true	Very little truth	Somewhat true	Quite a bit true	Very much true
a. Other people care about you	○	○	○	○	○
b. Your parents care about you	○	○	○	○	○
c. Your friends care about you	○	○	○	○	○
d. Your family pays attention to you	○	○	○	○	○

4. How much is each statement below like you? Is each one not at all like you, a little like you, or very much like you?

	Not at all like me	A little like me	Very much like me
a. You are proud of who you are	○	○	○
b. You are a nice person	○	○	○
c. You can make friends easily	○	○	○
d. You can tell other people your age how you feel when they upset you or hurt your feelings	○	○	○
e. You feel useful and important	○	○	○
f. You feel your life is full of interesting things to do	○	○	○
g. You can handle most things that come your way	○	○	○
h. You know how to get the information you need	○	○	○
i. You can get school staff or other adults to listen to you	○	○	○

Physical Health

5. In general, would you say your health is...

○ Poor
○ Fair
○ Good
○ Very good
○ Excellent

6. Does your health now limit you in these activities? If so, how much?

	Yes, limited a lot	Yes, limited a little	No, Not limited at all
a. Vigorous activities, such as running, lifting heavy objects, participating in strenuous sports	○	○	○
b. Moderate activities, such as moving a table, pushing a vacuum cleaner, bowling or playing golf	○	○	○
c. Lifting or carrying groceries	○	○	○
d. Climbing several flights of stairs	○	○	○
e. Bending, kneeling or stooping	○	○	○
f. Bathing or dressing yourself	○	○	○

7. How much bodily pain have you had during the past month?

○ None
○ Very mild
○ Mild
○ Moderate
○ Severe
○ Very severe

8. During the past month, how much did pain interfere with school or your normal work (including both work outside the home and housework)?

○ Not at all
○ A little bit
○ Moderately
○ Quite a bit
○ Extremely

9. How true or false is each of the following statements about you?

	Definitely false	Mostly false	Don't know	Mostly true	Definitely true
a. I seem to get sick a little easier than other people	○	○	○	○	○
b. I am as healthy as anybody I know	○	○	○	○	○
c. I expect my health to get worse	○	○	○	○	○
d. My health is excellent	○	○	○	○	○

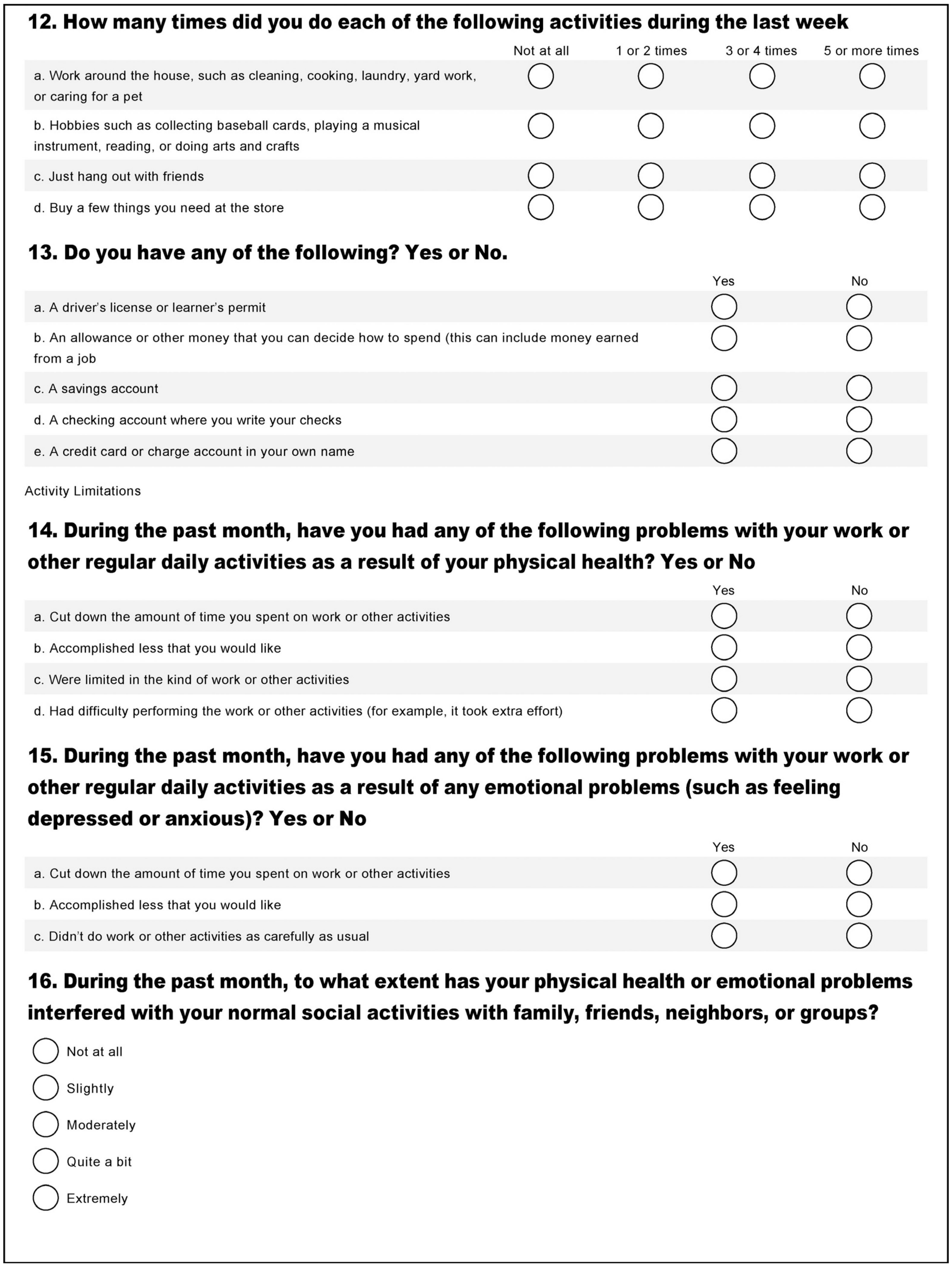
Independence

10. During the last year, about how many days a week have you usually gotten together with friends, outside of time you might spend at school and outside of organized activities or groups?

○ Never
○ Sometimes, but not every week
○ 1 day a week
○ 2 or 3 days a week
○ 4 or 5 days a week
○ 6 or 7 days a week

11. During the last year, about how often have friends called you on the phone?

○ Never
○ Rarely/less than once a month
○ A few times a month/but not every week
○ About once a week
○ Several days a week
○ Every day



12. How many times did you do each of the following activities during the last week

	Not at all	1 or 2 times	3 or 4 times	5 or more times
a. Work around the house, such as cleaning, cooking, laundry, yard work, or caring for a pet	○	○	○	○
b. Hobbies such as collecting baseball cards, playing a musical instrument, reading, or doing arts and crafts	○	○	○	○
c. Just hang out with friends	○	○	○	○
d. Buy a few things you need at the store	○	○	○	○

13. Do you have any of the following? Yes or No.

	Yes	No
a. A driver's license or learner's permit	○	○
b. An allowance or other money that you can decide how to spend (this can include money earned from a job	○	○
c. A savings account	○	○
d. A checking account where you write your checks	○	○
e. A credit card or charge account in your own name	○	○

Activity Limitations

14. During the past month, have you had any of the following problems with your work or other regular daily activities as a result of your physical health? Yes or No

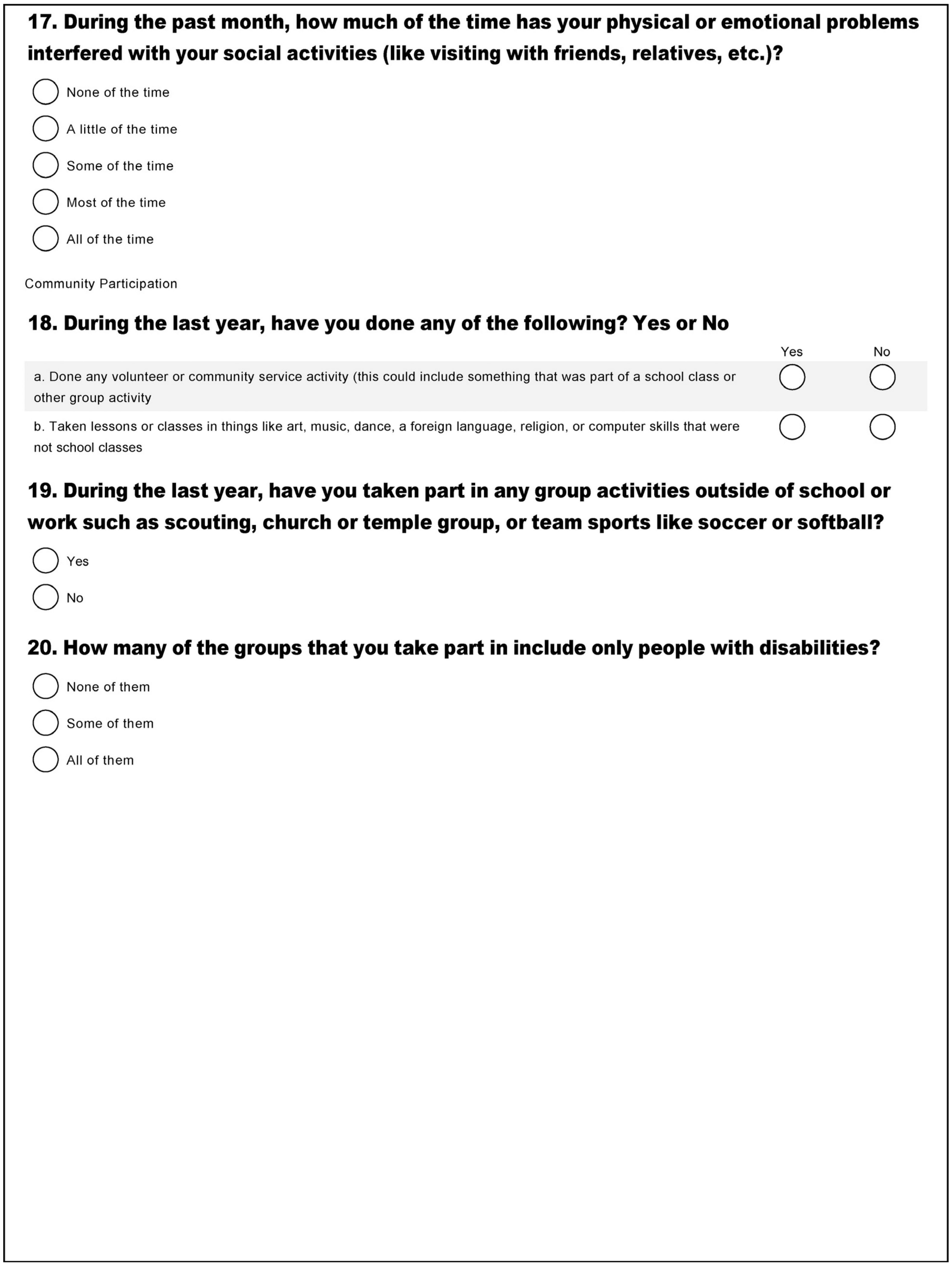
	Yes	No
a. Cut down the amount of time you spent on work or other activities	○	○
b. Accomplished less that you would like	○	○
c. Were limited in the kind of work or other activities	○	○
d. Had difficulty performing the work or other activities (for example, it took extra effort)	○	○

15. During the past month, have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)? Yes or No

	Yes	No
a. Cut down the amount of time you spent on work or other activities	○	○
b. Accomplished less that you would like	○	○
c. Didn't do work or other activities as carefully as usual	○	○

16. During the past month, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbors, or groups?

○ Not at all
○ Slightly
○ Moderately
○ Quite a bit
○ Extremely



17. During the past month, how much of the time has your physical or emotional problems interfered with your social activities (like visiting with friends, relatives, etc.)?

○ None of the time

○ A little of the time

○ Some of the time

○ Most of the time

○ All of the time

Community Participation

18. During the last year, have you done any of the following? Yes or No

	Yes	No
a. Done any volunteer or community service activity (this could include something that was part of a school class or other group activity	○	○
b. Taken lessons or classes in things like art, music, dance, a foreign language, religion, or computer skills that were not school classes	○	○

19. During the last year, have you taken part in any group activities outside of school or work such as scouting, church or temple group, or team sports like soccer or softball?

○ Yes

○ No

20. How many of the groups that you take part in include only people with disabilities?

○ None of them

○ Some of them

○ All of them

Appendix B

Scoring for survey tool (sum each section for factor score and sum each factor score for total score)

Emotional health score						
Q1: a, d, e, h	None = 0	A little = 1	Some = 2	A good bit = 3	Most = 4	All = 5
Q1: b, c, f, g, i	None = 5	A little = 4	Some = 3	A good bit = 2	Most = 1	All = 0

Q2: a, c	Never = 1	Sometimes = 2	A lot = 3	Most or all = 4		
Q2: b, d, e	Never = 4	Sometimes = 3	A lot = 2	Most or all = 1		
Q3: all	Not at all = 1	Very little = 2	Somewhat = 3	Quite a bit = 4	Very much = 5	
Q4: all	Not at all = 0	A little = 1	Very much = 2			
Physical health score						
Q5	Poor = 1	Fair = 2	Good = 3	Very good = 4	Excellent = 5	
Q6: all	Yes, a lot = 1	Yes, a little = 2	No, not at all = 3			
Q7	None = 6	Very mild = 5	Mild = 4	Moderate = 3	Severe = 2	Very severe = 1
Q8	Not at all = 5	A little bit = 4	Moderately = 3	Quite a bit = 2	Extremely = 1	
Q9: a, c	Definitely false = 2	Mostly false = 1	Do not know = 0	Mostly true = −1	Definitely true = −2	
Q9: b, d	Definitely false = −2	Mostly false = −1	Do not know = 0	Mostly true = 1	Definitely true = 2	
Independence score						
Q10	Never = 0	Sometimes = 1	1 day a week = 2	2–3 a week = 3	4–5 a week = 4	6–7 a week = 5
Q11	Never = 0	Rarely = 1	A few times = 2	once a week = 3	Several days = 4	Every day = 5
Q12	Not at all = 0	1–2 times = 1	3–4 times = 2	5 + times = 3		
Q13	No = 0	Yes = 1				
Activity limitations score						
Q14 & Q15	No = 1	Yes = 0				
Q16	Not at all = 5	Slightly = 4	Moderately = 3	Quite a bit = 2	Extremely = 1	
Q17	None = 5	A little = 4	Some = 3	Most = 2	All of the time = 1	
Community participation score						
Q18 & Q19	No = 0	Yes = 1				
Q20	None = 1	Some = 2	All = 3			

Sum each section for a factor score. Sum each factor score for total score. A higher score on any factor is considered a good score. The lower the score, the more problems or difficulty the individual reported.

References

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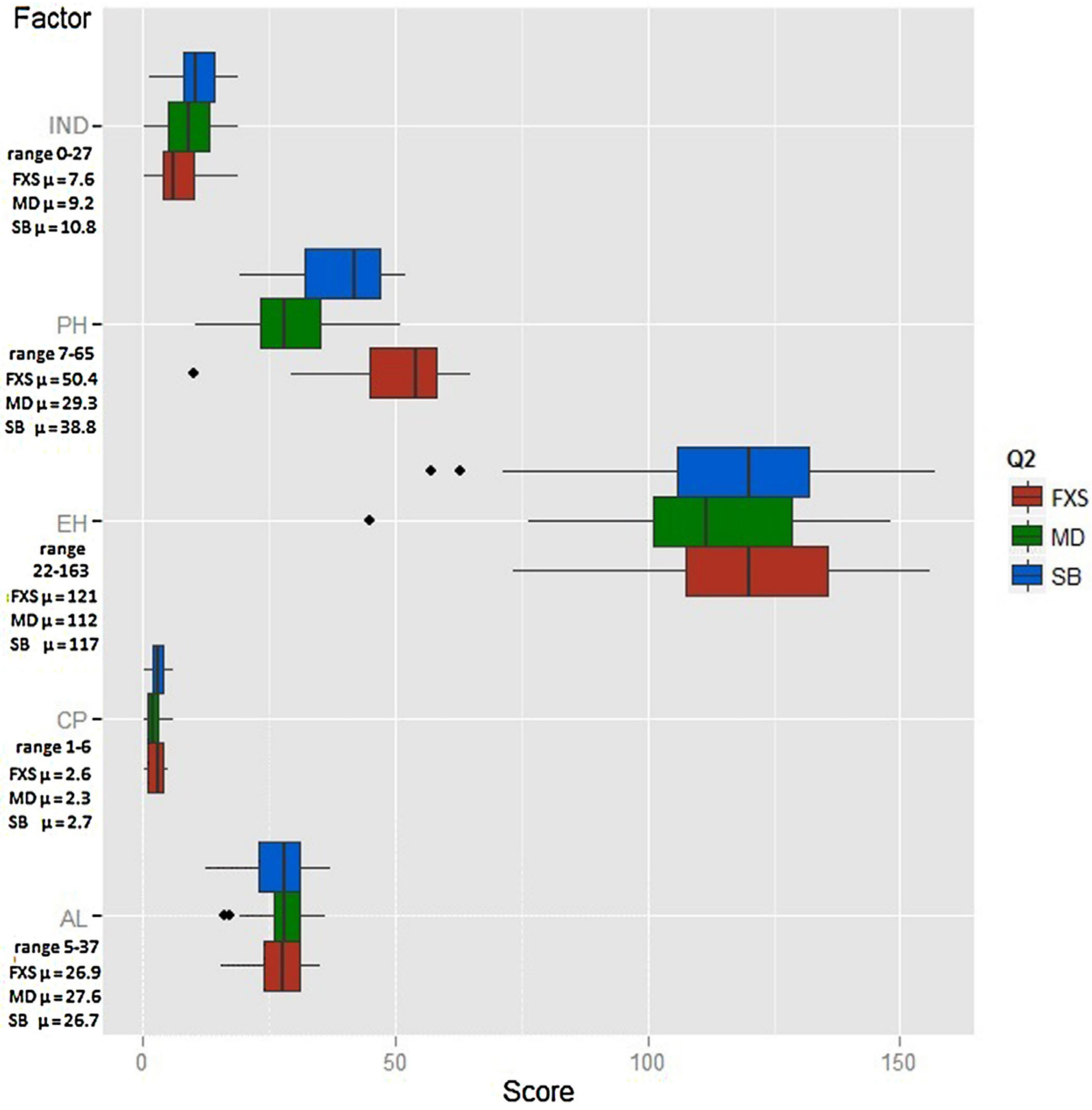

Fig. 1.

Distribution of factor scores by condition. EH: emotional health; PH: physical health; IND: independence; AL: activity limitations; CP: community participation. A higher score on any factor is considered a good score. The lower the score, the more problems or difficulty the group expressed.

Table 1

Maximum number of questions answered by participant type.

	Number of questions
Everyone	64
Additional questions	
If in high school	8
If not in high school	4
Graduate high school, not in college	0
Graduated high school and in college	36
If not graduated high school	5
If never had a job	1
If had job or has job now	36
Total number of questions in survey	154

Table 2

Characteristics of survey participants.

	***N* = 174**	**Percent**
Gender		
Male	112	64.4
Female	62	35.6
Disability		
Spina bifida	66	37.9
Muscular dystrophy	62	35.6
Fragile X syndrome	46	26.4
Age group		
15–19	98	56.3
20–24	76	43.7
Race		
White	138	79.3
Black	28	16.1
Other	8	4.6

Table 3

Loading and communality values of survey items for the five-factor model (*N* = 174) from the factor analysis.

Rotated factors	**Loading**[a]	**Communality**[b]
Factor 1: emotional health		
Q1: mood and feelings in past month	0.70	0.76
Q2: feelings in past week	0.85	0.79
Q3: perception of others	0.85	0.74
Q4: personal feelings about social interaction	0.80	0.74
Factor 2: physical health		
Q5: overall health	0.69	0.55
Q6: health limitations in activities	0.71	0.61
Q7: bodily pain during past month	0.79	0.69
Q8: pain interference with school or work	0.75	0.63
Q9: self-reported health statements	0.78	0.61
Factor 3: independence		
Q10: frequency gotten together with friends outside school/work	0.74	0.56
Q11: how often friends called on the phone past year	0.54	0.54
Q12: frequency of activities during last week	0.76	0.61
Q13: driver'slicense, allowance, credit card, bank account	0.74	0.61
Factor 4: activity limitations		
Q14: work or daily activity limitations due to emotional health	0.94	0.91
Q15: work or daily activities problems due to physical health	0.95	0.91
Q16: physical or emotional health interferes with social activities	0.56	0.62
Q17: social and recreational activity	0.57	0.60
Factor 5: community participation		
Q18: community service, lessons, and classes	0.78	0.62
Q19: group activities	0.80	0.67
Q20: groups with disability	0.69	0.52

[a] A loading value is the correlation between a question and the underlying factor (a higher the correlation implies the question is more strongly related to the factor).

[b] A communality estimate is the variance of an observed variable that is accounted for by the common factors (a communality of .69 for Q10 means that 69% of the variance in this question is accounted for by the common factor of Physical Health).

Table 4

Participants mean score[a] and overall factor score compared by disability group.

Factor (% variance explained[b])	Question #	Score range	FXS mean	MD mean	SB mean	*p*-Value[c]
Emotional health (20.53%)	Q1	0–45	31.8	27.3	27.7	0.0028[*]
	Q2	5–20	17.0	15.7	16.1	0.0666
	Q3	5–20	18.5	17.7	17.7	0.1974
	Q4	0–18	12.0	12.4	13.3	0.2004
	Overall score	10–103	80.0	73.6	74.8	0.0561
Physical health (20.14%)	Q5	1–5	3.9	3.0	3.2	<0.0001[*]
	Q6	7–21	18.5	9.1	13.5	<0.0001[*]
	Q7	1–6	5.4	4.1	4.3	<0.0001[*]
	Q8	1–5	4.7	4.1	4.0	0.0003[*]
	Q9	−8 to 8	2.8	−0.5	0.9	<0.0001[*]
	Overall score	2–45	35.5	19.8	25.8	<0.0001[*]
Independence (11.82%)	Q10	0–5	1.3	1.7	1.6	0.2229
	Q11	0–5	1.2	2.0	2.5	0.0002[*]
	Q12	0–12	3.6	3.7	4.0	0.6122
	Q13	0–5	1.4	2.1	2.7	<0.0001[*]
	Overall score	0–27	7.6	9.2	10.8	0.0027[*]
Activity limitations (17.8982%)	Q14	0–4	3.2	2.5	2.7	0.0498[*]
	Q15	0–3	2.3	2.2	2.2	0.7765
	Q16	1–5	4.0	3.5	3.9	0.0479[*]
	Q17	1–5	4.1	3.7	4.0	0.1137
	Overall score	2–17	13.6	11.9	12.7	0.1079
Community participation (8.98)	Q18	0–3	0.8	0.7	0.8	0.9109
	Q19	0–1	0.4	0.4	0.5	0.2588
	Q20	1–3	1.4	1.2	1.4	0.747
	Overall score	1–7	2.6	2.3	2.7	0.2138

[*] Significance difference at the .05 level.

[a] A larger mean value indicates good health/participation while a smaller mean value indicates worse health/less participation.

[b] Percent variance explained in overall survey by each factor.

[c] *p*-Values from one-way ANOVA comparison of mean scores of the three disability groups.

Table 5

Spearman/Pearson correlation coefficients for factor scores and related health information for survey participants linked to Medicaid data.

	All conditions	**FXS**	**MD**	**SB**
EH & Avg. severe MH counts	−0.14	**0.54**	**−0.81**	**−0.45**
EH & Avg. any MH counts	0.15	−0.15	−0.11	0.23
EH & Behavioral Health	−0.12	0.01	−0.28	−0.19
PH & Avg. IP visits/years enrolled	−0.23	−0.30	−0.22	**−0.46**
PH & Avg. ER/years enrolled	−0.10	−0.19	−0.13	**−0.25**
PH & Avg. medical visits	−0.14	0.09	0.18	0.02
IND & Start 12th grade	0.14	0.29	−0.36	**0.46**
AL & Avg. DME	**−0.25**	−0.17	0.11	−0.09
CP & Home Health	0.20	0.26	−0.02	0.22
CP & CLTC	0.14	0.25	−0.02	0.03

EH: emotional health; PH: physical health; IND: independence; AL: activity limitations; CP: community participation; MH: mental health; IP: inpatient; ER: emergency room; DME: durable medical equipment; CLTC: community long term care. Bolded correlations indicate significant associations.