Title: A network analysis of potential antecedents and consequences of pain-related activity-avoidance and activity-engagement in adolescents

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Abstract

Objective: This study sets out to identify potential daily antecedents and consequences of pain-related activity-avoidance and -engagement behavior in adolescents with chronic pain.

Methods: Adolescents ($N = 65$; $M_{age} = 14.41$) completed baseline self-reports and a diary for 14 days. Afternoon and evening reports were used to infer a network structure of within-day associations between pain intensity, pain-related fear, pain catastrophizing, affect, and pain-related activity-avoidance and -engagement behavior. Baseline psychological flexibility was examined as a potential resilience factor. **Results:** Activity-avoidance in the evening was predicted by pain-related fear and avoidance earlier that afternoon. Activity-engagement was predicted by positive affect and activity-engagement in the afternoon. Pain-related behavior in the afternoon was not related to subsequent changes in pain intensity, pain-related fear, pain catastrophizing, and affect. Pain-related fear in the afternoon was predictive of increased levels of pain and pain catastrophizing in the evening. Both pain-related fear and pain catastrophizing in the evening were predicted by negative affect in the afternoon.

Psychological flexibility was associated with lower levels of daily activity-avoidance, and buffered the negative association between pain intensity and subsequent activity-engagement.

Conclusion: This study provides insight into unique factors that trigger and maintain activity-avoidance and -engagement, and into the role of psychological flexibility in pediatric pain.

Future work should focus on both risk and resilience factors, and examine the role of psychological flexibility in chronic pediatric pain in greater detail.

Keywords: chronic pediatric pain; avoidance; activity-engagement; diary; network analyses

Introduction

Adolescents with chronic pain often experience moderate to severe restrictions in their daily functioning (1). Several cognitive, affective, and behavioral factors have now been identified which undermine adolescents' physical, emotional, school, and/or social functioning (2–6). One factor in particular, avoidance of pain-related activities, has received considerable attention in the literature (2,4). According to the Fear-Avoidance Model, which was developed in the context of adult pain [FAM; (7,8)], unwanted experiences such as anticipated pain, catastrophic thoughts, and pain-related fear, set the stage for subsequent avoidance behavior (2–4). This model predicts that avoidance of pain-related events serves to reduce contact with pain, fear, and catastrophizing. As a result, the probability that an individual will avoid these and other pain-related events across time increases (7,9). While certainly useful in the short-term, avoidance has many negative consequences in the long-term, such as increased disability, which heightens one's risk of maladaptive functioning (e.g., depression) (10–12). A similar process has also been found to operate in pediatric pain (2–4).

Chronic pain research has long focused on the role of avoidance and poor functional outcomes. However, many adolescents report intense, persistent pain and yet few impairments in their daily functioning (1,13). This has led researchers to search for cognitive, affective, and behavioral factors that promote adaptive outcomes. One such factor, engagement in valued or important activities regardless of the pain level, may be central to successful functioning in adolescents with chronic pain (12,14–16). For instance, activity-engagement in the presence of pain has been associated with improvements in disability and depression (16,17). So far the Fear-Avoidance Model has mainly focused on thoughts and behaviors (e.g., catastrophizing, avoidance) that increase the probability of maladaptive outcomes rather

than thoughts and behaviors which increase the probability of adaptive outcomes. However, recent updates to the model are starting to emphasize that the path to recovery begins by prioritizing valued life goals and engaging in previously avoided, important activities (18). This new focus on activity-engagement calls for research to identify other factors that decrease the risk of maladaptive outcomes and increase the probability of sustained adaptive functioning (13,19–23). One promising factor is psychological flexibility, which is broadly defined as being aware of, and open to, unwanted and uncontrollable inner experiences (such as chronic pain), while still being able to act in-line with what one values in life (15,24). Psychological flexibility is a central process in Acceptance and Commitment Therapy (25), a therapy which has proven valuable in increasing adaptive functioning in adults with chronic pain (26). Recent studies show similar effects in adolescents with chronic pain (27,28).

In this study we sought to identify the potential antecedents and consequences of pain-related activity-avoidance and activity-engagement using a daily diary methodology (29) in combination with a network analytic approach (30–33). This methodology captures momentary thoughts, feelings, and actions as they occur in the daily life of adolescents with chronic pain. We collected daily assessments of pain intensity, pain-related fear, pain catastrophizing, and (positive/negative) affect (2,4) and examined their respective relations with daily activity-avoidance and -engagement. We also examined if psychological flexibility influenced the strength of these daily associations with pain-related behavior. Drawing on the Fear-Avoidance model, we forward a number of hypotheses. First, higher levels of pain intensity, pain catastrophizing, pain-related fear, and negative affect should predict higher levels of activity-avoidance. Second, higher levels of activity-avoidance should predict lower levels of pain, pain catastrophizing, pain-related fear, and negative affect at a later point in time. We had no *a priori* hypotheses about how these same factors would relate to activity-

engagement given this has not been examined previously. Thus these latter relations are examined exploratory. Finally, we examined the potential resilience-enhancing role of psychological flexibility. We expected, based on previous cross-sectional work (34,35), that higher levels of psychological flexibility would predict lower levels of activity-avoidance and higher levels of activity-engagement on a daily basis. We also hypothesized that psychological flexibility would moderate the impact of pain, pain-related fear, and pain catastrophizing on activity-avoidance and activity-engagement at the within-day level. To the best of our knowledge, this is the first study to longitudinally examine the potential role of psychological flexibility as a resilience factor in the daily lives of adolescents with chronic pain.

Methods

Participants

Participants were adolescents with mixed chronic pain conditions recruited from two pediatric pain clinics in the USA. Recruitment occurred when they presented for initial clinical evaluation in the Pain Treatment Service at Boston Children's Hospital (BCH) between February 2017 and December 2017, and in the Pediatric Pain Management Clinic at Stanford Children's Health (SCH) between February 2017 and February 2018. Institutional Review Board (IRB) approval was granted at each site prior to the start of the study (BCH IRB#P0020989; Stanford IRB#39092). The present study is part of a larger research project (Child Pain In Context (CP-IC) study) for which the primary caregiver (e.g., parent/guardian) of each adolescent was also asked to participate (see <http://hdl.handle.net/1854/LU-8578159> for the complete study protocol). For the present study we only examined adolescent data.

Eligibility criteria for participation were [1] being 11 to 17 years old, [2] reporting persistent or recurrent pain for 3 months or longer, [3] having internet access at home or on an accessible mobile phone, [4] no significant cognitive impairments (e.g., intellectual disability, severe brain injury), and [5] no severe psychiatric or neurological conditions.

Eighty-four adolescents who fulfilled the inclusion criteria (i.e., 95%) agreed to participate, and were asked to complete a set of baseline self-report questionnaires followed by a 14-day diary assessment period. Prior to the self-reports, one participant was excluded due to a lack of interest in taking part, another for failing to respond after the first contact, and a further two due to retraction of consent for unknown reasons. Another seventeen withdrew during the self-report phase. Of these, thirteen were unresponsive following repeated prompts to complete the self-reports, one reported difficulty in completing the questions, one reported loss of interest in taking part, one withdrew due to the parent's concerns, and one withdrew for unknown reasons. Two participants withdrew after completing the self-reports due to severe health issues or absence of pain at the start of the diary. This left a final sample of sixty-five adolescents with chronic pain.

Study procedure

Participants received an online link to access the baseline self-report questionnaires (either via text message or e-mail). Once self-reports were completed, the diary period was scheduled to begin the following week. Automatic messages containing the diary surveys were sent to participants each afternoon and evening for 14 consecutive days, either via text message or e-mail. Study data were collected and managed using the REDCap (Research Electronic Data Capture) (36) tool hosted at BCH and Stanford University. REDCap is a secure, web-based application designed to support data capture for research studies.

Afternoon surveys were sent at 2 pm and deactivated at 6 pm, and evening diaries were sent at 6 pm and deactivated at 10 am the next day. In line with recommendations by Nezlek (34; p.46), all surveys completed between these time windows were treated as valid reports.

Participants who did not complete two consecutive diary assessments were contacted by the research assistant to prompt completion. All communication with the under-aged participants was carried out via the primary caregiver. If participants did not complete any of the required diary assessments on three consecutive days despite reminder calls, they were given the option of withdrawing from the study. If they decided to continue and failed to provide data on any additional days after this final reminder, their participation was terminated and they received no further diary invitations.

Participants who started the two-week diary period received a 10-dollar gift voucher at the end of the first week irrespective of the number of completed days. This was intended to serve both as a sign of appreciation for their participation, as well as an incentive to complete diary assessments in the second week. Participants received a 20-dollar gift voucher at the end of week two unless they withdrew from the study during the first week.

Measures

Baseline questionnaires

Participants completed a set of questionnaires measuring demographic information and baseline levels of several variables prior to the diary start-point.

Demographics were obtained by asking both the adolescent and their participating

parent/caregiver to complete a short questionnaire assessing age, gender, ethnicity, race, and schooling grade. Other demographic information (e.g., pain location, duration, and treatment) was gathered by means of a screening form to ensure participant eligibility for the study.

Pain intensity and disability were assessed using items from the child version of the Graded Chronic Pain Scale [GCPS; (1,38)]. Current and average pain intensity in the past six months were rated on a 11-point numerical scale (0 = *no pain*, 10 = *worst possible pain*). Disability was measured in terms of disability points. These points reflect a sum score of points allocated to the total number of days on which the adolescent was prevented from carrying out usual activities in the past six months (0: < 7 days; 1: ≥ 7 and < 15 days; 2: ≥ 15 and < 31 days; 3: ≥ 31 days) and points allocated to the degree to which pain caused difficulties in performing their usual activities in that same period (0 = *no difficulties at all*; 10 = *impossible to do activities*; 0: < 3; 1: ≥ 3 and < 5; 2: ≥ 5 and < 7; 3: ≥ 7). Based on the scores for pain intensity and disability, the adolescent's pain experience can be classified into 5 pain grades (0 = pain free; I = low disability [< 3], low intensity [< 5]; II = low disability [< 3], high intensity [≥ 5]; III = moderate disability [3 or 4], regardless of pain intensity; IV = high disability [≥ 5] regardless of pain intensity) which was used to describe the sample (1). The GCPS is a valid measure of pain severity in primary care, chronic pain, and general population samples (39–41). The child version has shown good psychometric properties in a general population sample (42).

Psychological flexibility was measured using the Avoidance and Fusion Questionnaire for Youth [AFQ-Y; (34)]. This 17-item scale was originally constructed to identify levels of psychological inflexibility characterized by experiential avoidance (i.e., avoidance of unwanted, negative private experiences such as thoughts and feelings) and cognitive fusion (i.e., being 'fused' or entangled with the content of one's thoughts or feelings). Items were

rated on a 5-point rating scale (0 = *not at all true*; 4 = *very true*). In line with previous research (see (43)), items were reverse-scored so that higher scores reflect a higher level of psychological flexibility. The AFQ-Y has been shown to be a valid measure of psychological (in)flexibility in a sample of children and adolescents from the general population (34,35,43).

Diary measures

Participants were asked to report on the period “since the previous diary entry” and to rate each item on the following five-point scale: 0 (*not at all true*), 1 (*a little true*), 2 (*somewhat true*), 3 (*mostly true*), and 4 (*totally true*) (unless stated otherwise). Diary items were validated using the Discriminant Content Validity (DCV) method of Johnston et al. (44). Prior to diary development, five psychologists with expertise in the field of pediatric pain research rated the extent to which each item measured the predetermined constructs. None of the items measuring the constructs required reformulation. Total diary scale scores were calculated by taking the average of the single item responses (i.e., if the scale consisted of two or three items), but only if at least 75% of the items were completed. If less than 75% of the items were completed, the total scale score was not calculated and considered as missing.

Positive and negative affect were assessed using items from the child version of the Positive and Negative Affect Schedule (PANAS-C) (45). Participants were asked to rate the degree to which they experienced a given affective state in the period since the last diary entry. Five positive mood adjectives (*joyful, cheerful, happy, lively, proud*) provided a measure of positive affect (PA), and four negative mood adjectives (*miserable, blue, afraid, scared*) provided a measure of negative affect (NA). Higher scores indicate higher levels of affect. The PANAS-C has shown good psychometric properties when PA and NA were measured in clinic-referred and school-based samples of youth (45,46).

The overall pain intensity level in the hours preceding the moment of diary completion was measured using a single item (i.e., “What was your overall level of pain?”). Responses ranged from 0 (*no pain*) to 10 (*worst possible pain*). An 11-point scale is a valid self-report tool to measure pain intensity in adolescents with chronic pain (47).

In each diary assessment, participants completed three items assessing their level of *pain catastrophizing* in the hours preceding the assessment: “I thought something serious might happen to me because of the pain”, “I kept thinking about how much pain I was experiencing” and “I felt I couldn’t go on much longer because of the pain.”. These items are based on the 3-item state version (i.e., situation-specific) of the Pain Catastrophizing Scale for Children [PCS-C; (48,49)]. The state PCS-C scale is a reliable and valid measure in children and adolescents aged between 8 and 18 years from the general population (48).

Participants were asked to report on their *pain-related fear* in the period since the last diary entry. Three items from the ‘fear of pain’ subscale of the Fear of Pain Questionnaire for Children [FOPQ-C; (50)] were adjusted to measure momentary pain-related fear (“My pain has caused my heart to beat fast or race”, “Feelings of pain were scary for me” and “I worried about my pain”). The items were selected to reflect physical, emotional, and cognitive aspects of experiencing pain-related fear and were evaluated positively by experts during the content validation procedure. The ‘fear of pain subscale’ of the FOPQ-C has proven valid and reliable in a sample of youth with chronic pain (50).

Pain-related activity-avoidance in the hours preceding the diary assessment was measured using three items derived from the ‘avoidance of activities’ subscale of the FOPQ-C (50) and

adjusted for use in the diary: “I skipped my planned activities because I expected them to trigger or increase my pain.”, “I stopped what I was doing because my pain started to get worse”, “I spent my time resting instead of doing my activities, because of my pain”. These items were selected to reflect different types of avoidance strategies in agreement with the author of the original FOPQ-C and were evaluated positively by the experts during the content validation procedure. Good internal consistency and reliability have been found for the ‘avoidance of activities’ subscale in pediatric chronic pain samples (50).

In each diary assessment, participants reported on their *activity-engagement in the presence of pain*. Following items were used: “I have put effort into completing activities that I find important or fun, while I was in pain”, and “I persisted in carrying out my planned activities while I was in pain”. These items were developed based on items of the ‘activity-engagement’ subscale of the Chronic Pain Acceptance Questionnaire for Adolescents (51). The CPAQ-A has proven to be a valid and reliable measure of pain acceptance (i.e., pain willingness and activity-engagement) in youth with chronic pain (17,51). The items of the activity-engagement scale were only presented to those who rated their pain intensity level in the same period to be at least one or higher as we aimed at measuring the extent to which adolescents engage in activities in the presence of pain.

Data analytic strategy

To answer our research questions network analyses were performed by means of the *lme4* package (52) in R (53). A multilevel approach to vector autoregressive (VAR) modelling (31) was used. Multilevel models can account for the hierarchical data structure (i.e., multiple observations nested within individuals) without violating the assumption of independence of observations and assume that observations are missing at random (54). In a VAR model

variable Y (i.e., the dependent variable) at moment t (in this study: the evening) is regressed on lagged versions of that same variable Y and all other independent variables in the model at moment $t - 1$ (in this study: the afternoon). Two network models of six variables were inferred – one for activity-avoidance and another for activity-engagement.

For our first research objective, i.e. to examine if pain-related activity-avoidance behavior in the evening was predicted by any other variable included in the network (in the afternoon), lagged versions of the level-1 predictors (i.e., pain intensity, pain-related fear, pain catastrophizing, positive and negative affect) were created. In a next step, activity-avoidance assessed in the evening was regressed on activity-avoidance assessed in the afternoon simultaneously with all other predicting variables in the afternoon. Next, similar multilevel VAR models were fitted with every independent variable now considered as an outcome. The same procedure was followed to explore if activity-engagement in the evening was predicted by any other variable in the afternoon. Activity-avoidance and activity-engagement were therefore never incorporated into the same model. Age and gender (level-2 predictors) were included as possible confounders in all models. Normality of the residuals was checked and all variables were standardized (i.e. Z-scores) prior to the analyses. In all models random intercepts were assumed, all slopes were fixed because preliminary analyses of the variances of the effects showed no evidence against the assumption of homogeneous effects. Estimating the fixed effects resulted in a weighted network structure which was visualized by means of the *qgraph* package in R (55). A template model with guidelines on how to interpret the resulting network model is presented in Figure 1.

-Insert Figure 1 about here -

For our second research objective, four additional models were fitted to test the predictive and/or moderating role of psychological flexibility for activity-avoidance and activity-engagement. All models included the same predicting variables as outlined above with psychological flexibility as an additional level-2 predictor. To test if psychological flexibility moderated the strength of within-day associations between pain intensity, pain-related fear, pain catastrophizing, and pain-related behavior, cross-level interaction terms between each of the three predictors (level 1) and psychological flexibility (level 2) were created and added as predictors in these models. Finally, we performed post-hoc calculations mimicking the observed data structure to assess the power to detect small to moderate main and interaction effects of psychological flexibility (see Supplementary File 1).

Results

Sample characteristics

The final sample consisted of 65 participants ($M_{age} = 14.41$ years, $SD = 1.95$; 54 girls [83.1%]). The majority of participants self-identified as white (64.6%), 3.1% as black or African-American, and 3.1% indicated that they were multiracial. Two participants explicitly chose not to answer the question asking about their race, while 16 (24.6%) did not provide it. Thirty-six participants (55.4%) reported musculoskeletal pain (i.e., pain in the back, neck, shoulders, arms, hands, hip, ankles, or feet) as their most dominant pain, 14 reported abdominal pain (21.5%), whereas eight reported headaches as their primary pain (12.3%). Seven participants (10.7%) reported other pains (e.g., pelvic, chest). The mean pain duration at the start of the study was 27.05 months (*range*: 3 – 96 months; $SD = 22.65$). The average pain intensity level during the past six months was moderate ($M = 6.40$; $SD = 1.76$).

Furthermore, the distribution of pain grades [based on the Graded Chronic Pain Scale (GCPS;

see ‘Method’)] in this sample was as follows: 11 % ($n = 7$) of participants in pain grade I; 11% ($n = 7$) in pain grade II; 19 % ($n = 12$) in pain grade III; and 56 % ($n = 36$) in grade IV (data of 3 participants was missing).

Descriptive statistics

Means, ranges, standard deviations, and Pearson correlation coefficients for age, gender, psychological flexibility at baseline, and aggregated diary scores are reported in Table 1. Results showed that participants, on average, reported moderate pain intensity levels during the two-week diary period ($M = 5.10$, $SD = 2.27$).

With regard to the diary items, acceptable ($> .50$) to excellent ($>.80$) scale reliabilities are observed. Reliability checks of the diary items were based on a multilevel confirmatory factor analysis which makes it possible to inspect level-specific reliabilities (56) (see Supplementary File 2). Of 1820 potential diary observations (i.e., 65 individuals $\times$ 14 days $\times$ 2 assessments/day), 1195 were completed (65.7%). Incomplete diary entries (i.e., not all diary items at a single assessment time were completed) were also included in further analyses: 97.3% of the diary assessments were complete, 2.7 % were missing data on at least one but not all diary items. On average 94% (*range*: 16-100%, $SD = 17\%$) of pain intensity scores during the two-week period were rated at 1 or higher. Pain intensity levels of 5 or higher were on average observed in 64% (*range*: 0-100%, $SD = 36\%$) of the pain ratings during the two-week period. Within-individual differences accounted for 55% of the variance in activity-avoidance and 42% of the variance in activity-engagement assessed in the evening.

- Insert Table 1 about here –

Activity-avoidance network

Figure 2 shows the weighted network of afternoon-evening associations with activity-avoidance as the central outcome. In line with our hypothesis, pain-related fear in the afternoon was a significant predictor of higher levels of activity-avoidance later that evening ($b = 0.11$, $t(222.52) = 2.24$, $p < 0.05$). Unexpectedly, pain intensity, pain catastrophizing, negative or positive affect (assessed in the afternoon) did not predict activity-avoidance in the evening above and beyond activity-avoidance and pain-related fear in the afternoon. Further exploration of the network model showed a positive ‘self-loop’ or ‘autoregressive effect’ for each variable (i.e., the level of each variable assessed in the afternoon was predictive of an increased level of that same variable in the evening). The significant self-loop for activity-avoidance in the evening indicates that it was predicted by activity-avoidance earlier that afternoon ($b = 0.61$, $t(418.73) = 18.13$, $p < 0.001$). With regard to consequences of activity-avoidance, results were not in line with our expectations. Activity-avoidance in the afternoon was not related to subsequent pain intensity, pain catastrophizing, pain-related fear, or affect in the evening.

In addition to exploring potential antecedents and consequences of activity-avoidance, we also examined the relationship between these variables themselves. Here we found that adolescents who reported higher pain-related fear in the afternoon also reported higher pain intensity ($b = 0.13$, $t(347.23) = 2.51$, $p < 0.05$) and higher catastrophizing about pain ($b = 0.19$, $t(381.87) = 4.40$, $p < 0.001$) in the evening. A higher degree of pain-related fear in the evening was in turn predicted by higher levels of negative affect in the afternoon ($b = 0.09$, $t(162.68) = 2.88$, $p < 0.01$). The model suggests that pain-related fear was an important variable, predicting most other variables in the model. We also observed a bidirectional positive relation between negative affect and pain catastrophizing such that negative affect in

the afternoon was predictive of pain catastrophizing in the evening ($b = 0.08$, $t(356.07) = 2.14$, $p < 0.05$) while pain catastrophizing in the afternoon was predictive of negative affect in the evening ($b = 0.09$, $t(136.53) = 2.06$, $p < 0.05$). Finally, positive affect in the evening was not predicted by any other variable in the model except by the level of positive affect in the afternoon ($b = 0.70$, $t(144.51) = 20.01$, $p < 0.001$).

– Insert Figure 2 about here –

Activity-engagement network

The weighted network of associations with activity-engagement as the outcome of interest is represented in Figure 3. Similar self-loops or auto-regressive effects for all variables were observed as in the activity-avoidance network (Figure 2). Note that we had no *a priori* expectations about the potential antecedents and consequences of activity-engagement. Exploration of the associations in the model showed that activity-engagement in the evening was only predicted by higher levels of positive affect ($b = 0.11$, $t(172.70) = 2.50$, $p < 0.05$) and activity-engagement ($b = 0.57$, $t(313) = 15.84$, $p < 0.001$) in the afternoon. Pain intensity, pain catastrophizing, pain-related fear, and negative affect in the afternoon did not predict engagement in activities in the evening. With regard to the possible consequences of activity-engagement, no significant associations emerged between activity-engagement in the afternoon and pain intensity, pain-related fear, pain catastrophizing, or affect in the evening.

Further exploration of the relationships between the other variables within the activity-engagement network (Figure 3) yielded two negative associations compared to the activity-avoidance model (Figure 2). Higher pain intensity levels in the afternoon were predictive of lower levels of positive affect in the evening ($b = -0.07$, $t(149.80) = -2.01$, $p < 0.05$). Pain

catastrophizing in the afternoon was negatively associated with pain intensity in the evening ($b = -0.10$, $t(390.39) = -2.06$, $p < 0.05$). In contrast to what was found in the activity-avoidance network, we did not find a positive association between pain catastrophizing in the evening and negative affect in the afternoon. Finally, a positive association between pain-related fear and pain intensity, and between negative affect and pain-related fear emerged.

- Insert Figure 3 about here –

The role of psychological flexibility

To examine if psychological flexibility was predictive of daily pain-related behavior, we first constructed a model to test the direct effect of psychological flexibility on daily activity-avoidance, while controlling for the impact of the activity-avoidance, age, gender, and all other level-1 predictors in the afternoon (see left side of Table 2). Results indicated that baseline psychological flexibility predicted lower levels of daily activity-avoidance as assessed in the evening ($b = -0.12$, $t(53.62) = -2.39$, $p < 0.05$). Second, we constructed a similar model to test the direct effect of psychological flexibility on daily activity-engagement, while controlling for the impact of activity-engagement, age, gender, and all other level-1 predictors in the afternoon (see right side of Table 2). Results revealed that psychological flexibility did not predict levels of daily activity-engagement as assessed in the evening ($b = 0.05$, $t(47.70) = 0.79$, $p = 0.43$).

Next, we examined if psychological flexibility was a moderator of the within-day associations between pain intensity, pain-related fear, pain catastrophizing, and activity-avoidance. We found that psychological flexibility did not moderate within-day associations between pain intensity ($b = -0.05$, $t(111.66) = -1.24$, $p = 0.22$), pain-related fear ($b = -0.05$, $t(292.01) =$

0.90, $p = 0.37$), and pain catastrophizing ($b = -0.09$, $t(365.30) = -1.94$, $p = 0.053$) with activity-avoidance. In short, the strength of these associations did not depend on adolescents' level of psychological flexibility.

Finally, we examined if psychological flexibility was a moderator of the within-day associations between pain intensity, pain-related fear, pain catastrophizing, and activity-engagement. Results indicated that psychological flexibility moderated the association between pain intensity in the afternoon and activity-engagement in the evening ($b = 0.13$, $t(114.72) = 2.85$, $p < 0.01$). Figure 4 shows a significant negative association between pain intensity and subsequent activity-engagement for adolescents who showed the lowest levels of psychological flexibility at baseline. However, this association was no longer significant for higher levels of psychological flexibility. Finally, levels of psychological flexibility did not moderate the association between pain-related fear in the afternoon and activity-engagement in the evening ($b = -0.10$, $t(269.99) = -1.81$, $p = 0.07$), nor between pain catastrophizing and activity-engagement ($b = 0.02$, $t(353.75) = 0.46$, $p = 0.64$).

– Insert Table 2 about here –

– Insert Figure 4 about here –

Discussion

The current study sought to identify variables that influence daily activity-avoidance and activity-engagement in the presence of pain in adolescents with chronic pain. We tested specific hypotheses about the antecedents and consequences of activity-avoidance, and carried

out an exploratory analysis of the potential antecedents and consequences of activity-engagement. Two network models were created to examine these relationships: one focused on whether factors such as pain intensity, pain-related fear, pain catastrophizing, positive and negative affect, were predictive of activity-avoidance and another on whether those same factors were predictive of activity-engagement. Our secondary objective was to determine if psychological flexibility represents a resilience-enhancing factor which predicts lower daily activity-avoidance and higher daily activity-engagement, and buffers against the adverse impact of pain-related experiences on such behaviors. To achieve these objectives, we conducted a daily diary study in combination with a network analytic approach.

With regard to our first objective we found the following. On the one hand, the avoidance network showed that pain-related fear in the afternoon predicts avoidance of pain-related activities in the evening. Activity-avoidance was also more likely to occur in adolescents who avoided activities earlier that day. Pain intensity levels were, however, not predictive of changes activity-avoidance. These findings are generally in line with the Fear-Avoidance Model and prior research (50,57) and suggest that pain-related fear is a better predictor of pain-related functioning than pain itself. Unexpectedly, however, activity-avoidance in the afternoon was not related to subsequent levels of pain intensity, pain-related fear, pain catastrophizing, or affect later that evening.

We also found that both pain-related fear and pain catastrophizing in the evening were predicted by higher levels of negative affect earlier that same day. These findings are also consistent with the Fear-Avoidance Model's position on negative affect as a factor that increases the chance of pain-related fear and catastrophic interpretations (58).

Further, whereas prior research mainly found concurrent links between fear and pain in children and adolescents with chronic pain [e.g., (50,59)], this is the first study to demonstrate the temporal predictive value of pain-related fear in the afternoon for higher pain intensity levels in the evening. We also found that pain-related fear in the afternoon predicts higher levels of pain catastrophizing later that day, but not the reverse. Such findings are inconsistent with the Fear-Avoidance Model's assumption that increased levels of pain catastrophizing necessarily predicts higher levels of fear. This could suggest that the relation between catastrophic thoughts and fear is not unidirectional, but that fearful feelings can elicit catastrophizing as well. Future research could examine if such a bidirectional relationship between fear and catastrophizing exists. An alternative explanation might be that momentary states of catastrophizing and pain-related fear may have different effects compared to when they are considered or measured as a fixed trait (i.e., by means of a questionnaire).

On the other hand, the engagement network showed that activity-engagement was positively predicted by earlier activity-engagement and positive affect. Such a finding is consistent with the Broaden and Build Theory of Positive Emotions (60) which argues that positive affect might broaden one's range of behavioral options in a (stressful) situation (e.g., dealing with chronic pain). This may explain why adolescents who experienced more positive feelings in the afternoon engaged more in activities despite the pain later that day. Activity-engagement in the afternoon was not related to subsequent levels of pain intensity, pain-related fear, pain-catastrophizing, or affect in the evening.

In the engagement network the same positive relations between pain-related fear in the afternoon and subsequent pain intensity and pain catastrophizing, and between negative affect in the afternoon and pain-related fear and pain catastrophizing in the evening, were observed.

However, the engagement network also displayed some unique relationships not observed in the context of avoidance. For instance, we observed a negative relation between pain intensity in the afternoon and positive affect later that day, which could suggest that pain leads to a reduction in positive rather than an increase in negative feelings. We also found a negative relation between pain catastrophizing in the afternoon and levels of pain in the evening, which seems counterintuitive. One post-hoc explanation might be that high levels of catastrophizing in the afternoon were followed by intermediate coping strategies which led to reduced levels of pain in the evening. For instance, these adolescents might have tried to shift their attention away from the negative thoughts about the pain (and potentially also the pain). Potentially, these differences could also be explained by the fact that catastrophizing was measured as a momentary state as opposed to previous work which mainly focused on catastrophizing as a trait. However, given that this relation was only found in the engagement network, future studies should replicate this and search for potential intervening variables to illuminate this finding before making firm conclusions.

With regard to our second objective, to investigate if psychological flexibility represents a resilience-enhancing factor, the following findings emerged. We found that psychological flexibility indeed predicted lower levels of daily pain-related activity-avoidance. This is consistent with our theory-based expectations (15,24,25) and previous research [e.g., (17,61)]. Adolescents who are generally more psychologically flexible are expected to be less avoidant of unwanted, negative experiences or events (such as chronic pain). However, based on theory we would also expect that these adolescents engage more in pain-related activities as compared to their less psychologically flexible peers. Yet our findings showed no significant associations between adolescent psychological flexibility and their daily activity-engagement. We also expected it to buffer against expected adverse associations between pain intensity,

pain-related fear, catastrophizing, and subsequent activity-avoidance or -engagement. We found that the negative relation between pain intensity and engagement behavior indeed was not present for those adolescents who reported higher levels of psychological flexibility. This suggests that activity-engagement in this group is less influenced by their pain level relative to their peers who scored lower in psychological flexibility. Taking a step back, this might signify that their decision to engage in activities is driven by other factors than pain (e.g., values), which is one of the core predictions within psychological flexibility theories in the context of pain (15). That said, psychological flexibility did not moderate the relationship between pain-related fear or pain catastrophizing and activity-engagement, nor between pain intensity, pain-related fear, pain catastrophizing, and activity-avoidance. Correlational analyses showed that psychological flexibility was associated with lower daily levels of pain catastrophizing and pain-related fear, and that lower daily levels of pain catastrophizing and pain-related fear were in turn correlated with lower daily activity-avoidance. It could be that the adaptive effects of psychological flexibility in relation to these cognitive/emotional factors are better explained by a mediating rather than a moderating pathway. Future research could test if psychological flexibility leads to reduced levels of activity-avoidance via lower levels of pain catastrophizing, or pain-related fear, by testing specific mediating pathways. Although no correlational patterns were found to suggest this, it might be worthwhile to explore if similar processes mediate the potential association between psychological flexibility and activity-engagement.

In short, the hypothesized resilience-enhancing role of psychological flexibility in the daily life of adolescents with chronic pain was only partially supported by our findings: it acted as a predictor of lower levels of daily activity-avoidance, and as a buffer for the negative impact of pain intensity on activity-engagement. However, it was not predictive of daily activity-engagement, nor did it moderate associations between pain catastrophizing or pain-related

fear and activity-avoidance or -engagement. Future research should examine the underlying mediating or moderating mechanisms of the adaptive effects of psychological flexibility on adolescents' daily pain-related behavior and long-term outcomes.

Limitations and future directions

The current work has a number of limitations and opens up new directions for future research. One immediate issue is that only afternoon and evening reports, and no morning reports on the pain-related factors were used in this study. Previous work suggests that morning pain intensity may be the best predictor of pain-related functioning throughout the day (62). Future diary studies should try to incorporate morning measures as well. Second, one should be aware of the fact that self-report measures are often subject to social desirability. Although all participants were informed that their answers would be anonymized and would never affect their further treatment, there is still a chance that they reported what they wanted their health care providers to hear rather than what they actually felt or thought. This risk was especially high because they were recruited in a hospital setting at initial clinical evaluation. Future research could test this possibility by replicating our work across different samples and settings, or by employing manipulations or measures that reduce the impact of social desirability on responding. For instance, observational (e.g., parent proxy reports) or activity monitoring assessments could be used to obtain a more objective perspective on the adolescent's daily activity-avoidance or -engagement. A third issue is that a variety of chronic pain conditions were included in this study. Future work could examine if distinct associations with activity-avoidance or activity-engagement emerge as a function of chronic pain type. It may be that adolescents with musculoskeletal pain avoid different activities (e.g., physically demanding activities) than those suffering from persistent headaches (e.g., loud and noisy activities).

In future research, researchers could draw from other conceptual models to further identify potential antecedents and consequences of activity-avoidance and -engagement beyond those central to the Fear-Avoidance Model. For instance, a goal pursuit perspective draws attention to goal achievement or frustration [see (63)] while the Psychological Flexibility Model in chronic pain (15) would argue that pain acceptance needs to be considered (17,64). Future studies could also examine the behavior of peers, parents, or other caregivers as potential antecedents or consequences for the adolescent's pain-related activity-avoidance and -engagement given the essential role of interpersonal influences in pediatric pain (2,3,65). Although this is one of the core predictions of the Fear-Avoidance Model, the present study did not focus on the long-term (mal)adaptive outcomes of pain-related activity-avoidance and -engagement. Given that previous work in adults showed that not only persistent avoidance, but also rigid engagement in all movements or activities (i.e., without evaluation of the consequences of doing so) can lead to maladaptive outcomes (66), future research could examine which daily behavioral dynamics are predictive of either long-term disability or adaptation to pain. Even though the Fear-Avoidance Model portrays avoidance of, or engagement in activities as two opposite pathways in responding to pain, it would be interesting to examine how they co-vary. Finally, future diary studies could explore how psychological flexibility operates in daily life rather than including it as a baseline variable. The variability or flexibility in activity-avoidance or -engagement across days, factors relating to flexibility versus persistence in different behavioral patterns, and how different patterns relate to long-term outcomes could be examined.

Clinical implications

If replicated, our findings would have implications for clinical practice. First, they would

reaffirm the importance of pain-related fear in driving avoidance of daily activities, a central assumption in most cognitive behavioral therapies for youth with chronic pain (see (67)).

They also suggest that strategies aimed at increasing activity-engagement in the presence of pain may need to focus on enhancing positive emotions [see (68,69)]. The network analytic techniques used here could have potential as a therapeutic tool to help explore, discuss, and target factors that are associated with an adolescent's pain-related behavior (31). Finally, our examination of psychological flexibility and daily pain-related behavior could inform future research on the processes central to Acceptance and Commitment Therapy [ACT; (25)], a treatment strategy focused on enhancing psychological flexibility. Whereas available evidence speaks to the effectiveness of ACT in increasing adaptive functioning in adolescents with chronic pain (27,28,70), more in-depth research is needed to identify the exact processes that underpin changes in psychological flexibility.

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Figures and Tables

Table 1. Means, Standard Deviations, Range and Pearson Correlations Coefficients for all Baseline and Diary Variables

[illegible]

Table 2. *Models Fitted to Test the Predictive Effect of Psychological Flexibility on Daily Activity-avoidance and Activity-engagement*

Outcome: Avoidance (<i>t</i>)				Outcome: Engagement (<i>t</i>)		
Predictors: Fixed Effects	B	SE (B)	95% CI	B	SE (B)	95% CI
Level 1 (within-individual)						
Intercept	-0.03	0.29	[-0.56 – 0.51]	0.03	0.37	[-0.65 – 0.71]
Pain Intensity (<i>t</i> – 1)	0.03	0.04	[-0.04 – 0.10]	-0.06	0.05	[-0.15 – 0.03]
Pain-related Fear (<i>t</i> – 1)	0.09	0.05	[-0.01 – 0.19]	0.03	0.05	[-0.09 – 0.13]
Pain Catastrophizing (<i>t</i> – 1)	-0.02	0.05	[-0.13 – 0.07]	-0.04	0.05	[-0.13 – 0.09]
Positive Affect (<i>t</i> – 1)	0.03	0.04	[-0.04 – 0.11]	0.11*	0.05	[0.02 – 0.20]
Negative Affect (<i>t</i> – 1)	0.01	0.05	[-0.07 – 0.11]	-0.02	0.05	[-0.11 – 0.08]
Engagement (<i>t</i> – 1)	n/a	n/a	n/a	0.57***	0.04	[0.50 – 0.68]
Avoidance (<i>t</i> – 1)	0.61***	0.03	[0.55 – 0.69]	n/a	n/a	n/a
Level 2 (between-individual)						
Age	-0.01	0.02	[-0.04 – 0.03]	-0.001	0.03	[-0.05 – 0.05]
Gender	0.13	0.11	[-0.07 – 0.33]	-0.003	0.14	[-0.26 – 0.25]
PF	-0.12*	0.05	[-0.21 – -0.02]	0.05	0.06	[-0.06 – 0.16]
Predictors: Random Effects	S²					
Level 1 (intercept)	0.05	0.10				

Level 2 (residual)	0.35	0.32
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B = unstandardized beta coefficients; PF = Psychological Flexibility (as measured by the Avoidance and Fusion Questionnaire for Youth[1]).

* $p < .05$. ** $p \leq .01$. *** $p \leq .001$

Figure 1. Template Network Model

The nodes in the network represent the key variables, the arrows between the nodes display the associations between the variables. The weighted network structure represents autoregressive fixed effects (i.e., self-loops) and cross-regressive effects (i.e., between two different variables). An arrow between node *a* and node *b* represents the relationship between variable *a* in the afternoon and variable *b* in the evening. The strength of an association is reflected by the thickness of the arrow (i.e., a thicker line signals a stronger association), whereas the shape of the arrow indicates the direction of the association (i.e., dashed line represents negative association; solid line represents positive association). Intercepts are not represented in the network structure.

Figure 2. Activity-avoidance Network. A total number of 563 observations were included in this model. Only significant associations ($p < .05$) are displayed. AVOID. BEH. = activity-avoidance behavior; PAIN = pain intensity; FEAR = pain-related fear; PAIN CATA. = pain catastrophizing; POS. AFF. = positive affect; NEG. AFF. = negative affect. Instructions on how to interpret this model are presented in a template model (Figure 1).

Figure 3. Activity-engagement Network. A total number of 502 observations were included in this model. Only significant associations ($p < .05$) are displayed. ENG. BEH. = activity-engagement behavior; PAIN = pain intensity; FEAR = pain-related fear; PAIN CATA. = pain catastrophizing; POS. AFF. = positive affect; NEG. AFF. = negative affect. More instructions on how to interpret these models are presented in a template model (Figure 1).

Figure 4. Interaction plot for the interaction effect of pain intensity (PI) and psychological flexibility (PF) on activity-engagement. Regression lines for the effect of PI are shown for the mean value of PF (= 0; bottom right), 1 standard deviation below the mean (PF = -1; bottom left), and 1 standard deviation above the mean (PF = 1; upper left). Psychological flexibility was a significant moderator of the within-day association between pain intensity and activity-engagement. Specifically, the significant negative association between pain intensity and activity-engagement at low levels (PF = -1) of psychological flexibility ($b = -0.20$, 95% CI from -0.35 to -0.06, $p = .006$) was no longer significant for higher levels

(PF = 0 & PF = 1) of psychological flexibility ($b = -0.07$, 95% CI from -0.17 to 0.03, $p = .16$; and $b = 0.06$, 95% CI from -0.06 to 0.19, $p = .31$).

Supplementary Files

Supplementary File 1

Post-hoc power calculations

Post-hoc power analyses mimicking the observed data were performed. We assumed a first-order autoregressive process for the level-1 predictor and outcome with mean 0, variance 1, and autocorrelation 0.60; and a standardized level-2 predictor with mean 0 and variance 1. In a sample of 65 patients with 14 afternoon and evening measurements, the study had about 70% power to detect a main effect of size 0.10 (i.e., beta coefficient) of the upper level predictor on the outcome in the VAR model, and about 80% power to detect an effect size of 0.10 for the moderation of the upper-level predictor on the association between the lower-level predictor and the outcome in a VAR model.

Supplementary File 2. *Within- and Between-Person Reliabilities for the Diary Scales in the Afternoon and in the Evening*

	Pain Catastrophizing		Pain-related Fear		Activity- Avoidance		Activity- Engagement		Positive Affect		Negative Affect	
	Afternoon	Evening	Afternoon	Evening	Afternoon	Evening	Afternoon	Evening	Afternoon	Evening	Afternoon	Evening
Within- person α	.64	.65	.62	.59	.82	.80	.67	.74	.82	.86	.59	.68
Between- person α	.90	.89	.74	.78	.95	.74	.92	.93	.97	.98	.88	.88

Note. Reliabilities were estimated by a multilevel confirmatory factor analysis framework by Gheldof et al., (2014)