Understanding why cognitive—behavioral therapy is an effective treatment for adolescents with juvenile fibromyalgia

Recent studies have demonstrated that cognitive-behavioral therapy (CBT) is an effective treatment for children and adolescents with juvenile fibromyalgia. Unfortunately, the specific psychological changes that occur during treatment that explain why CBT works are not well understood. Historically, the increased use of specific coping strategies learned during CBT was thought to be the primary reason for why CBT was effective. However, evidence to support the notion that increases in adaptive coping directly lead to patient improvement is minimal. Instead, a growing number of studies in adults suggest that CBT results in more global changes of psychological perceptions of the pain itself (cognitive appraisals) and one's ability to manage the pain. This report discusses the role of coping and aspects of cognitive appraisal as potential psychological changes that explain CBT-related improvements for youths with juvenile fibromyalgia.

KEYWORDS: adaptive coping = catastrophic thinking = chronic pain = cognitive appraisal = cognitive-behavioral therapy = coping self-efficacy = juvenile fibromyalgia

Cognitive-behavioral therapy works for patients with fibromyalgia

Cognitive-behavioral therapy (CBT) is known to be an effective treatment for adults [1,2] and children with fibromyalgia [3]. Among youths with juvenile fibromyalgia (JFM), treatment with CBT combined with usual medical care results in significant and clinically relevant improvement in a number of domains such as daily functioning - that is, participation in everyday activities in home, school and social settings - as well as improved mood, and to a lesser extent, reduced pain levels by the end of treatment [3-5]. Effects on functioning and mood in one study of CBT for adolescents were maintained over 6 months [3]. It should be noted that JFM is a complex pain condition involving several associated symptoms, including fatigue and sleep disturbance, in addition to pain; however, behavioral treatment is most often focused on teaching patients pain management strategies and coping tools to help them function better in their daily lives. This is also in keeping with guidelines for pediatric pain trials that identify physical functioning, pain intensity and mood as key outcome measures to aid comparison across pain conditions [6-10]. Although the evidence for the effectiveness of CBT in the treatment of JFM in improving daily functioning and depressed mood is strong, the specific psychological changes that occur during CBT that make it an effective treatment are not well understood.

Based on the theory behind CBT, the negative impact of chronic pain in a person's life is maintained, in large part, by a combination of maladaptive thoughts, feelings and behaviors. Therefore, CBT treatment focuses on training patients in the use of specific cognitive and behavioral strategies to improve pain coping and reduce pain-related disability. CBT has been found to improve several psychological processes, such as increasing adaptive coping, reducing catastrophic thinking about pain and increasing coping self-efficacy (i.e., one's perceived control over pain) in both adults and youths with fibromyalgia [1,2,11,12]. At present, very few studies have specifically addressed the psychological changes that are associated with improvements in children and adolescents who received CBT for pain management [13]. More attention to how psychological treatments work for pediatric patients with chronic pain is needed owing to the unique developmental issues and specific needs that are relevant to this age group. This report will draw upon what is known from both adult and pediatric CBT treatment studies to identify and discuss potential psychological changes that lead to improvement in functioning, mood and pain for youths with JFM.

Overall goals & treatment components of CBT

The primary goal of CBT for children and adolescents with JFM is to reduce pain-related

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disability in order to help patients get back into their daily activities, such as school, peer and family activities, despite pain. Reduction of emotional distress and pain severity are also important secondary goals. These goals are accomplished by training children and adolescents in the use of active and adaptive coping skills to better manage their pain. Several components of CBT help patients achieve these goals, including: education about how the body processes pain and psychological factors that impact pain perception; learning to identify and change negative thoughts and helpless feelings associated with pain (e.g., by using positive and calming self-statements, distraction and problem solving) and behavioral skills (e.g., relaxation and activity pacing); training parents in behavior management strategies (e.g., reducing attention for negative pain behaviors, supporting the child's use of coping skills in daily life and encouraging independent coping); and learning self-monitoring skills to identify internal (e.g., avoiding activities due to fear of pain) and environmental (e.g., overdoing activity and tests at school) triggers for pain episodes [14,15]. Finally, they learn to monitor their own progress to maintain treatment gains over time. Collectively, these treatment components can replace unhelpful and maladaptive coping strategies with more helpful and adaptive ones, thus modifying the thoughts and behaviors that contribute to the maintenance of chronic pain and disability.

Does CBT work because patients learn new adaptive coping skills during treatment?

Improving patients' coping with chronic pain is one psychological process that historically has received a lot of attention from both clinicians and researchers as the key technique that helps change pain behaviors and contributes to beneficial improvements [16-18]. However, research in adults has shown that the increased use of specific coping strategies taught during treatment, such as relaxation or activity pacing, actually had surprisingly little direct impact on patient improvement in CBT treatment studies [18-22]. This suggests that perhaps improving adaptive coping skills is not the primary explanation for how CBT works - at least in adults; rather, there might be other underlying psychological changes related to CBT that play a stronger role in explaining patients' improvement in functioning.

Is improvement from CBT related to broader changes in psychological coping and beliefs about pain? Clues from a conceptual model of pain, coping & appraisal

To better understand other potential psychological changes that explain why CBT is an effective treatment for children and adolescents, we drew upon past models of stress and coping [23] and applications of this model to chronic pain [24] to create a conceptual model of coping for youths with chronic pain (FIGURE 1). The classic stress and coping model proposes that two critical psychological processes may explain the relationship between psychological stress (such as chronic pain) and an individual's wellbeing and functioning [23]: the cognitive appraisal, which is an evaluative process that determines the perceived severity of the stressor/threat (chronic pain) and the person's available resources and abilities to deal with it: and coping, which are cognitive and behavioral strategies that are purposefully used to manage the stress (chronic pain). The appraisal process can result in negative or benign-positive appraisals of pain. For example, catastrophic thinking about pain and negative beliefs about pain (e.g., belief that one is disabled by pain and pain signals damage in the body) are part of the appraisal process [24] and influence the evaluation of one's ability to cope [25]. An individual's coping self-efficacy or perceived control of pain (part of the secondary appraisal process) can guide the use of specific coping strategies. Therefore, the cognitive appraisal of pain may play an influential role in the selection of adaptive or maladaptive coping strategies, which further impacts physical and emotional functioning.

Importance of changing cognitive appraisals as a result of CBT

Treatment studies have expanded their focus beyond increases in the use of specific coping skills to evaluate other psychological changes that occur during treatment. Specifically, research has demonstrated that individuals' cognitive appraisals of pain, such as lower levels of catastrophic thinking and beliefs about pain (primary appraisals), and enhanced coping self-efficacy (secondary appraisal), were related to favorable improvements in functioning in studies of adults with fibromyalgia [1,2,20] and other chronic pain conditions [17,26]. An examination of these cognitive appraisals in adult CBT treatment studies has found that



Data taken from [23,24].

a reduction in catastrophic thinking [26] and improvement in pain coping self-efficacy [19] were the strongest psychological changes that occurred during treatment that explained why CBT was effective. Thus, despite the longstanding emphasis on teaching specific strategies to cope with chronic pain, studies in adult patients suggest that targeting the underlying cognitive appraisal process by changing patient beliefs about pain and improving self-efficacy may be the more important targets of CBT treatment and teaching coping skills is just a means whereby these changes are facilitated. Interestingly, recent brain imaging studies have begun to elucidate the neurobiological correlates of cognitive appraisals and different aspects of the pain experience. Research in adults with

fibromyalgia has documented abnormalities in pain processing within the CNS, which suggests that central sensitization may contribute to the biological basis of fibromyalgia [27,28]. At least one neuroimaging study using functional MRI in adults with fibromyalgia suggests that catastrophic thinking was associated with increased brain activity that sharpens attention to pain and anticipation of pain, and enhances emotional responses to pain [29]. Therefore, it is conceivable that psychological treatments that effectively reduce catastrophic thinking can also alter brain activation in a way that attenuates the 'suffering' component of chronic, unremitting pain. More studies are needed in this area, especially in pediatric patients with chronic pain, to better understand how CBT may affect the neurobiology of pain in

children when they are equipped with stronger psychological coping skills to modify cognitive appraisals of pain.

How does CBT work in children & adolescents with JFM?

It is important to recognize that the psychological changes that lead to patient improvement in functioning may work differently in children and adolescents compared with adults. Unlike adults, children and adolescents are still in the process of fully developing their cognitive skills and abilities. In particular, the development of cognitive and behavioral coping strategies increases with age, such that the number of different coping strategies used in response to a painful stressor increases as children mature into late adolescence and young adulthood [30]. For example, the use of behavioral distraction, positive self-statements and relaxation in response to a painful stressor significantly increases between the ages of 8 and 18 years. Despite the developmental changes that occur in coping, engagement in catastrophic thinking in response to pain was found to remain high across all ages [30]. This suggests that throughout adolescence, youths are becoming more mature and self-aware of their own abilities to think about how they cope with pain and recognize their emotional reactions and beliefs related to pain. In addition, adults and caregivers often have a stronger influence on shaping youths' perception of how well they can manage their own pain and related symptoms. Therefore, the unique developmental issues faced by children and adolescents may influence the psychological changes that occur during CBT.

Understanding the changes that occur in coping and cognitive appraisals during CBT requires consideration of these processes within the context of adolescents' skills and abilities. Considering the cognitive developmental changes that occur throughout childhood, it is often easier for children and adolescents to begin changing their behavior and coping in response to guided instruction during CBT before directly modifying their thoughts about pain. Comparable with CBT treatment for depression in youths, targeting changes in behavior is encouraged early in treatment as a means to achieve modifications of maladaptive thoughts and beliefs [31]. Therefore, in children and adolescents with JFM, it is possible that the coping improvements that occur during CBT also help strengthen changes in cognitive appraisals [14,32]. For example, coping strategies such as relaxation skills, pacing and distraction

are concrete skills that children can more easily master first, which may help further reduce their catastrophic thinking about pain, engage in more calming or neutral thoughts about managing pain and have greater perceived control over their pain. Therefore, children and adolescents with JFM may need to focus on improving their pain behaviors and coping early in treatment to help further support successful modification of their cognitive appraisals of pain.

Only one study to date has tested the psychological changes underlying how CBT works for children and adolescents with JFM [12]. Consistent with CBT studies with adults, there was an increase in the use of specific coping strategies taught during CBT, but these coping improvements did not directly explain improvements in functioning and mood for youths with fibromyalgia. However, the role of cognitive appraisals for youths with JFM was surprisingly different from CBT studies with adults. That is, changes in cognitive appraisals, such as a reduction in catastrophic thinking and increased coping self-efficacy, were evident during treatment but did not explain CBT efficacy in youths as it did for adults. This does not necessarily suggest that coping and cognitive appraisals, such as catastrophizing and self-efficacy, are unimportant psychological changes to consider in understanding how CBT works for youths, but rather highlights several areas of interest that are in need of further research.

The relative importance of changing cognitive appraisals versus improving adaptive coping during CBT treatment in pediatric pain conditions remains unclear. Pain beliefs (part of the cognitive appraisal process) are an emerging key explanation of how CBT treatment works for adults [19,22,26], but have not been directly assessed in CBT studies for pediatric chronic pain. However, pain beliefs have been a primary target of change in treatment studies evaluating the effectiveness of Acceptance and Commitment Therapy (ACT), an extension of CBT. A primary goal of ACT involves improving functioning by increasing psychological flexibility, which is one's ability to act effectively with respect to one's personal values even when faced with interfering or negative thoughts, emotions or bodily sensations, such as pain [33,34]. A smallscale (n = 30) randomized study of ACT for the treatment of pediatric chronic pain suggested that changes in pain beliefs and reactions to pain helped to partially explain the effectiveness of ACT on improving depressive symptoms (with preliminary support on reducing disability/pain

impairment) [34]. Therefore, future CBT studies for youths need focused assessments on both coping strategies and cognitive appraisals, especially pain beliefs, to help clarify the role of each of these psychological changes during treatment. In addition, reports by parents and caregivers should be included to help obtain a more accurate perspective of adolescents' actual use of coping strategies and whether skills are practiced regularly, as intended.

Lastly, it is possible that the current methodology of treatment studies has not fully captured the temporal relationship of psychological changes that occur during CBT. That is, the widely spaced assessments (at 3-6-month intervals) typically utilized in treatment studies may overlook treatment-related changes that are ongoing during treatment. Therefore, monitoring treatment outcomes more regularly (weekly or biweekly) throughout treatment may reveal clearer patterns of how psychological changes occur with time during the course of CBT.

Conclusion

In summary, CBT is an effective treatment for children and adolescents with JFM, but the underlying psychological changes that lead to patient improvements from CBT are not well understood. Although training in adaptive pain coping skills has long been considered the

Executive summary

Cognitive-behavioral therapy works for patients with fibromyalgia

- Cognitive-behavioral therapy (CBT) treatment focuses on training patients in the use of specific cognitive and behavioral strategies to improve pain coping and reduce pain-related disability.
- Additional studies are needed to understand how psychological treatments work for pediatric patients with chronic pain.

Overall goals & treatment components of CBT

- The primary goal of CBT is to reduce pain-related disability in order to help patients get back into their daily activities despite pain.
- Components of CBT include education about how the body processes pain, behavioral skills, learning to identify and change negative thoughts and helpless feelings associated with pain, training parents in behavior management strategies and support, and self-monitoring skills.

Does CBT work because patients learn new adaptive coping skills during treatment?

- Studies provide no direct evidence to support that increases in adaptive coping result in improved treatment outcomes.
- Other underlying psychological changes that occur during CBT may play a stronger role in explaining patients' improvement in functioning.

Is improvement from CBT related to broader changes in psychological coping & beliefs about pain? Clues from a conceptual model of pain, coping & appraisal

- A classic stress and coping model was applied to chronic pain to create a conceptual model to better understand coping in youths with chronic pain.
- Cognitive appraisals and coping are two critical psychological processes that might explain the relationship between chronic pain and an individual's wellbeing and functioning.
- Cognitive appraisals of pain may play an influential role in the selection of adaptive or maladaptive coping strategies, which further с. impacts physical and emotional functioning.

Importance of changing cognitive appraisals as a result of CBT

- Research has found that a reduction in catastrophic thinking and improvement in pain coping self-efficacy were the strongest psychological changes that occurred during treatment that explained why CBT was effective in adults.
- Targeting the underlying cognitive appraisal process by changing patient beliefs about pain and improving self-efficacy may be the more important targets of CBT treatment.

How does CBT work in children & adolescents with juvenile fibromyalgia?

- As they are still developing cognitive abilities, youths with juvenile fibromyalgia may need to focus on improving their pain behaviors and coping early in treatment to help further support successful modification of their cognitive appraisals of pain.
- The only pediatric CBT study that evaluated psychological changes during treatment did not find support for adaptive coping or cognitive appraisals (catastrophic thinking or coping self-efficacy) as explanations for why CBT was effective.
- Additional pediatric studies are needed with focused assessments on both coping strategies and cognitive appraisals to help clarify the role of each of these psychological changes during treatment.

Conclusion

- CBT is an effective treatment for youths with juvenile fibromyalgia, but the underlying psychological changes that lead to patient improvements are not well understood.
- Additional research is needed to examine cognitive appraisals of pain and coping as potential psychological changes that explain how CBT is effective for youths.

primary potential explanation through which CBT is effective, there is no direct evidence to support that increases in adaptive coping result in improved treatment outcomes. Rather, research suggests that cognitive appraisals of pain, such as pain beliefs, catastrophic thinking about pain and coping self-efficacy, are important psychological changes that are related to the improvements from CBT for adults with chronic pain. Additional research is warranted to examine cognitive appraisals of pain and coping as potential psychological changes that explain how CBT is effective for children and adolescents.

Future perspective

To date, randomized controlled trials of CBT have established that CBT is effective for chronic pain in adult and pediatric samples. Additional trials are needed to determine whether treatment-related improvements are

References

Papers of special note have been highlighted as: • of interest

- of considerable interest
- Bernardy K, Fuber N, Kollner V, Hauser W. Efficacy of cognitive–behavioral therapies in fibromyalgia syndrome – a systematic review and metaanalysis of randomized controlled trials. *J. Rheumatol.* 37(10), 1991–2005 (2010).
- 2 Glombiewski JA, Sawyer AT, Gutermann J, Koenig K, Rief W, Hofmann SG. Psychological treatments for fibromyalgia: a meta-analysis. *Pain* 151(2), 280–295 (2010).
- 3 Kashikar-Zuck S, Ting TV, Arnold LM *et al.* Cognitive behavioral therapy for the treatment of juvenile fibromyalgia: a multisite, single-blind, randomized, controlled clinical trial. *Arthritis Rheum.* 64(1), 297–305 (2012).
- Randomized controlled trial demonstrating the efficacy of cognitive-behavioral therapy for youths with juvenile fibromyalgia.
- 4 Kashikar-Zuck S, Swain NF, Jones BA, Graham TB. Efficacy of cognitive–behavioral intervention for juvenile primary fibromyalgia syndrome. *J. Rheumatol.* 32(8), 1594–1602 (2005).
- 5 Degotardi PJ, Klass ES, Rosenberg BS, Fox DG, Gallelli KA, Gottlieb BS. Development and evaluation of a cognitive–behavioral intervention for juvenile fibromyalgia. *J. Pediatric Psychol.* 31(7), 714–723 (2006).
- 6 Miró J, Huguet A, Nieto R. Predictive factors of chronic pediatric pain and disability: a Delphi poll. J. Pain 8(10), 774–792 (2007).

- 7 McGrath PJ, Walco GA, Turk DC *et al.* Core outcome domains and measures for pediatric acute and chronic/recurrent pain clinical trials: PedIMMPACT recommendations. *J. Pain* 9(9), 771–783 (2008).
- 8 Dworkin RH, Turk DC, Farrar JT *et al.* Core outcome measures for chronic pain clinical trials: IMMPACT recommendations. *Pain* 113(1–2), 9–19 (2005).
- 9 Eccleston C, Palermo TM, Williams AC, Lewandowski A, Morley S. Psychological therapies for the management of chronic and recurrent pain in children and adolescents. *Cochrane Database Syst. Rev.* (2), CD003968 (2009).
- 10 Palermo TM, Eccleston C, Lewandowski AS, Williams AC, Morley S. Randomized controlled trials of psychological therapies for management of chronic pain in children and adolescents: an updated meta-analytic review. *Pain* 148(3), 387–397 (2010).
- 11 Nielson WR, Jensen MP. Relationship between changes in coping and treatment outcome in patients with Fibromyalgia Syndrome. *Pain* 109(3), 233–241 (2004).
- 12 Kashikar-Zuck S, Sil S, Lynch-Jordan A et al. Changes in pain coping, catastrophizing and coping efficacy after cognitive-behavioral therapy in children and adolescents with juvenile fibromyalgia. J. Pain (2013) (In Press).
- Evaluates the psychological changes that occur during cognitive-behavioral therapy for youths with juvenile fibromyalgia.
- 13 Morley S. Process and change in cognitive behaviour therapy for chronic pain. *Pain* 109(3), 205–206 (2004).

maintained long-term, beyond 6–12 months. Future studies will likely include more systematic and planned tests of theoretically driven psychological processes to increase the therapeutic impact of CBT for chronic pain. By doing so, CBT treatments may lead to greater patient improvements, result in more costeffective and efficient treatment, and provide a better match of treatments to patient needs.

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- 14 Kashikar-Zuck S, Lynch-Jordan A. Psychological interventions for chronic pain. In: *Pain in Children: a Practical Guide for Primary Care*. Walco GA, Goldschneider K (Eds). Humana Press, NJ, USA, 145–152 (2008).
- 15 Palermo TM. Cognitive–Behavioral Therapy for Chronic Pain in Children and Adolescents. Oxford University Press, NY, USA (2012).
- 16 Eccleston C, Morley S, Williams A, Yorke L, Mastroyannopoulou K. Systematic review of randomised controlled trials of psychological therapy for chronic pain in children and adolescents, with a subset meta-analysis of pain relief. *Pain* 99(1–2), 157–165 (2002).
- 17 Jensen MP, Turner JA, Romano JM. Changes in beliefs, catastrophizing, and coping are associated with improvement in multidisciplinary pain treatment. J. Consult. Clin. Psychol. 69(4), 655–662 (2001).
- 18 Morley S, Eccleston C, Williams ADC. Systematic review and meta-analysis of randomized controlled trials of cognitive behaviour therapy for chronic pain in adults, excluding headache. *Pain* 80, 1–13 (1999).
- 19 Turner JA, Holtzman S, Mancl L. Mediators, moderators, and predictors of therapeutic change in cognitive–behavioral therapy for chronic pain. *Pain* 127(3), 276–286 (2007).
- Demonstrates that adults' perceived control over pain helps to explain cognitive–behavioral therapy-related patient improvement.
- 20 Curran C, Williams AC, Potts HW. Cognitive–behavioral therapy for persistent pain: does adherence after treatment affect

outcome? Eur. J. Pain 13(2), 178–188 (2009).

- Demonstrates that the use of coping strategies taught during treatment do not directly explain improvements in treatment outcomes.
- 21 Jensen MP, Turner JA, Romano JM, Karoly P. Coping with chronic pain: a critical review of the literature. *Pain* 47(3), 249–283 (1991).
- 22 McCracken LM, Eccleston C. Coping or acceptance: what to do about chronic pain? *Pain* 105(1–2), 197–204 (2003).
- 23 Lazarus RS, Folkman S. *Stress, Appraisal, and Coping.* Springer Publishing Company, NY, USA (1984).
- Classic work that presents a detailed theory of how coping and cognitive appraisal impacts psychological stress.
- Turner JA, Jensen MP, Romano JM.
 Do beliefs, coping, and catastrophizing independently predict functioning in patients with chronic pain? *Pain* 85(1–2), 115–125 (2000).
- Study examining the relative contributions of patient beliefs, catastrophizing and coping

on physical and psychosocial functioning in adults.

- 25 Lazarus RS. Coping theory and research: past, present, and future. *Psychosomatic Med.* 55, 234–247 (1993).
- 26 Smeets RJ, Vlaeyen JW, Kester AD, Knottnerus JA. Reduction of pain catastrophizing mediates the outcome of both physical and cognitive–behavioral treatment in chronic low back pain. *J. Pain* 7(4), 261–271 (2006).
- Demonstrates that reducing catastrophic thinking about pain helps to explain patient improvement in response to cognitive-behavioral therapy.
- 27 Blumenstiel K, Gerhardt A, Rolke R *et al.* Quantitative sensory testing profiles in chronic back pain are distinct from those in fibromyalgia. *Clin. J. Pain* 27(8), 682–690 (2011).
- 28 Woolf C. Central sensitization: implications for the diagnosis and treatment of pain. *Pain* 152(3), S2–S15 (2011).
- 29 Gracely RH, Geisser ME, Giesecke T *et al.* Pain catastrophizing and neural responses to

pain among persons with fibromyalgia. *Brain* 127, 835–843 (2004).

- 30 Brown JM, O'Keeffe J, Sanders SH, Baker B. Developmental changes in children's cognition to stressful and painful situations. *J. Pediatric Psychol.* 11, 343–357 (1986).
- 31 Reinecke M, Ginsburg G. Cognitive-behavioral treatment of depression during childhood and adolescence. In: *Handbok of Depression in Children and Adolescents*. Abela J, Hankin B (Eds). Guilford Press, NY, USA, 179–206 (2008).
- 32 Vlaeyen JW, Morley S. Cognitive–behavioral treatments for chronic pain: what works for whom? *Clin. J. Pain* 21, 1–8 (2005).
- 33 Wicksell RK, Olsson GL, Hayes SC. Psychological flexibility as a mediator of improvement in acceptance and commitment therapy for patients with chronic pain following whiplash. *Eur. J. Pain* 14(10), 1059.e1–1059.e11 (2010).
- 34 Wicksell RK, Olsson GL, Hayes SC. Mediators of change in acceptance and commitment therapy for pediatric chronic pain. *Pain* 152, 2792–2801 (2011).