Maintaining Lasting Improvements: One-Year Follow-Up of Children With Severe Chronic Pain Undergoing Multimodal Inpatient Treatment

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Objective To investigate the long-term effectiveness of a 3-week multimodal inpatient program for children and adolescents with chronic pain. Methods 167 adolescents were evaluated at pretreatment baseline, 3-, and 12-month follow-up. Long-term effectiveness was investigated for pain-related variables (pain-related disability, school absence, pain intensity) and emotional distress. Results We found statistically and clinically significant changes in all variables. After 1 year, the majority (56%) showed overall improvement as indexed by decreased pain-related disability or school absence. 22% had an unsuccessful treatment outcome. Those showing only short-term improvements had higher levels of emotional distress at baseline. Conclusions 1 year after completing a multimodal inpatient program adolescents report less chronic pain, disability, and emotional distress. Clinically significant changes remain stable. Adolescents with high levels of emotional distress at admission may require special attention to maintain positive treatment outcomes. Specialized inpatient therapy is effective for children with chronic pain. Key words adolescents; clinical significance; headache; inpatient pain treatment; somatoform pain disorder.

Introduction

Chronic pain is a pervasive problem for the health care system that may begin in childhood and adolescence with ~5% suffering from severe and disabling chronic pain (Huguet & Miro, 2008).

Intensive multimodal pain treatment is considered as an adequate treatment option for these severely disabled children because then professionals from several disciplines can see the child daily in various situations, and a systematic change in pain-related disability and distress can be achieved. There are only few inpatient programs or intensive day care programs to date, and their conceptualization and labeling is diverse (Eccleston, Bruce, & Carter, 2006; Hechler, Dobe, & Zernikow, 2010b; Logan et al., 2012a; Logan, Conroy, Sieberg, & Simons, 2012b). Here, we follow the definition of a multimodal inpatient treatment (MIT) set out by the German Pain Society (Arnold et al., 2009) defined as a treatment provided by a minimum of three health care disciplines including medical, psychological, and physical therapy, which
are provided concurrently, generally lasting for 3 weeks. This treatment addresses underlying pain mechanisms, adapts pain medication, treats specific symptoms such as emotional distress, and teaches active coping skills (Hechler et al., 2010c; Maynard, Amari, Wieczorek, Christensen, & Slifer, 2009a).

Effectiveness of MIT has rarely been investigated and if so, comparison is hampered by the diversity of the programs. The few studies are diverse in terms of setting (some programs provide inpatient and outpatient treatment [Chalkiadis, 2001]), length of stay (ranging from 3-8 days to 3 weeks), and interventions provided (exercise therapy [Sherry, Wallace, Kelley, Kidder, & Sapp, 1999] compared with multimodal interventions [Eccleston, Malleson, Clinch, Connell, & Sourbut, 2003]). The program described by Maynard and colleagues can be classified as MIT. There are, however, other intensive multidisciplinary rehabilitation programs, which can not be classified as MIT as defined earlier but which share similarities in therapeutic approaches and program philosophy (e.g. [Logan et al., 2012a]). In addition, long-term follow-up has rarely been investigated. However, extended follow-ups are necessary to investigate the stability of treatment effects and factors that influence the stability of treatment effects (Maynard et al., 2009a). The only study that investigated long-term effects has been conducted by Sherry and colleagues (1999). This study followed 96 children for up to 7 years after a short exercise therapy and found high stability of treatment results, i.e. after 2 years, 88% of the children were still symptom free. However, their program was an exercise-based program, focused on children with complex regional pain syndrome (CRPS), and suffered from a large amount of dropout (50% after 2 years).

It is yet unclear which factors influence long-term treatment outcome. Given that there is numerous empirical support for the role of emotional variables on the maintenance and exacerbation of pediatric chronic pain and disability (e.g., [Cohen, Vowles, & Eccleston, 2010; Kashikar-Zuck, Goldschneider, Powers, Vaught, & Hershey, 2001; Sieberg, Williams, & Simons, 2011; Vervoort et al., 2011]), emotional distress may be an important factor. Vervoort et al. (2011) demonstrated recently that pain catastrophizing in schoolchildren was related to pain and disability 6 months later. They also found that children with increased anxiety at baseline were more inclined to report catastrophizing at follow-up. Whether this pattern holds true for children with chronic pain has not yet been investigated.

In addition, very few of the previous treatment studies analyzed the clinical significance (Jacobson & Truax, 1991) of treatment results as recommended by the Initiative for Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT) (Dworkin et al., 2009). Jacobson and Truax (1991) proposed a method for defining clinically significant change and suggest two criteria: (1) the magnitude of change between pre- and post-treatment scores should be statistically reliable. (2) By the end of the therapy, patients should move from a dysfunctional to a functional level to render them indistinguishable from healthy people. Thus far, only one study reported on the clinical significance of their treatment results 3 months after inpatient treatment (Hechler et al., 2009). They found that 3 months after treatment, 72% of the patients had clinically significant changes in pain intensity, and 45% had clinically significant changes in pain-related disability.

The present article aims to add to our limited knowledge on the long-term effects of MIT by investigating long-term treatment outcome 12 months after treatment compared with the pre-treatment status at admission. There were four objectives of the study: First, to assess 12 month outcomes of three primary outcomes (pain intensity, pain-related disability, school absences), with a focus on clinically significant changes. We expected significant and clinically relevant decreases in primary outcomes from admission to 12-month follow-up. Second, to classify children into two groups based on the composite measure of ‘‘overall improvement.’’ Third, to predict clinically relevant levels of overall improvement. We hypothesized that patient- and pain characteristics at admission would predict overall improvement at 12-month follow-up. Fourth, to assess the stability of change over time. Therefore, we defined the following groups based on overall improvement status at 3- and 12-month follow-up: (1) ‘‘stable long-term improvers’’ (improved on both assessments), (2) ‘‘short-term improvers’’ (improved at 3-month follow-up only), (3) ‘‘long-term improvers’’ (improved at 12-month follow-up only), and (4) ‘‘children with unsuccessful treatment results’’ (not improved at both follow-ups). We also aimed to investigate the influence of pain characteristics and emotional distress at admission on stability of change.

Materials and Methods
Participants
In the present study, we report data from extended 12-months follow-up of our earlier study (Hechler et al., 2009). Data from this sample have been described previously (Dobe, Damschen, Reiffer-Wiesel, Sauer, & Zernikow, 2006; Dobe, Hechler, Behlert, Kosfelder, & Zernikow, 2011; Hechler et al., 2009; Hechler, Blankenburg, Dobe, Kosfelder, Hübner, & Zernikow, 2010a; Hechler et al., 2010c), but this is the first time that the
clinical significant long-term outcomes are described. The study was a naturalistic longitudinal effectiveness study, with assessment at admission, 3 and 12 months after treatment. Between January 2004 and December 2006, adolescents that completed the 3-week inpatient chronic-pain treatment program at the German Paediatric Pain Centre were included. All children and their guardians were informed and agreed to their data being anonymously stored and processed for research purposes. The study was approved by the Ethics Committee of the Witten/Herdecke University.

During the study, 200 children and adolescents completed the program. Thirty-three were excluded from this analysis because they were aged <11 years. At admission, the 167 adolescents included in the final analysis were between the ages 11 and 18.3 years, mostly female (62%), and the majority (50%) had recurrent headache (see Table I for complete demographic information, and Table II for information about the level of functioning). In all, 141 (84%) provided data at the 3 months follow-up, and 136 (81%) provided data at the 12 months follow-up (see Figure 1). Participants completed the 12 month follow-up data on average 394 days after therapy.

Comparing those who dropped out of the study at 12-month follow-up to those who remained in the study, we found that dropouts were significantly older than completers (14 vs. 15 years; \(p < .01\)) and had higher levels of school aversion compared to dropouts (51 vs. 58; \(p < .01\)). There were no differences in pain characteristics at admission (all \(p > .1\)). At the 12-month follow-up, data were collected via mail for \(n = 105\) patients (63%), via telephone for \(n = 13\) patients (8%), and during a scheduled appointment at the German Paediatric Pain Centre for \(n = 18\) (11%) patients. Chi-square tests found no differences in sample characteristics (age, sex, diagnosis) and the way of responding (mail, phone, in-person, or drop-out) at the 12-month follow-up. We found however a difference between the way of responding and overall improvement (\(\chi^2 = 12.91; p = .002\); responses given via telephone indicated less improvement than those given via post and in-person.

### Outcome Measures

We assessed the following three measures as primary outcome measures: (1) mean pain intensity in the past 7 days, (2) pain-related disability comprising disability in daily life assessed by the Pediatric Pain Disability Index (Hübner et al., 2009), and (3) school absence assessed via parental report through the German Pain Questionnaire for Children and Adolescents (DSF-KJ) (Schroeder et al., 2010). Emotional distress and use of analgesics were secondary outcome measures. We thus cover three core outcome domains (pain intensity, role functioning, and emotional functioning) from the recent IMMPACT recommendations for pediatric patients (PedIMMPACT) (McGrath et al., 2008).

### Pain Intensity

Adolescents’ pain-intensity was assessed using a 11-point numeric rating scale (NRS) (Denecke & Hünseler, 2000) ranging from 0 (no pain) to 10 (maximum pain). This scale has been shown to be valid and sensitive to change in pediatric samples (von Baeyer et al., 2009).

### Pain-Related Disability

Pain-related disability was assessed using the P-PDI (Hübner et al., 2009). The P-PDI assesses disability in daily activities owing to pain on 12 items rated on a 5-point scale (1 [never] to 5 [always]). Daily activities comprise activities such as going to school, doing homework, engaging in physical activity, and sleeping. The total score is computed by summing all items and ranges from 12 to 60 (Cronbach’s alpha in this sample = .85). The 2-way imputation method (Sijtsma & van der Ark, 2003) was

<table>
<thead>
<tr>
<th>Variable</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (mean ± SD, range)</td>
<td>14.1 ± 1.91 (11–18.3)</td>
</tr>
<tr>
<td>Sex (n [%])</td>
<td>103 (62)</td>
</tr>
<tr>
<td>Diagnosis (n [%])</td>
<td></td>
</tr>
<tr>
<td>Headache</td>
<td>84 (50)</td>
</tr>
<tr>
<td>Recurrent abdominal pain</td>
<td>22 (13)</td>
</tr>
<tr>
<td>Back pain</td>
<td>16 (10)</td>
</tr>
<tr>
<td>Diffuse idiopathic pain</td>
<td>15 (9)</td>
</tr>
<tr>
<td>Disease-related pain</td>
<td>11 (7)</td>
</tr>
<tr>
<td>Fibromyalgia</td>
<td>8 (5)</td>
</tr>
<tr>
<td>Localized idiopathic pain</td>
<td>6 (4)</td>
</tr>
<tr>
<td>CRPS</td>
<td>5 (3)</td>
</tr>
<tr>
<td>Pain duration in months (mean ± SD, range)</td>
<td>42 ± 37</td>
</tr>
<tr>
<td>Patients on pain medication at preassessment n [%]</td>
<td>70 (62)</td>
</tr>
<tr>
<td>Mean pain intensity (mean ± SD, range)</td>
<td>6.7 ± 1.7</td>
</tr>
<tr>
<td>Pain intensity categories [%]</td>
<td></td>
</tr>
<tr>
<td>Mild pain</td>
<td>4</td>
</tr>
<tr>
<td>Moderate pain</td>
<td>42</td>
</tr>
<tr>
<td>Severe pain</td>
<td>55</td>
</tr>
</tbody>
</table>

Note: NRS = Numeric Rating Scale.

* Cumulative percentage > 100% owing to rounding up.

*The NRS was categorized into four categories: no pain (NRS 0), mild-to-moderate pain (NRS 1–3), severe pain (NRS 4–6), and very severe pain (NRS 7–10).
used to impute missing values when at least 9 of 12 items were responded to.

School Absence

Parents were asked how many days their child missed school owing to ongoing pain within the past 4 weeks (i.e., 20 school days excluding weekends and holidays) as part of the DSF-KJ (Schroeder et al., 2010). Thus, for children admitted short after public holidays, some parents had to report school absence before the holidays, but the number of expected school days was similar for all children, i.e., 20 days. Logan and colleagues provided evidence for a high correlation between parent report and official school attendance records (Logan, Conroy, Sieberg, & Simons, 2008b).

Emotional Distress

Emotional distress was assessed using the Anxiety Questionnaire for Pupils (AFS; [Wieczerkowski, Nickel, Janowksi, Fittkau, & Rauer, 1981]) and the Depression Inventory for Children and Adolescents (DIKJ; [Stiensmeier-Pelster, Schürmann, & Duda, 2000a]). Both measures are frequently used in adolescent samples and allow a norm-based comparison with non-clinical school children. We used these norms to transform individual raw-scores to T values with a mean of 50 and standard deviation of 10. We investigated three subscales of the AFS; (1) fear of exams, (2) generalized anxiety, and (3) school aversion, which all have good internal consistency (Cronbach’s alpha between .75 and .85; Wieczerkowski et al., 1981) The DIKJ consists of 26 items measuring depression with good reliability (internal consistency: 0.84) and validity (Stiensmeier-Pelster, Schürmann, & Duda, 2000b). Raw scores > 18 are indicative of depression (Stiensmeier-Pelster et al., 2000b). Individual missing items (up to two for the DIKJ and the scale school aversion, and up to three for the scales fear of exams and generalized anxiety) were replaced as suggested by the test authors.

Use of Analgesics

Parents indicated whether the children used analgesics before and after treatment as part of the DSF-KJ. Use of analgesics was then classified into five groups: (1) non-steroidal anti-inflammatory drugs (NSAID), (2) other non-opioid analgesics, (3) triptans, and (4) opioids.

Procedure

Data on the initial status were collected by sending a battery of questionnaires via mail before admission. Data for the follow-up were collected at the 3-month follow-up, which was regularly scheduled with the patients. Data for the 12-month follow-up were collected in three different ways: (a) during a scheduled appointment at the German Paediatric Pain Centre for those participants who were still in treatment, (b) via mail, or (c) via structured telephone interview for those participants who did not return their questionnaires. Children still in treatment were seen for follow-up visits scheduled 3, 6, or 12 months after inpatient treatment (see also Hechler et al.,

<table>
<thead>
<tr>
<th>Variable</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>P-PDI (n = 160; mean ± SD, range)a</td>
<td>40 ± 9</td>
</tr>
<tr>
<td>School absence of &gt;2 days (10%) within the preceding 4 weeks (n [%])b</td>
<td>122 (73)</td>
</tr>
<tr>
<td>School absence because of pain (n [%])</td>
<td></td>
</tr>
<tr>
<td>&lt;1% (3 days) school absence</td>
<td>45 (27)</td>
</tr>
<tr>
<td>15–50% (3–10 days) school absence</td>
<td>57 (34)</td>
</tr>
<tr>
<td>No school attendance at all</td>
<td>42 (25)</td>
</tr>
<tr>
<td>Significant emotional distress (n [%])</td>
<td></td>
</tr>
<tr>
<td>Fear of examination (n = 167)c</td>
<td>35 (21)</td>
</tr>
<tr>
<td>General anxiety (n = 167)c</td>
<td>61 (36)</td>
</tr>
<tr>
<td>School aversion (n = 167)c</td>
<td>34 (26)</td>
</tr>
<tr>
<td>Depression (n = 161)d</td>
<td>51 (31)</td>
</tr>
<tr>
<td>Note: P-PDI = Pediatric Pain Disability Index.</td>
<td></td>
</tr>
<tr>
<td>aCumulative percentage &gt;100% owing to rounding up. P-PDI values ranged between 12 and 60; the higher the value, the more disabled the child.</td>
<td></td>
</tr>
<tr>
<td>bSchool absence was assessed as days absent from school within the past 4 weeks (parental report).</td>
<td></td>
</tr>
<tr>
<td>cSignificant values in the anxiety questionnaire were assigned if the patient demonstrated T &gt; 60 compared with a non-clinical population.</td>
<td></td>
</tr>
<tr>
<td>dSignificant values in the depression questionnaire were assigned if the patient demonstrated raw values &gt; 18.</td>
<td></td>
</tr>
</tbody>
</table>
Responses were included if they were given maximum 18 months post discharge. The structured interview was conducted by a research assistant not involved in treatment.

Pain intensity and pain-related disability were assessed via self-report. School absence and use of analgesics were assessed by the parents. The sample size differs between the variables (see Table II) owing to >2 missing items on the questionnaires.

**MIT at the German Paediatric Pain Centre**

The MIT at the German Paediatric Pain Centre lasts 3 weeks. Children are referred to our specialized treatment center by primary physicians due to persisting pain. The treatment is based on operant and cognitive behavioral principles throughout all modules. Parents are actively engaged in the treatment as part of weekly family sessions and coaching sessions during which parents were taught to actively support their child to engage in healthy daily activities. Furthermore, reintegration into the child’s daily life is initiated from the second week onwards comprising visits home and to their home school on one appointed day. Patients complete various treatment modules such as education, individual sessions on pain-related coping and related problems, family sessions on parental cognitions and reactions towards the child’s pain, and group sessions on peer relationships that sum up to ~5–8 hr per day. The treatment is provided by a multiprofessional team including clinical psychologists, pediatricians, children and adolescent psychiatrists, pediatric nurses, physiotherapists, occupational therapists, and social workers. A scheduled appointment is arranged for each child and its family 3 months after discharge. More information can be taken from our earlier study (Hechler et al., 2009).

**Statistical Analysis**

Descriptive statistics (means, SD, and frequencies) of patients’ characteristics at admission were computed. Group differences between the different ways of responding at the 12 months follow-up were tested with chi-square tests.

**Treatment Effects at 12-Month Follow-Up—Statistical Significance**

Statistically significant changes between admission and 12-month follow-up in all continuous variables (pain intensity, pain-related disability, school absence, fear of exam, generalized anxiety, school aversion, and depression) were assessed using repeated-measures analysis of variance (ANOVA). Effect sizes were calculated according to Grawe and colleagues by dividing the difference between 12-month follow-up scores from prescores by the SD of the scores at admission (Grawe, Bernauer, & Donati, 1994). Effect sizes were interpreted according to established standards (Cohen, 1988). Significant changes in the use of analgesics (yes or no) were assessed using McNemar’s test for paired proportions (McNemar, 1947).

**Treatment Effects at 12-Month Follow-Up—Clinical Significance**

The clinically significant changes in the core variables were defined using the method developed by Jacobson and Truax (1991). This procedure entails two steps; (1) Computation of the reliable change index (RCI) to estimate whether the change for an individual patient is due to chance fluctuations or real change; (2) Calculation of cut-off points that need to be crossed to count as clinically relevant. The RCI was calculated using Jacobsen and Truax’s formular and resulted in three outcome groups: “no reliable change,” “reliable deterioration,” and “reliable improvement” for each patient and test. Cut-off points were defined using different methods, depending on whether normative data for a test existed.

1. If no normative data were available, a cut-off point at 90% of the patient population (mean before treatment + /- 1.6 SD) was used.
2. If a cut-off point already existed that indicates a clinically relevant point, (e.g., a t-value, which indicates that a score lies outside of the distribution of the healthy population), this was used.
3. If no cut-off score existed, but distribution of the healthy population was known, a cut-off point was chosen, at which a value is more likely to belong to a healthy than a clinical population.

In the present article, criterion 1 was used for pain-related disability, resulting in a cut-off score of 25.58. Criterion 2 was used for pain intensity, school absence, and the three anxiety scales. Specifically, we used the same the cut-off point for pain-intensity that has been identified in adult research, where a raw change of –1.74 on the 11-point NRS was found to be clinically significant (Farrar, Young, LaMoreaux, Werth, & Poole, 2001). This resulted in a cut-off point of 5 (6.82 /–1.74) for our sample.

The cut-off point for school absence could not be based on a clear definition, as there are no official data on school absence in Germany, and was thus based on our previous definition that school absence of >10% (i.e., 2 days) during a 4-week period would be considered as unacceptable by the teaching staff. The cut-off point for the three anxiety scales (60) was taken from published research with these scales (Wieczerkowski et al., 1981).
Criterion 3 was used for depression where data on the distribution of a healthy sample were available, and a raw score of $>18$ has been indicative of depression (Stiensmeier-Pelster et al., 2000b). Using these cut-offs together with the RCI, five outcome groups were defined for all variables:

1. “Clinically significant change”: Patients who reliably improved and whose post-scores were below the cut-off.
2. “Significant but not clinically relevant”: Patients who reliably improved but did not cross the cut-off.
3. “No change unproblematic”: Patients who showed no reliable change and had prescores below or equal to the cut-off.
4. “No change problematic”: Patients who showed no reliable change and had prescore above the cut-off.
5. “Reliable deterioration”: Patients who showed change in the undesired direction that was larger than the RCI.

To make our results comparable with other reports using only individual reductions in pain ratings as index for clinically significant change, we also calculated the number of patients who reduced their pain rating by at least 2 points on the NRS and at least 50% of their initial score.

Treatment Effects at 12-Month Follow-Up—Overall Improvement

We also computed an overall improvement score based on the clinically significant changes between admission and 12-month follow-up in the primary outcome variables (pain intensity, pain-related disability, school absence [Hechler et al., 2009]). Patients were classified as showing overall improvement if they did not show deterioration in any pain-related variable (i.e., in pain intensity, pain-related disability, and school absence) and had clinically significant improvements in one or both disability-related variables (pain-related disability or school absence). All other patients were classified as showing no overall improvement.

Furthermore, we compared the levels of overall improvement for patient and pain characteristics. Chi-square tests were used to compare diagnostic groups. Owing to the low group size, a mixed group of adolescents with fibromyalgia, localized idiopathic pain, and CRPS ($n = 19$) was formed in addition to the other five groups (headache, recurrent abdominal pain, back pain, diffuse idiopathic pain, disease-related pain). A logistic regression was used to predict overall improvement with pain intensity, pain-related disability, and emotional distress at admission, as well as sociodemographic variables such as age and sex.

Maintenance Groups Over the Two Time Points (3- and 12-Month Follow-Up)

First, we investigated whether there was a systematic change in the overall improvement status of the adolescents from 3- to 12-month follow-up using McNemar’s test. Specifically, we investigated whether more adolescents changed from no overall improvement at 3-month follow-up to overall improvement at 12-month follow-up or vice versa.

Second, we subdivided the children into four maintenance groups according to the stability of their overall improvement status at 3- and 12-months follow-up:

1. “stable long-term improvers”: Patients who showed stable overall improvement both at 3- and 12-month follow-up.
2. “long-term improvers”: Patients who showed no overall improvement at 3-month follow-up, but overall improvement at 12-month follow-up.
3. “short-term improvers”: Patients who showed overall improvement at 3-month follow-up, but no overall improvement at 12-month follow-up.
4. “children with unsuccessful treatment results”: Patients who showed stable no overall improvement both at 3- and 12-month follow-up.

To investigate pre-treatment influences on maintenance status, univariate ANOVA were used to compare the four maintenance groups in terms of pain-related and demographic variables; post-hoc tests (Tukey’s honestly significant difference [HSD]) were computed to test which of the four groups differed. Data were analyzed using R. We defined $p < .05$ as significant.

Results

Treatment Effects at 12-Month Follow-Up—Statistical Significance

Participants showed statistically significant reductions in all variables 12 months after treatment (see Table III). Participants showed statistically significant reductions in all variables 12 months after treatment such that they reported less pain, disability, and fewer school absences. Similarly, adolescents reported less emotional distress with lower levels of fear of exams, generalized anxiety,
school aversion, and depression. Effect-sizes for primary outcome measures were large. Effect-sizes for secondary outcome measures were of medium size.

We found a significant decrease in the number of adolescents taking pain medication (McNemar’s $\chi^2 = 10.26; p = .001$; Table IV).

**Treatment Effects at 12-Month Follow-Up—Clinical Significance**

In terms of the clinical significance of the changes, 74% of patients showed clinically significant changes in pain intensity, 53% in pain-related disability, and 39% in school absence (see Figure 1). For the secondary outcome variables, “unchanged unproblematic” was the most frequent category. Specifically, there were 27% who improved clinically significant in depression and 24% in generalized anxiety. Across all measures, between 1% (for fear of exams) and 7% (for depression), showed reliable deterioration.

There were 62% of the children who showed a decrease of at least 50% from the initial NRS score and 80% of the children who showed reductions of $-1.74$ from the initial NRS score.

**Maintenance Groups Over 3- and 12-Month Follow-Up**

Over the two time points, 53 (46%) of the adolescents were “stable long-term improvers,” 16 (14%) were “long-term improvers,” 22 (19%) were “short-term improvers,” 25 (22%) had an “unsuccessful treatment outcome.” There was no systematic change in the overall improvement status (either decrease or increase) between 3 and 12 months follow-up (McNemar’s $\chi^2 = .66; p = .4$).

Specifically, there were as many short-term improvers, as there were long-term improvers.

There were no differences between the maintenance groups in terms of diagnosis ($\chi^2 = 16.76; p = .33$) and those who participated versus those who did not participate in the 3-month follow-up ($\chi^2 = 0.56; p = .45$). ANOVAs comparing the four improvement groups in terms of pretreatment psychosocial variables found that the four groups significantly differed in school absence ($F = 3.03; p < .05$), generalized anxiety ($F = 2.90; p < .05$), and depression ($F = 2.95; p < .05$). Post-hoc tests revealed that “stable long-term improvers” adolescents had significantly higher rates of school absence at admission than “unsuccessful” adolescents. Furthermore, “short-term improvers” had higher levels of generalized anxiety and depression than “stable long-term improvers” adolescents (both $p < .05$).

**Discussion**

The present study presents data on the statistical and clinical significance of treatment effects 1 year after an interdisciplinary MIT (Hechler et al., 2009). Our results show that the majority of children exhibit statistically and clinically significant changes in core outcome measures 12 months after treatment. Changes in secondary outcomes were also observable albeit smaller. Importantly, the majority of adolescents reported no significant emotional distress at admission. The majority of children showed overall

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**Table III. Statistically Significant Changes in Outcomes From Admission to One-Year Follow-Up**

<table>
<thead>
<tr>
<th></th>
<th>Admission</th>
<th>3 months follow-up</th>
<th>12 months follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$M$ (SD)</td>
<td>$M$ (SD)</td>
<td>$M$ (SD)</td>
</tr>
<tr>
<td>Pain intensity (n = 120)</td>
<td>6.83 (1.78)</td>
<td>2.83 (2.66)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Pain-related disability (n = 109)</td>
<td>39.19 (8.51)</td>
<td>22.82 (9.06)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>School absence (n = 120)</td>
<td>9.25 (7.76)</td>
<td>1.8 (3.92)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Fear of exams (n = 72)</td>
<td>47.43 (12.37)</td>
<td>43.18 (11.05)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Generalized anxiety (n = 72)</td>
<td>52.96 (12.73)</td>
<td>46.6 (11.67)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>School aversion (n = 72)</td>
<td>51.94 (10.87)</td>
<td>47.92 (9.95)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Depression (n = 73)</td>
<td>13.19 (7.89)</td>
<td>8.59 (6.92)</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Statistically Significant Changes in Outcomes From Admission to One-Year Follow-Up</td>
<td>$\chi^2 = 10.26; p = .001$</td>
<td>$\chi^2 = 0.56; p = .45$</td>
<td>$\chi^2 = .66; p = .4$</td>
</tr>
</tbody>
</table>
The most likely explanation for this is that only a minority of our sample exhibited clinically relevant levels of depression and anxiety at admission and thus could not show clinical improvements. Also, the program may have prevented an aggravation of emotional distress so that most children could maintain a low level of emotional distress.

We extend previous studies (Hechler et al., 2009; Eccleston et al., 2003; Lanzi et al., 2007; Maynard et al., 2009a; Palermo & Scher, 2001) by showing that adolescents are able to maintain these positive treatment effects over a period of 1 year. Here, positive long-term outcomes for a 12-month period could be replicated with a lower drop-out rate (32% compared with 50% by Sherry et al., 1999) and a stricter definition of overall improvement (based on the concept of clinical significance of Jacobson and Truax) compared with previous studies (Sherry et al., 1999). Hence, in line with existing studies, the present study highlights the importance of MITs for severely disabled children. Several possible explanations accounting for the effectiveness have previously been discussed: The treatment intensity of the inpatient setting, changes in coping strategies, the ability to engage in activities with peers, and experiences of success (Eccleston et al., 2003; Hechler et al., 2009; Hechler et al., 2010c; Lanzi et al., 2007; Maynard, Amari, Wieczorek, Christensen, & Slifer, 2009b; Palermo & Scher, 2001; Sherry et al., 1999). Future studies are warranted to examine the exact mechanisms of change, i.e., which changes for instance in cognitive or emotional variables such as pain catastrophising (Vervoort et al., 2011), or treatment adherence (Simons, Logan, Chastain, & Cerullo, 2010) may moderate changes in primary outcomes (Keefe, Buffington, Studts, & Rumble, 2002).

In addition, future research should address two practical issues (Jensen & Foster, 2010): First, the cost-benefit ratio needs to be assessed in a large group of patients to make possible financial incentives more obvious (Maynard et al., 2009b; Palermo & Scher, 2001; Sherry et al., 1999). Future studies are warranted to examine the exact mechanisms of change, i.e., which changes for instance in cognitive or emotional variables such as pain catastrophising (Vervoort et al., 2011), or treatment adherence (Simons, Logan, Chastain, & Cerullo, 2010) may moderate changes in primary outcomes (Keefe, Buffington, Studts, & Rumble, 2002).

### Table V. Predictors of Overall Amelioration at 12 Months Follow-Up

<table>
<thead>
<tr>
<th>Predictors</th>
<th>Coefficient</th>
<th>SE</th>
<th>OR (standardized OR)</th>
<th>95% CI lower</th>
<th>95% CI upper</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>School absence at admission</td>
<td>0.07</td>
<td>0.03</td>
<td>1.07 (1.73)</td>
<td>1.02</td>
<td>1.14</td>
<td>&lt;.05</td>
</tr>
<tr>
<td>Pain intensity</td>
<td>−0.1</td>
<td>0.13</td>
<td>0.91 (.84)</td>
<td>0.7</td>
<td>1.16</td>
<td>n.s.</td>
</tr>
<tr>
<td>Pain-related disability</td>
<td>0.03</td>
<td>0.03</td>
<td>1.03 (1.34)</td>
<td>0.98</td>
<td>1.09</td>
<td>n.s.</td>
</tr>
<tr>
<td>Fear of examinations</td>
<td>0</td>
<td>0.02</td>
<td>1 (.99)</td>
<td>0.95</td>
<td>1.05</td>
<td>n.s.</td>
</tr>
<tr>
<td>General anxiety</td>
<td>−0.01</td>
<td>0.03</td>
<td>0.99 (.86)</td>
<td>0.93</td>
<td>1.04</td>
<td>n.s.</td>
</tr>
<tr>
<td>School aversion</td>
<td>−0.03</td>
<td>0.03</td>
<td>0.97 (.74)</td>
<td>0.92</td>
<td>1.02</td>
<td>n.s.</td>
</tr>
<tr>
<td>Depression</td>
<td>−0.02</td>
<td>0.04</td>
<td>0.98 (.84)</td>
<td>0.89</td>
<td>1.07</td>
<td>n.s.</td>
</tr>
<tr>
<td>Age</td>
<td>−0.22</td>
<td>0.12</td>
<td>0.8 (67)</td>
<td>0.63</td>
<td>1.02</td>
<td>n.s.</td>
</tr>
<tr>
<td>Sex</td>
<td>−0.08</td>
<td>0.5</td>
<td>0.92 (.96)</td>
<td>0.34</td>
<td>2.47</td>
<td>n.s.</td>
</tr>
</tbody>
</table>
et al., 2009a). Second, it is important to investigate inpatient treatment in the context of other treatment options. Most participants proceed through the health care system in various steps of treatment intensity, such as starting with primary care, and proceeding to specialized treatment if the pain persists. Here, they may be referred to outpatient or intensive inpatient treatment dependent on their clinical status (Hechler et al., 2011a; Von Korff & Moore, 2001). Further research should investigate the feasibility of a stepped-care approach in which treatment can be intensified over time (Hechler et al., 2011a). Within such a framework, screening tools that detect children at risk of developing chronic pain and explicit decision criteria for treatment allocation can be developed (Vowles, Jordan, & Eccleston, 2009).

**Maintenance of Improvement**

We found that 50% showed improvement at both follow-ups, 20% were not significantly improved at any of the two follow-ups, and ~30% of the participants showed improvement 3 months after treatment, but not 12 months after treatment or vice versa. Our analysis of pre-admission differences between these groups revealed important differences. First, stable long-term improvers had the highest levels of school absence at admission. This is in accordance with the finding that adolescents with high school absence at admission were more likely to display overall improvement at 3-month follow-up (Hechler et al., 2009). These children may have been entrenched in the vicious cycle of pain, disability, and school absence at admission. As demonstrated by Logan et al. (2008b), their academic achievement may have decreased and contributed to long-term disability. These children may, however, be highly academically competent (Logan et al., 2008b). Once these children have surmounted their disability and pain, they may therefore be able to quickly reintegrate into school functioning. This may lead to experiences of success, positive contact with peers, and teachers, which may prevent deterioration and relapse over time. These suggested mechanisms need to be further explored in future studies.

Second, short-term compared with stable long-term improvers had higher levels of emotional distress at admission. Possibly, deterioration in their emotional distress over time, especially from 3- to 12-month follow-up, may have lead to increases in pain and disability at 12-month follow-up. The relationship between increased emotional distress and pain and disability has been extensively studied in children (Cohen et al., 2010; Kashikar-Zuck et al., 2001). Our findings suggest that children with increased emotional distress at admission benefit from the intensive inpatient treatment as suggested by

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**Figure 2.** Percentage of adolescents demonstrating clinically significant changes in the seven outcome variables at 12-month follow-up. Displayed are the percentages of children for the five outcome groups: Clinically significant change; Positive change, but not clinically significant; No change—unproblematic; No change—problematic; Reliable deterioration.
improvements in primary outcomes. They deteriorate, however, with time. The patterns of these deteriorations need to be further investigated. On the basis of these findings, additional treatments for these children may need to be conceptualized specifically addressing the aspect of emotional distress to prevent relapse and deterioration such as how to cope with pain when emotional distress increases again.

Finally, children with unsuccessful outcome did not show particularly high levels of emotional distress or high levels in school absence. Different to children high in school absence at admission, they did not experience success when being able to attend school again. Whether these children really do not experience emotional distress, or whether these children are high in social desirability and deny emotional distress (Logan, Claar, & Scharff, 2008a) needs to be investigated in future studies. In addition, other potential candidates that may account for unsuccessful treatment results such as the child’s pain-specific emotions (fear of pain [Simons, Sieberg, Carpino, Logan, & Berde, 2011]) or pain-specific cognitions (Vervoort et al., 2011) and parental reactions such as adherence to treatment (Simons et al., 2010) or parental cognitions and reactions (Hechler et al., 2011b; Vervoort et al., 2011) warrant further investigation.

**Limitations**

There are some limitations inherent of the present study. First, the study is limited by the lack of a control group. However, there is little reason to suspect spontaneous changes in the participants we studied (Perquin et al., 2003), and the inclusion of a waiting-list control group would have been highly unethical, given the known effectiveness of our program. Second, we found unexpected differences in the overall improvement rates depending on the way in which participants responded. Improvement rates were lower for those who responded by phone, compared with those who responded in person at a scheduled follow-up visit. The latter group had contacted the tertiary institute for a follow-up visit. During this follow-up visit, the current status of the child is evaluated. Dependent on the child’s status, further pain management strategies are adjusted to the needs of the child (Hechler et al., 2011b). At this stage, we do not if the difference in improvement rates is due to the additional treatment or a social desirability effect. Possibly, families who were interviewed by a research assistant were more willing to report negative aspects than those faced with the clinicians in person. In light of the small sample size, only 13 responded by telephone; this pattern needs to be replicated before any strong conclusions can be drawn. Third, our outcome measures relied on self-report. More objective measures of pain-related disability have only recently been used in samples with chronic pain (Wilson & Palermo, 2012). Similarly, this study did not include a measure of the economic benefit. Fourth, we dichotomized our outcome variables. This greatly enhances the communication of results. On the other hand, this clearly leads to a drop in power. At present, there is no statistical method that maintains maximal power, and can be easily interpreted for binary outcomes and predictors; thus the use of the Jacobson and Truax method is recommended (Lambert & Ogles, 2009). It will be most important to develop methods that classify individuals’ change over time, and at the same time retain the comparability across studies. On a similar note, cut-off points for clinically relevant relative changes in pain intensity need to be determined for children and adolescents (Farrar et al., 2001). Also absolute levels of clinical significant pain that are required according to Jacobsen and Truax to differentiate the “no change problematic” from the “no change unproblematic” group are not established for children and adolescents. At present, it is not clear whether methods based on relative or absolute cut-offs are more relevant to patients. Although relative cutoffs may overestimate small changes in almost healthy patients, absolute cutoffs may overestimate small changes near the border between healthy and diseased. The application of the adult cut-off score may have hampered results. For better study comparison, we therefore integrated report on children displaying a 50% pain reduction. Finally, we assessed general emotional distress and no pain-specific emotions such as fear of pain (Simons et al., 2011). Future treatment studies should implement assessment of pain-specific emotions to investigate its potential influence on treatment results.

The clinical implications of our results can be summarized as follows: (1) the MIT is effective in improving the overall status of children over long periods. (2) However, there were 20% with an overall unsuccessful treatment result and 30% who showed only short-term or long-term improvement. (3) Emotional distress seems to play a crucial role for long-term improvement, given that adolescents with high levels of emotional distress displayed a risk to relapse after short-term improvements. These adolescents may require special attention and interventions that are useful in enhancing maintenance of positive outcomes for this particular group of patients.

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References


