DOI: 10.1002/ejp.1754

ORIGINAL ARTICLE

EJP European Journal of Pain

Long-term outcomes of children with severe chronic pain: Comparison of former patients with a community sample

Julia Wager[1,2,3] | **Ann-Kristin Ruhe**[1,2] | **Lorin Stahlschmidt**[1] | **Kathrin Leitsch**[2] | **Benedikt B. Claus**[3] | **Winfried Häuser**[4] | **Elmar Brähler**[5,6] | **Andreas Dinkel**[4] | **Rüya Kocalevent**[7] | **Boris Zernikow**[1,2,3]

[1]German Paediatric Pain Centre, Children's and Adolescents' Hospital, Datteln, Germany

[2]Department of Children's Pain Therapy and Paediatric Palliative Care, Faculty of Health, School of Medicine, Witten/Herdecke University, Witten, Germany

[3]PedScience Research Institute, Datteln, Germany

[4]Department of Psychosomatic Medicine and Psychotherapy, Klinikum rechts der Isar, School of Medicine, Technical University of Munich, Munich, Germany

[5]Department of Medical Psychology and Medical Sociology, University of Leipzig, Leipzig, Germany

[6]Clinic and Polyclinic for Psychosomatic Medicine and Psychotherapy, University Medical Center Mainz, Mainz, Germany

[7]Institute and Polyclinic of Primary Medical Care, University Medical Centre Hamburg-Eppendorf, Hamburg, Germany

Correspondence
Dr. Julia Wager, German Paediatric Pain Centre, Children's and Adolescents' Hospital, Datteln; Witten/Herdecke University; Dr.-Friedrich-Steiner Str. 5, Datteln, 45711, Germany.
Email: j.wager@deutsches-kinderschmerzzentrum.de

Funding information
None.

Abstract

Background: Findings on the short- and long-term effectiveness of intensive interdisciplinary pain treatment (IIPT) for children with severe chronic functional pain are promising. However, a definitive appraisal of long-term effectiveness cannot be made due to a lack of comparison groups. The aim of the present study was to compare the health status of former patients with the health status of an age- and sex-matched comparison group from the community.

Methods: Data from two samples, a clinical sample of former patients (n = 162; aged 14 to 26) and an age- and sex-matched community sample (n = 162), were analysed. Former patients provided data 7 years after IIPT. Pain characteristics, physical and mental health status, autonomy, coping and health care utilisation were compared between the two samples.

Results: Seven years after treatment, the majority (58%) of the clinical sample were completely pain-free. Compared to the community sample, the clinical sample demonstrated worse physical and mental health and continued to seek more frequent health care, irrespective of whether or not they experienced ongoing chronic pain. However, the clinical sample reported better coping strategies and a comparable level of autonomy.

Conclusion: Patients experiencing severe chronic pain in childhood who engage in IIPT are likely to have recovered from their pain in early adulthood. Long-term treatment effects may manifest in better coping strategies. However, reduced mental and physical health status may indicate a negative long-term effect of early chronic pain experiences or a general vulnerability in people developing a chronic pain condition in childhood.

Significance: The majority of severely impaired paediatric chronic pain patients no longer suffer from chronic pain seven years after intensive interdisciplinary pain treatment. However, former patients have worse physical and mental health status than a community sample, and continue to seek out more frequent health care utilisation, irrespective of whether or not they continue to experience chronic pain.

This accompanies the following article: Wallace, DP & Gauntlett-Gilbert, J. Broadening the scope of paediatric intensive interdisciplinary pain treatment to promote future resilience and psychological flexibility. Eur J Pain. 2021; 25: 1177–1178. https://doi.org/10.1002/ejp.1778

Therefore, potential negative long-term effects of childhood chronic pain experiences need specific attention early on.

1 | INTRODUCTION

Between 5% and 8% of children and adolescents experience severely disabling chronic pain resulting in high health care costs and serious limitations in functioning, everyday life and emotional well-being (Groenewald et al., 2014; Huguet & Miró, 2008; Könning et al., 2021). Most of these chronic pain conditions are functional, i.e. they cannot be explained by a defined medical illness (Zernikow et al., 2012). For severely impaired patients, first-line treatment options, ranging from primary care to specialized care by orthopaedists, neurologists, rheumatologists, gastroenterologists or surgeons are often inefficient (Kaufman et al., 2016; Könning, Rosenthal, Friese, et al., 2021; Wager et al., 2019). In case of treatment failure of single-disciplinary treatments the preferred therapy is an intensive interdisciplinary approach that addresses the complexity of a chronic pain condition (Simons et al., 2013). The short-term effectiveness of this kind of treatment has been well documented (Hechler et al., 2015; Stahlschmidt et al., 2016). Due to the high risk of paediatric chronic pain persisting into adulthood when treatment is ineffective (Brattberg, 2004; Brna et al., 2005; Hestbaek et al., 2006; Walker et al., 2010), it is of great interest whether positive treatment effects persist long-term. Few studies have analysed the long-term effects of intensive interdisciplinary pain treatment (IIPT) for children and adolescents with severe chronic pain, with all available studies employing pre/post comparisons to determine outcomes (Banez et al., 2014; Kashikar-Zuck et al., 2019; Randall et al., 2018; Zernikow et al., 2018). These studies generally indicate a reduction in pain intensity, functional disability and health care utilisation (Banez et al., 2014; Kashikar-Zuck et al., 2019; Randall et al., 2018; Zernikow et al., 2018). The proportion of former patients completely recovering from pain varied between studies. While one study on patients with fibromyalgia reported that 8 years after discharge 100% still had one or more fibromyalgia symptoms (Kashikar-Zuck et al., 2019), studies on patients with musculoskeletal or mixed functional chronic pain report that 30% (Randall et al., 2018) to 40% (Zernikow et al., 2018) were pain-free at the long-term follow-up. Even though it is a core goal of IIPT to return youth with pain to typical levels of functioning (Stahlschmidt et al., 2016), it remains uncertain whether the above-mentioned promising outcomes of former IIPT patients are comparable to the broader population.

The aim of the present study was to address the knowledge gap regarding returning to normalcy and long-term outcomes by determining whether former patients differ seven to eight years after IIPT from an age- and sex-matched community-based comparison group regarding their pain characteristics and secondary outcomes, such as physical and mental health status, autonomy, coping style and health care utilisation.

2 | METHOD

2.1 | Sample and Setting

We analysed data from two samples: a clinical sample and a community sample. The clinical sample comprised former patients who sought IIPT between 2009 and 2011. At this institution, IIPT is a manualized, inpatient-based treatment approach delivered by a multi-professional team with a three- to four-week duration. Children and adolescents with severe pain conditions who have not responded to single-disciplinary treatment are eligible for this cognitive behavioural therapy-focused programme. Treatment modules address patient education, realistic goal setting, pain management strategies, the treatment of co-occurring emotional distress, pharmacological treatment if applicable, physiotherapy and relapse prevention. The programme is described in detail in a treatment manual (Dobe & Zernikow, 2019).

At the time of data collection, the clinical sample ($n = 162$) was aged between 14 and 26 years and mostly female. They had received IIPT seven to eight years prior ($M = 7.35$, $SE = 0.06$). At admission, the pain condition lasted for an average of 33 months ($SE = 2.76$); the majority reported the head as the main pain location (46%), followed by musculoskeletal (27%) and abdominal pain (17%); 10% had multiple main pain locations and 50% had constant pain. The age- and sex-matched comparison group was drawn from a sample representative of the general population (Häuser et al., 2014). Table 1 provides an overview of the sociodemographic characteristics of the two samples. After matching, in both samples, the majority was female (70%; $\chi^2 = 0.0$; $p = 1.0$) and the mean age was 21 years ($Z = -0.020$; $p = .984$).

2.2 | Procedure

The clinical sample was first contacted via telephone. Of 296 potential study participants, 228 (77%) were reached. After consenting to participate, $n = 162$ (participation rate 71%) completed a short telephone interview with key outcome measures. These $n = 162$ participants compose the study sample for main analyses. Additional data were collected with an online survey that was sent via email to those who

TABLE 1 Sociodemographic characteristics of the clinical and community samples

	Clinical sample ($n = 162$)		Community sample ($n = 162$)	
Age (in years), Mean (SE)[a]	21.1	(0.01)	21.1	(0.02)
Sex (female), *n* (%)	113	(69.8%)	113	(69.8%)
German nationality, *n* (%)	161	(99.4%)	159	(98.1%)
Marital status, *n* (%)				
Married /live together	2	(1.2%)	8	(4.9%)
Married / separated	1	(0.6%)	0	(0.0%)
Single	159	(98.1%)	152	(93.8%)
Divorced	0	(0.0%)	2	(1.2%)
Employment, *n* (%)				
Full time (≥35 hr)	28	(17.3%)	50	(30.9%)
Part-time (15–34 hr)	6	(3.7%)	12	(7.4%)
Vocational training	44	(27.2%)	23	(14.2%)
School education	70	(43.2%)	54	(33.3%)
Other[b]	14	(8.6%)	23	(14.2%)

[a]Age range for both groups was 14 – 26 years.

[b]Hourly employment, military/community service, maternity/parental leave, unemployed, short-time work, housewife/-husband, incapacitated.

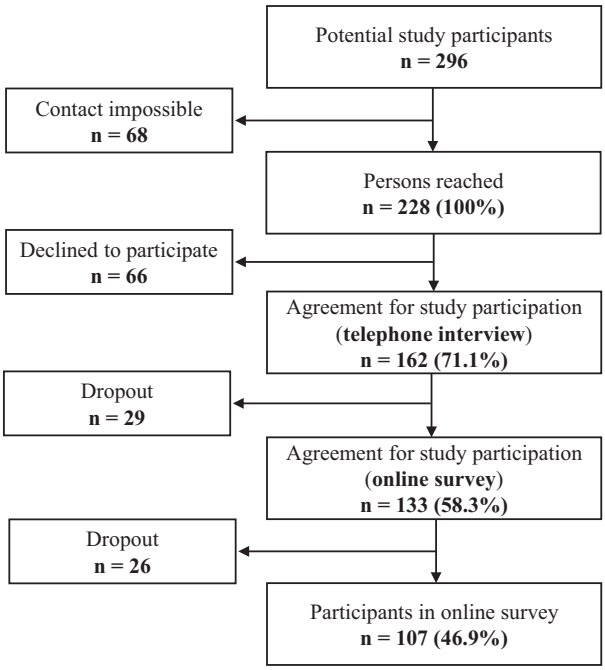

FIGURE 1 Flowchart of the recruiting of the clinical sample

agreed to provide more detail ($n = 133$); $n = 107$ participants completed the online survey (shown in Figure 1). Analyses including data from the online survey were only carried out with the $n = 107$ participants. A drop-out analysis revealed that former patients not participating in the study ($n = 134$) did not differ from participants ($n = 162$) regarding sex, age, pain location, pain intensity, presence of constant pain, pain-related disability, anxiety and depression at initial presentation. Also, no differences regarding those parameters were identified between participants taking part in the telephone interview only ($n = 55$) and those participating in the telephone interview and online survey ($n = 107$).

For the community sample, data collection was conducted by an independent institution for opinion and social research (USUMA, Berlin) between May and June 2013. For a detailed description of the study procedure, see the original publication (Häuser et al., 2014).

2.3 | Measures

The survey for the clinical sample was matched to questionnaires previously delivered to the community sample (shown in Table. 2).

Questions about **demographic characteristics** included age, sex, citizenship, marital status and employment status.

Regarding **pain characteristics**, participants were asked about the presence of constant or frequent pain in the past three months. Additionally, **pain characteristics** were assessed by means of the **Chronic Pain Grading (CPG)**

TABLE 2 Overview of the study measures and data collection method

Section	Questionnaire	Assessment method [a]
Demographics	Demographic characteristics	Phone
Pain	Short pain questionnaire (Klasen et al., 2004)	Phone
Physical and mental health	Self-administered comorbidity questionnaire (SCQ-D) (Sangha et al., 2003)	Online
	Giessen Symptom Questionnaire (GBB−8) (Kliem et al., 2017)	Phone
	Patient Health Questionnaire−4 (PHQ−4) (Löwe et al., 2010)	Online
Autonomy and coping	Perceived Autonomy (PA) (Warner et al., 2011)	Online
	Brief Resilient Coping Scale (BRCS) (Kocalevent et al., 2017)	Online
Health care utilisation	BGS98-short (German National Health Survey 1998) (Bergmann, 1999)	Phone

[a]Assessment method is for the clinical sample only. All measures were collected through online survey for the community sample.

(Klasen et al., 2004). If permanent or frequent pain was present in the prior 3 months, the CPG questionnaire measures the current, average and highest pain intensity in the past four weeks using numeric rating scales (NRS with 0 = no pain and 10 = worst pain), combined with impairment in everyday, leisure and work activities on a scale from 0 ('no impairment') to 10 ('no activity possible'). Additionally, the number of days in the past three months ordinary activities were impossible due to pain was recorded. Based on these responses, patients were classified into five chronic pain grades: CPG 0: no chronic pain; CPG I: low disability, low intensity; CPG II: low disability; high intensity; CPG III: high disability, moderately limiting; CPG IV: high disability, severely limiting. Psychometric properties of the German version of the CPG have been investigated in an adult sample aged ≥ 18 (Klasen et al., 2004). Even though a CPG version for adolescents exists (Wager et al., 2013), for reasons of comparability with the community sample, study participants below 18 years (n = 9) also completed the adult version. The CPG demonstrates good validity through high correlations with disability measures, measures of grading and staging chronic pain and the frequency of physician visits and analgesic use (Klasen et al., 2004).

Physical and mental health status were assessed by use of three self-report questionnaires:

- The **self-administered comorbidity questionnaire (SCQ-D)** (Sangha et al., 2003) as an indicator of current disease load measures the presence of 13 common diseases (e.g. hypertension, heart disease, depression) based on self-report. For each present comorbidity, the participant receives one point (range 0–13), so that higher values indicate a greater comorbid disease load (Sangha et al., 2003; Streibelt et al., 2012). The SCQ-D has been validated in an adult sample and shows moderate associations with the Charlson comorbidity index and health care utilisation as well as good predictive validity regarding health status and resource utilisation one year later. This measure was also applied to adolescents below 18 years in the community sample and accordingly in the clinical sample.
- The Giessen Symptom Questionnaire **(GBB-8)** (Kliem et al., 2017) provides subjective information about the current burden of eight different symptoms. The severity of each symptom is rated from 'not at all' (0) to 'very much' (4). The eight symptoms are assigned to four subscales of complaints: exhaustion, abdominal complaints, limb pain, heart problems. The GBB-8 total value ranges from 0 to 32; each of the four subscales ranges from 0 to 8. A higher score indicates a higher burden due to complaints. The GBB-8 is validated from the age of 14 years onwards. Construct validity has been shown by means of associations with severity of mental disorders and health care utilisation as well as by invariance of the factor structure across gender and age groups (Kliem et al., 2017).
- The **Patient Health Questionnaire-4 (PHQ-4)** (Löwe et al., 2010) is a valid and reliable short screening instrument for anxiety and depression in people aged 14 years and older. In two items for each subscale, it assesses the frequency of occurrence of the core symptoms of anxiety and depression over the last 2 weeks with the response options 'not at all' (0), 'several days' (1), 'more than half the days' (2) and 'nearly every day' (3). Accordingly, values range from 0 to 12, higher values indicate more symptoms; values ≥ 6 are increased (96th percentile of the normal population). Additionally, separate scores for anxiety (GAD-2) and depression (PHQ-2) can be calculated (range 0–6) (Löwe et al., 2010).

Perceived autonomy was assessed by a questionnaire with four items regarding the independent management of everyday life and shaping life in the way one chooses (e.g. 'I organize my life according to my own ideas'. or 'I live by my own choices'.). Items are rated from 'strongly disagree' (1) to 'strongly agree' (4) (total score range 4–16). Higher values indicate a more autonomous lifestyle. The questionnaire has shown good reliability in a sample of adults (Warner et al., 2011). This measure was also applied to adolescents in the community sample and accordingly in the clinical sample.

The **Brief Resilient and Coping Scale (BRCS)** is a behaviour-based self-report measure on the tendency to flexibly use active coping strategies (e.g. 'I look for creative ways to alter difficult situations' or 'I actively look for ways to replace the losses I encounter in life'). The four items on the current coping behaviour are answered on a 5-point Likert scale range from 'describes me not at all' (1) to 'describes me very well' (5). That results in a sum score ranging from 4 to 20; higher values indicate better coping abilities (Kocalevent et al., 2017; Sinclair & Wallston, 2004). The questionnaire has been validated in a sample aged ≥ 14 years (Kocalevent et al., 2017) showing good construct validity and a stable factor structure across age groups.

Health care utilisation was assessed by items from the **German Federal Health Survey** (Bergmann, 1999). Participants were asked if they visited a general practitioner, a psychotherapist, or another specialist in the past 12 months.

2.4 | Ethics

The current study was approved by the Ethics Committee of the Witten/Herdecke University (134/2018). Participants of the clinical sample provided informed consent for data collection, electronic storage and data analysis. For adolescents younger than 16 years parents additionally provided their informed consent. Ethical approval for the study collecting the

community data was provided by the University of Leipzig (Az 092–12–05032012). All participants provided informed consent. For adolescents under the age of 18 years, the parents or guardians also gave informed consent.

2.5 | Data analysis

Propensity score matching was used to assign each clinical individual to an appropriate individual from the community sample. We implemented this in R (R Core Team, 2019) with the package MatchIt (Ho et al., 2011; Imai, 2018). The matching method was set to 'optimal', requiring the additional package optmatch (Hansen et al., 2019; Hansen & Klopfer, 2006) with a ratio set to 1, thus assigning one clinical to exactly one individual from the community sample.

Differences in pain characteristics between the clinical and the community sample were tested with chi-squared (χ^2) tests for nominal variables and, due to skewed distributions, Yuen's non-parametric two-sample trimmed mean test (Yuen, 1974) for continuous or ordinal variables. The 20% trimmed mean (M_T) was preferred over the median as the latter assumes that all data, except for one point (the median itself), contain contaminated data. We refer to M_T as the trimmed mean, SE_T as the trimmed standard error and h as the number of observations left after trimming. Analyses were performed using R (R Core Team, 2019) with the WRS2 package for robust methods (Mair & Wilcox, 2019).

Differences between samples regarding secondary outcomes were analysed taking into account the pain status. Therefore, the two-way design hypothesis test based on 20% trimmed means (Wilcox, 2012) was calculated with the dependent variables 'sample' and 'pain'.

For nominal variables tested with χ^2-test, the Cramer's V effect size and the 95% confidence interval (CI) are reported. For ordinal and continuous data, we report the explanatory measure effect size ξ (Wilcox, 2012) as well as its 95% CI. For both effect sizes, values of 0.1, 0.3 and 0.5 can be interpreted as small, moderate and strong (Cohen, 1988). The significance level was set at $p < 0.05$.

3 | RESULTS

3.1 | Pain characteristics

Seven years after IIPT, 72% of the clinical sample had a CPG 0 or I, indicating no chronic pain or pain with low disability and low intensity. Regarding pain intensity, there was no significant difference between those individuals with chronic pain in the clinical and the community samples. However, due to higher functional impairment, the clinical sample was more frequently assigned to higher CPG compared to the community sample (shown in Table 3).

3.2 | Physical and mental health status

The clinical sample reported a significantly higher number of physical and mental health comorbidities compared to the community sample (shown in Table 4 SCQ-D, effect S) and participants with chronic pain had more comorbidities than those without pain (shown in Table 4: SCQ-D, effect P). No interaction effect between sample and pain status was identified (shown in Table 4: SCQ-D, effect S x P), indicating that comorbidities were significantly more prevalent in

TABLE 3 Pain characteristics of the clinical and the community samples

	Clinical sample	**Community sample**			
Pain characteristic	***n*** **(%)**	***n*** **(%)**		***p***	**Effect size [95% CI]**
Chronic pain	*n = 161* 67 (41.6%)	*n = 162* 22 (13.6%)	$\mathbf{X^2 = 31.79}$	**<0.001**	$\mathbf{V = 0.31}$ **[0.21, 0.42]**
Mean pain intensity[a], M_T (SE_T)	*h = 41* 5.05 (0.25)	*h = 14* 4.00 (0.54)	T_y (19.18) = 1.85	0.080	ξ = 0.35 [0.00, 0.68]
Chronic Pain Grade (CPG)	*n = 161*	*n = 162*	$\mathbf{T_y}$ **(96) = 4.29**	**<0.001**	$\boldsymbol{\xi}$ **= 0.50 [0.29, 0.64]**
0	94 (58.4%)	140 (86.4%)			
I	22 (13.7%)	10 (6.2%)			
II	21 (13.0%)	8 (4.9%)			
III	12 (7.5%)	4 (2.5%)			
IV	12 (7.5%)	0 (0.0%)			

Abbreviations: M_T, 20% trimmed mean, SE_T, trimmed standard error, h, number of observations left after trimming.

CPG 0: no chronic pain; CPG I: low disability, low intensity; CPG II: low disability; high intensity; CPG III: high disability, moderately limiting; CPG IV: high disability, severely limiting. Significant results are marked in bold.

[a]pain intensity in the last 4 weeks.

TABLE 4 Comparison of comorbidities and self-reported health status by sample (clinical or community) and pain status (chronic pain absent or present)

		M_T (SE_T)					
Measure		**Clinical**	**Community**	**Effect**	***Q***	***p***	**ξ [95%-CI]**
Comorbidities[a]	Total	0.97 (0.14)	0.02 (0.05)	**S**	**9.10**	**0.005**	**0.53 [0.45, 0.92]**
	No pain	0.50 (0.18)	0.00 (0.00)	**P**	**32.57**	**0.001**	**0.71 [0.52, 0.84]**
	Chronic Pain	1.62 (0.20)	1.00 (0.27)	S x P	0.11	0.747	
Total symptom burden[b]	Total	8.72 (0.52)	1.37 (0.32)	**S**	**51.77**	**0.001**	**0.70 [0.59, 0.78]**
	No Pain	6.50 (0.67)	0.74 (0.18)	**P**	**53.17**	**0.001**	**0.66 [0.53, 0.79]**
	Chronic Pain	11.50 (0.72)	6.57 (1.16)	S x P	0.35	0.561	
Abdominal complaints	Total	1.03 (0.17)	0.12 (0.11)	S	3.19	0.082	0.45 [0.23, 0.57]
	No Pain	0.83 (0.21)	0.04 (0.06)	**P**	**8.10**	**0.007**	**0.35 [0.16, 0.56]**
	Chronic Pain	1.41 (0.34)	1.14 (0.47)	S x P	0.76	0.388	
Limb pain	Total	2.89 (0.25)	0.35 (0.12)	**S**	**31.91**	**0.001**	**0.68 [0.55, 0.79]**
	No Pain	2.05 (0.27)	0.10 (0.06)	**P**	**60.78**	**0.001**	**0.69 [0.54, 0.81]**
	Chronic Pain	4.24 (0.34)	2.71 (0.46)	S x P	0.48	0.493	
Heart problems	Total	0.63 (0.12)	0.00 (0.00)	**S**	**7.41**	**0.010**	**0.35 [0.29, 0.65]**
	No Pain	0.36 (0.15)	0.00 (0.00)	**P**	**9.30**	**0.004**	**0.47 [0.28, 0.69]**
	Chronic Pain	1.17 (0.25)	0.43 (0.30)	S x P	0.88	0.354	
Exhaustion	Total	2.82 (0.19)	0.47 (0.12)	**S**	**25.05**	**0.001**	**0.65 [0.54, 0.76]**
	No Pain	2.29 (0.26)	0.29 (0.12)	**P**	**19.02**	**0.001**	**0.61 [0.42, 0.75]**
	Chronic Pain	3.51 (0.31)	2.07 (0.57)	S x P	0.68	0.418	

Abbreviations: CI, confidence interval, S, sample, P, pain status.

Significant results are marked in bold.

[a]Self-administered comorbidity questionnaire (SCQ-D), number of problems (range 0 – 13) (Sangha et al., 2003).

[b]Total value of the Giessen Symptom Questionnaire (GBB-8; range: 0 – 32); including the four symptoms exhaustion, abdominal complaints, limb pain and heart problems (range: 0–8) (Kliem et al., 2017).

the clinical sample irrespective of the pain status (shown in Figure 2a). Table 5 displays the frequencies of the different comorbidities in both samples. Specifically, the clinical sample reported more frequently to be suffering from the following diseases: depression, stomach disease, rheumatism and back problems.

Former patients also reported a higher total symptom burden (GBB-8), as well as the elevation of specific symptoms including exhaustion, limb pain and heart problems compared to the community sample (shown in Table 4). Again, group differences in these outcomes manifested between participants with and without chronic pain; no interaction effects between sample and pain status were identified (shown in Table 4 and Figure 2b-f).

Overall, the clinical sample reported significantly higher scores in the screening for anxiety and depression symptoms (PHQ-4) compared to the community sample. These differences specifically occurred due to higher values within the anxiety scale. Groups did not differ regarding reported depression symptoms (shown in Table 6). A total of 24% of the clinical sample and 6% of the community sample reported clinically relevant distress in the PHQ-4 (≥6) ($\chi^2 = 16.6$; $p < .001$). Values within the PHQ-4 and the anxiety subscale were significantly higher in the clinical sample irrespective of pain status (shown in Figure 3). Depression level in this measure did not differ between samples. No interaction effects between sample and pain status were identified for any mental health domain (shown in Table 6).

3.3 | Autonomy and coping

Perceived autonomy did not differ between the clinical and community samples (shown in Table 6). Regarding coping strategies, the clinical sample reported better coping strategies compared to the community sample (shown in Table 6). No differences of coping were found regarding the current presence of chronic pain for the clinical or the community sample.

3.4 | Health care utilisation

The reported hospital inpatient days in the past 12 months did not differ between the clinical and the community samples;

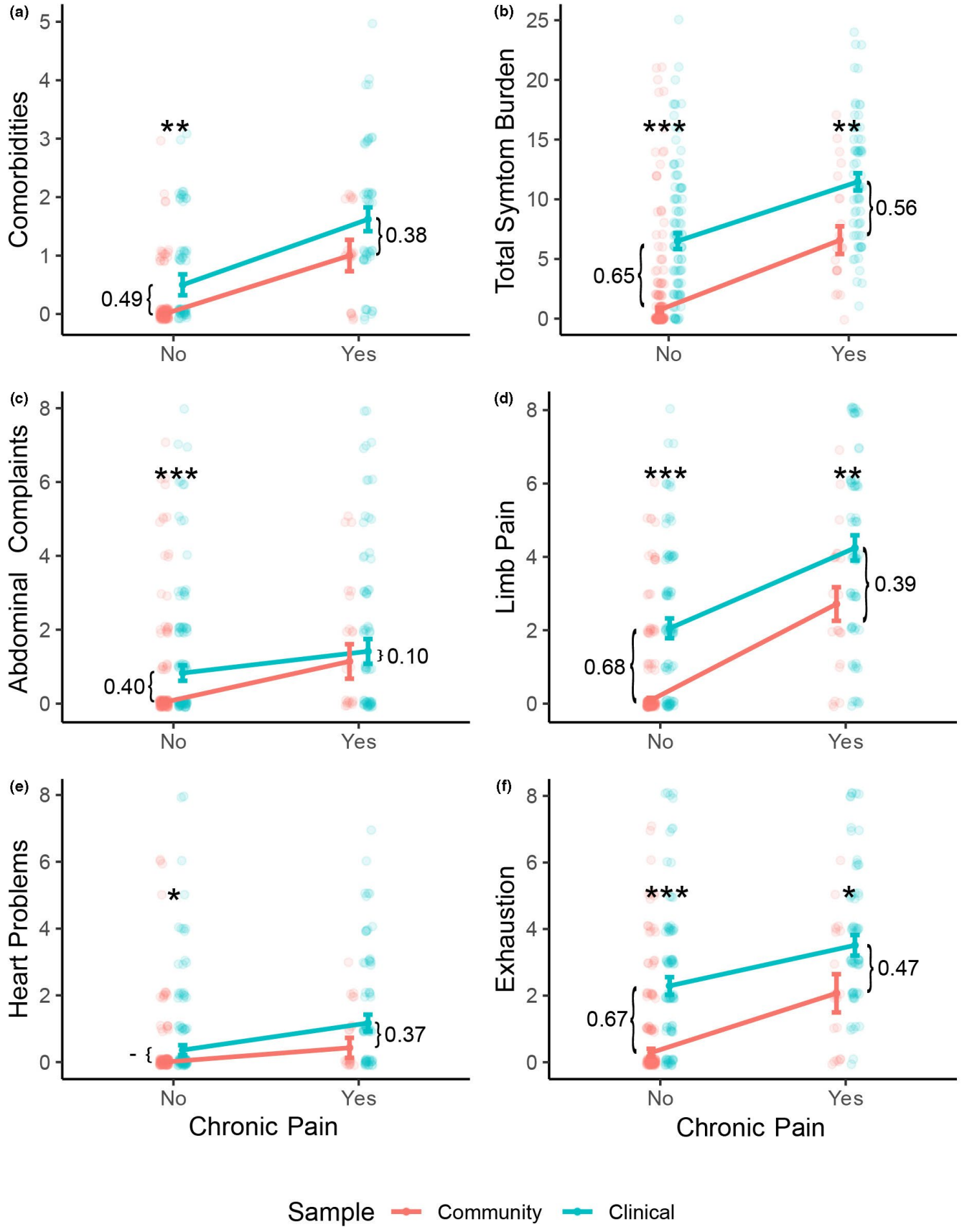

FIGURE 2 Number of comorbidities and degree of symptom burden in clinical and community samples with and without chronic pain. *Note:* Estimates beside brackets indicate the effect size ξ. $*p < 0.05$, $**p < 0.01$, $***p < 0.001$. The number of comorbidities was measured with the Self-administered Comorbidity Questionnaire (SCQ-D) (Sangha et al., 2003). Symptom burden was measured with the Giessen Symptom Questionnaire (GBB-8) (Kliem et al., 2017); total value includes the four symptoms exhaustion, abdominal complaints, limb pain and heart problems

TABLE 5 Presence of physical and mental comorbidities in the clinical and the community samples

SCQ-D Comorbidity	Clinical Sample		Community Sample				
	n	%	*n*	%	X^2	*p*	*V* [95% CI]
Back problems	52	48.6%	24	14.8%	36.28	**<0.001**	**0.37 [0.26, 0.48]**
Depression	28	26.2%	4	2.5%	34.53	**<0.001**	**0.36 [0.25, 0.45]**
Stomach disease	14	13.1%	5	3.1%	9.81	**0.002**	**0.19 [0.07, 0.29]**
Rheumatism	7	6.5%	0	0.0%	10.88	**0.001**[a]	0.20 [–, –][a]
Pulmonary disease	7	6.5%	2	1.2%	5.61	**0.032**[a]	**0.14 [0.03, 0.24]**
Hypertension	4	3.7%	3	1.9%	0.91	0.441[a]	0.06 [0.00, 0.18]
Blood disorder	4	3.7%	1	0.6%	3.44	0.083[a]	0.11 [–, –][a]
Heart disease	3	2.8%	0	0.0%	4.59	0.062[a]	0.13 [–, –][a]
Alcohol or drug abuse	2	1.9%	2	1.2%	0.18	0.651[a]	0.03 [–, –][a]
Diabetes	1	0.9%	4	2.5%	0.83	0.651[a]	0.06 [–, –][a]
Kidney disease	1	0.9%	0	0.0%	1.52	0.398[a]	0.08 [–, –][a]
Liver disease	1	0.9%	0	0.0%	1.52	0.398[a]	0.08 [–, –][a]
Cancer	0	0.0%	0	0.0%	–	–	–

Note:: Significant results are marked in bold. SCQ-D = Self-administered Comorbidity Questionnaire (Sangha et al., 2003).

[a]Bootstrapped confidence intervals not computable as too few participants had this criterion or one cell contained 0 counts.

in fact, mean inpatient days were 0 in both groups. The number of visits to the general practitioner and to specialists was significantly higher in the clinical sample compared to the community sample and in individuals with chronic pain compared to those without chronic pain (shown in Table 7). Regarding the subgroups, the clinical sample contacted their general practitioner ($\xi = 0.64$) and a specialist ($\xi = 0.42$) significantly more frequently compared to the community sample irrespective of whether they currently had chronic pain.

4 | DISCUSSION

The aim of the present study was to analyse the long-term outcomes of paediatric patients seven years after IIPT by comparing former patients to a community sample. At the seven-year follow-up, the majority of former patients were either pain-free (58%) or experienced low intensity and low disability pain (14%), indicating that paediatric chronic patients receiving specialized treatment have a high likelihood of recovering from their chronic disabling pain condition after IIPT. However, results of this study also demonstrated that compared to community members, former patients have poorer physical and mental health status and higher health care utilisation – irrespective of whether or not they have chronic pain at this time point. Concurrently, former patients have better coping strategies.

Promisingly, our study found that almost three-quarters of former patients reported no chronic pain or pain with low intensity and low disability seven years after IIPT. These results are consistent with those of previous IIPT follow-up studies, which report long-term improvements regarding pain intensity, disability and school absence (Banez et al., 2014; Randall et al., 2018; Zernikow et al., 2018). This finding demonstrates that chronic pain may not be a life-long condition and that children in need of IIPT can recover from pain. However, some level of risk appears to remain. In our study, close to 15% experienced disabling chronic pain seven years after IIPT. This proportion is comparable to a US-based five-year follow-up study, which found 28% of former patients continued to experience moderately or severely disabling pain (Randall et al., 2018). In patients with juvenile fibromyalgia, more than half continued to suffer the full range of fibromyalgia symptoms eight years after treatment, indicating that this pain diagnosis poses a higher risk for worse outcomes (Kashikar-Zuck et al., 2019).

Former IIPT patients appear to have poorer physical and mental health compared to the broader community. In our study, former patients reported a higher number of comorbidities and a greater degree of physical and mental health complaints. Regarding mental health specifically, the clinical sample particularly differed from the community sample in self-reported generalized anxiety rather than depression symptoms. In line with this finding, a follow-up study of IIPT paediatric patients found worse anxiety outcomes compared to depression outcomes four years post-treatment (Zernikow et al., 2018). Likewise, a large Norwegian population-based

TABLE 6 Mental health status, autonomy and coping in the clinical and community samples

		M_T (SE_T)[a]					
Measure		**Clinical**	**Community**	**Effect**	***Q***	***p***	**ξ [95%-CI]**
Anxiety/Depression symptoms[b]	Total	3.11 (0.32)	0.87 (0.21)	**S**	**10.56**	**0.002**	**0.51 [0.38, 0.66]**
	No Pain	2.50 (0.36)	0.61 (0.17)	**P**	**19.14**	**0.001**	**0.56 [0.36, 0.74]**
	Pain	4.00 (0.56)	3.00 (0.60)	S x P	1.00	0.323	
Anxiety symptoms[c]	Total	1.75 (0.14)	0.42 (0.11)	**S**	**12.44**	**0.002**	**0.67 [0.46, 0.79]**
	No Pain	1.56 (0.18)	0.28 (0.07)	**P**	**10.94**	**0.003**	**0.48 [0.30, 0.71]**
	Pain	2.00 (0.21)	1.50 (0.44)	S x P	2.37	0.136	
Depression symptoms[d]	Total	1.31 (0.19)	0.38 (0.11)	S	3.67	0.065	0.46 [0.29, 0.62]
	No Pain	0.92 (0.19)	0.25 (0.12)	**P**	**12.56**	**0.002**	**0.51 [0.34, 0.77]**
	Pain	1.97 (0.30)	1.43 (0.53)	S x P	0.04	0.835	
Autonomy[e]	Total	3.47 (0.06)	3.50 (0.05)	S	0.23	0.635	0.04 [0.00, 0.19]
	No Pain	3.50 (0.08)	3.50 (0.06)	P	0.23	0.635	0.07 [0.00, 0.28]
	Pain	3.41 (0.12)	3.50 (0.08)	S x P	0.26	0.611	
Coping[f]	Total	16.20 (0.37)	17.70 (0.26)	**S**	**6.40**	**0.016**	**0.34 [0.17, 0.48]**
	No Pain	16.70 (0.43)	14.70 (0.28)	P	1.00	0.326	0.05 [0.00, 0.28]
	Pain	15.6 (0.57)	14.70 (0.88)	S x P	1.07	0.308	

Abbreviations: CI, confidence interval, P, pain status S, sample.

Significant results are marked in bold.

[a]20% trimmed means (20% trimmed standard deviations).

[b]Patient Health Questionnaire (PHQ-4); values range from 0 to 12, higher values indicate more symptoms (Löwe et al., 2010).

[c]2-item anxiety scale (GAD-2); values range from 0 to 6 (Löwe et al., 2010).

[d]2-item depression scale (PHQ-2); values range from 0 to 6 (Löwe et al., 2010).

[e]Perceived Autonomy (Warner et al., 2011); sum score ranging from 4 to 16.

[f]4-item Brief Resilient and Coping Scale (BRCS); sum score ranging from 4 to 20 (Kocalevent et al., 2017).

study also reported a higher risk of anxiety disorders, compared to mood disorders, in young adults who experienced musculoskeletal pain during their teens (Eckhoff et al., 2017). However, the clinical sample in our study reported the presence of comorbid depression more often compared to the community. Taken together, these findings suggest a generally reduced mental health status in former pain patients.

Several explanations may account for these findings. Severe childhood pain may have a negative long-term effect on mental health outcomes. This effect may be particularly pertinent to certain subpopulations. Kashikar-Zuck and colleagues (2019) found different trajectories of emotional distress after IIPT in juvenile fibromyalgia patients. While some patients experienced an improvement in depression symptoms, a similar number of patients experienced worsening of symptoms. However, this study did not control for the patients' pain status at follow-up; therefore, this work does not provide insights into whether different trajectories may occur independent of the pain status. Studies monitoring chronic pain and mental health outcomes after treatment consistently show that anxiety and depression decrease simultaneously with the improvement of pain characteristics (Benore et al., 2015; Hechler et al., 2014; Hirschfeld et al., 2013). This supports the hypothesis of mutual maintenance rather than a negative long-term effect of pain (Asmundson & Katz, 2009; Soltani et al., 2019).

A general vulnerability to mental/psychosomatic disorders may provide a second explanation, in that individuals are more likely to experience chronic pain but also other mental conditions during the course of their lives. This has been proposed for different mental health conditions (Copeland et al., 2009, 2013; Gundel et al., 2018; Steinhausen, 2013). For chronic pain specifically, some preliminary evidence on two-sided causality exists. Evidence suggests that negative emotional symptoms are risk factors for the onset of chronic pain (Huguet et al., ,2016, 2017) and that chronic pain predicts mental disorders (Fearon & Hotopf, 2001; Shelby et al., 2013). While a satisfactory explanation of the cause of higher mental distress in former patients cannot be provided based on current research, a clear clinical implication can be drawn. In order to improve the mental health status of former patients long-term, better aftercare to treat mental comorbidities as well as regular checkups, even if the patient is pain-free, are indicated.

In contrast to the physical and mental health status, coping strategies were significantly better in the clinical sample compared to the community sample, indicating that former

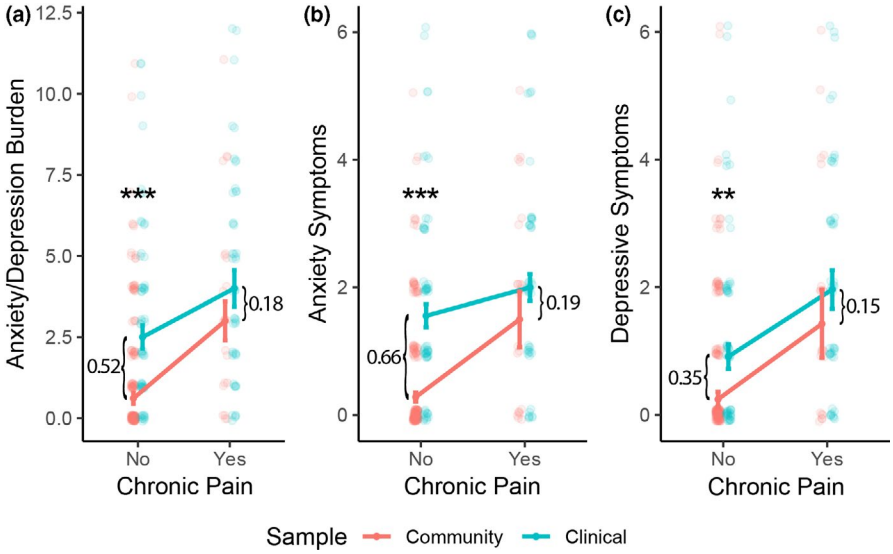

FIGURE 3 Mental health symptoms in the clinical and community samples with and without chronic pain. *Note:* Estimates beside brackets indicate the effect size ξ. $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$. Anxiety and depression symptoms were measured with the Patient Health Questionnaire (PHQ-4) (Löwe et al., 2010)

TABLE 7 Number of visits to health care providers in the clinical and community samples

	M_T (SE_T)					
Health care provider	**Clinical**	**Community**	**Effect**	***Q***	***p***	**ξ [95%-CI]**
General Practitioner	4.93 (0.41)	2.27 (0.15)	**S**	**9.70**	**0.004**	**0.75 [0.53, 0.79]**
No Pain	4.07 (0.32)	2.08 (0.12)	**P**	**17.87**	**0.001**	**0.63 [0.42, 0.79]**
Pain	6.70 (0.82)	4.77 (0.91)	S x P	0.002	0.966	
Psychotherapist	1.18 (0.16)	1.00 (0.00)	S	1.27	0.266	–[a]
No Pain	1.03 (0.14)	1.00 (0.00)	P	0.99	0.326	–[a]
Pain	1.55 (0.50)	1.00 (0.00)	S x P	0.99	0.326	
Specialist	1.00 (0.26)	1.38 (0.11)	**S**	**13.25**	**0.001**	**0.46 [0.35, 0.62]**
No Pain	2.50 (0.35)	1.34 (0.12)	**P**	**4.21**	**0.049**	**0.45 [0.23, 0.65]**
Pain	3.73 (0.40)	1.83 (0.64)	S x P	0.78	0.386	

Abbreviations: CI, confidence interval, P, pain status, S, sample.

Significant results are marked in bold.

[a]not computable as one *SD* is equal to 0.

patients may have better strategies for dealing with difficult situations. IIPT has a strong cognitive behavioural therapy focus; patients learn different behavioural and cognitive techniques for pain management (Dobe & Zernikow, 2019). Furthermore, many patients seek recommended outpatient psychotherapy after IIPT, in which they may learn further cognitive or behavioural coping techniques (Hechler et al., 2014). The clinical sample may transfer the acquired techniques to other situations and challenges, even years after treatment. These strategies may be important resilience factors in former patients. If these were less developed, former patients might have worse health outcomes than those reported here.

From several studies, it is well known that children with chronic pain frequently access a variety of health care services (Groenewald et al., 2014; Könning, Rosenthal, Friese, et al., 2021; Ruhe et al., 2013; Toliver-Sokol et al., 2011). Following IIPT, health care utilisation decreases (Evans et al., 2016; Zernikow et al., 2018), particularly in patients with an overall improvement in their chronic pain condition (Zernikow et al., 2018). However, in this study, even former patients who no longer experienced chronic pain visited their general practitioner and specialists more often than the community sample, indicating that reasons other than pain may be triggering physician consultations. The higher level of emotional distress in the clinical sample may be cause for greater health care utilisation (Benuto et al., 2020).

The results of this study need to be interpreted with consideration to some limitations. First, our comparison group consisted of data from a representative community sample that were collected separately from our study (Häuser et al., 2014). Therefore, only the predefined questionnaires could be selected for the follow-up survey of the clinical sample. Three questionnaires were only validated for adults.

However, all questionnaires were also applied to adolescents younger than 18 years ($n = 9$ in each sample). Second, the questionnaires only referenced the prior year or, in some items, the past four weeks. Longitudinal information regarding patients' health status and health care utilisation were not assessed. Third, information contained in the self-administered comorbidity questionnaire was not verified. It is unclear if diagnoses indicated by participants were based on an assessment by a physician or if it was self-diagnosed by the study participants. Fourth, data of the community sample were collected about 5 years earlier than data in the clinical sample. Therefore, time effects cannot be fully ruled out. Last, the participation rate for this study was 71% for the telephone interview and 47% for the additional online survey. These numbers are comparable to prior long-term studies with retention rates between 46% and 74% (Banez et al., 2014; Kashikar-Zuck et al., 2019; Randall et al., 2018; Zernikow et al., 2018). Furthermore, dropout analyses did not identify any group differences at admission.

5 | CONCLUSION

Our study suggests that patients experiencing severe chronic pain in childhood who engage in IIPT are likely to recover from their pain condition by early adulthood. Long-term treatment effects manifest as better coping strategies. However, former patients' mental and physical health statuses are reduced compared to a community sample. This indicates either the negative long-term effect of early chronic pain experiences or a vulnerability to experiencing mental and physical comorbidities in individuals who develop a chronic pain condition during childhood. Future longitudinal research should attempt to parse out the bidirectional effects between chronic pain and emotional distress amongst youth across time.

ACKNOWLEDGEMENT

We would like to thank Donnamay Brown and Alexandra van der Valk for editing the manuscript for English language.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Study conception and design included JW, AR, LS and BZ. LS and KL collected clinical sample data; WH, EB, AD and RK collated the community-sample data. BBC, JW and AR conducted the data analysis, and JW, AR and LS contributed to drafting the manuscript. All authors reviewed the manuscript and provided important intellectual content. All authors have significantly contributed and approved the final manuscript.

REFERENCES

Asmundson, G. J., & Katz, J. (2009). Understanding the co-occurrence of anxiety disorders and chronic pain: State-of-the-art. *Depression and Anxiety*, *26*, 888–901. https://doi.org/10.1002/da.20600

Banez, G. A., Frazier, T. W., Wojtowicz, A. A., Buchannan, K., Henry, D. E., & Benore, E. (2014). Chronic pain in children and adolescents: 24–42 month outcomes of an inpatient/day hospital interdisciplinary pain rehabilitation program. *J Pediatr Rehabil Med*, *7*, 197–206. https://doi.org/10.3233/PRM-140289

Benore, E., D'Auria, A., Banez, G. A., Worley, S., & Tang, A. (2015). The influence of anxiety reduction on clinical response to pediatric chronic pain rehabilitation. *Clinical Journal of Pain*, *31*, 375–383. https://doi.org/10.1097/AJP.0000000000000127

Benuto, L. T., Casas, J., Gonzalez, F., & Newlands, R. (2020). The behavioral model of health: Education, behavioral health factors, and stigma as predictors of help-seeking attitudes. *Community Mental Health Journal*, *56*(7), 1275–1283. https://doi.org/10.1007/s10597-020-00601-y

Bergmann, E. K. P. (1999). Use of medical services. *Gesundheitswesen*, *61*, S138–S144.

Brattberg, G. (2004). Do pain problems in young school children persist into early adulthood? A 13-year follow-up. *European Journal of Pain*, *8*, 187–199. https://doi.org/10.1016/j.ejpain.2003.08.001

Brna, P., Dooley, J., Gordon, K., & Dewan, T. (2005). The prognosis of childhood headache: A 20-year follow-up. *Archives of Pediatrics and Adolescent Medicine*, *159*, 1157–1160. https://doi.org/10.1001/archpedi.159.12.1157

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences*. Lawrence Earlbaum Associates.

Copeland, W. E., Adair, C. E., Smetanin, P., Stiff, D., Briante, C., Colman, I., Fergusson, D., Horwood, J., Poulton, R., Jane Costello, E., & Angold, A. (2013). Diagnostic transitions from childhood to adolescence to early adulthood. *Journal of Child Psychology and Psychiatry*, *54*, 791–799. https://doi.org/10.1111/jcpp.12062

Copeland, W. E., Shanahan, L., Costello, E. J., & Angold, A. (2009). Childhood and adolescent psychiatric disorders as predictors of young adult disorders. *Archives of General Psychiatry*, *66*, 764–772. https://doi.org/10.1001/archgenpsychiatry.2009.85

Dobe, M., & Zernikow, B. (2019). *Practical treatment options for chronic pain in children and adolescents: An interdisciplinary therapy manual*. Springer.

Eckhoff, C., Straume, B., & Kvernmo, S. (2017). Multisite musculoskeletal pain in adolescence and later mental health disorders: A population-based registry study of Norwegian youth: The NAAHS cohort study. *British Medical Journal Open*, *7*, e012035.–https://doi.org/10.1136/bmjopen-2016-012035

Evans, J. R., Benore, E., & Banez, G. A. (2016). The cost-effectiveness of intensive interdisciplinary pediatric chronic pain rehabilitation. *Journal of Pediatric Psychology*, *41*, 849–856. https://doi.org/10.1093/jpepsy/jsv100

Fearon, P., & Hotopf, M. (2001). Relation between headache in childhood and physical and psychiatric symptoms in adulthood: National birth cohort study. *BMJ*, *322*, 1145. https://doi.org/10.1136/bmj.322.7295.1145

Groenewald, C. B., Essner, B. S., Wright, D., Fesinmeyer, M. D., & Palermo, T. M. (2014). The economic costs of chronic pain among a cohort of treatment-seeking adolescents in the United States. *The Journal of Pain*, *15*, 925–933. https://doi.org/10.1016/j.jpain.2014.06.002

Gundel, L. K., Pedersen, C. B., Munk-Olsen, T., & Dalsgaard, S. (2018). Longitudinal association between mental disorders in childhood and subsequent depression–A nationwide prospective cohort study. *Journal of Affective Disorders*, *227*, 56–64. https://doi.org/10.1016/j.jad.2017.10.023

Hansen, B. B., Fredrickson, M., Buckner, J., Errickson, J., Rauh, A., & Solenberger, P. (2019). R Package 'optmatch' version 0.9-11.

Hansen, B. B., & Klopfer, S. O. (2006). Optimal full matching and related designs via network flows. *Journal of Computational and Graphical Statistics*, *15*, 609–627. https://doi.org/10.1198/106186006X137047

Häuser, W., Wolfe, F., Henningsen, P., Schmutzer, G., Brähler, E., & Hinz, A. (2014). Untying chronic pain: Prevalence and societal burden of chronic pain stages in the general population - a cross-sectional survey. *BMC Public Health*, *14*, 352. https://doi.org/10.1186/1471-2458-14-352

Hechler, T., Kanstrup, M., Holley, A. L., Simons, L. E., Wicksell, R., Hirschfeld, G., & Zernikow, B. (2015). Systematic Review on Intensive Interdisciplinary Pain Treatment of Children With Chronic Pain. *Pediatrics*, *136*, 115–127. https://doi.org/10.1542/peds.2014-3319

Hechler, T., Ruhe, A., Schmidt, P., Hirsch, J., Wager, J., Dobe, M., Krummenauer, F., & Zernikow, B. (2014). Inpatient-based intensive interdisciplinary pain treatment for highly impaired children with severe chronic pain: Randomized controlled trial of efficacy and economic effects. *Pain*, *155*, 118–128. https://doi.org/10.1016/j.pain.2013.09.015

Hestbaek, L., Leboeuf-Yde, C., Kyvik, K. O., & Manniche, C. (2006). The course of low back pain from adolescence to adulthood: Eight-year follow-up of 9600 twins. *Spine*, *31*, 468–472. https://doi.org/10.1097/01.brs.0000199958.04073.d9

Hirschfeld, G., Hechler, T., Dobe, M., Wager, J., von Lützau, P., Blankenburg, M., Kosfelder, J., & Zernikow, B. (2013). Maintaining lasting improvements: One-year follow-up of children with severe chronic pain undergoing multimodal inpatient treatment. *Journal of Pediatric Psychology*, *38*, 224–236. https://doi.org/10.1093/jpepsy/jss115

Ho, D. E., Imai, K., King, G., & Stuart, E. A. (2011). Matchit: Nonparametric preprocessing for parametric causal inference. *Journal of Statistical Software*, *42*(8). https://doi.org/10.18637/jss.v042.i08

Huguet, A., & Miró, J. (2008). The severity of chronic pediatric pain: An epidemiological study. *The Journal of Pain*, *9*, 226–236. https://doi.org/10.1016/j.jpain.2007.10.015

Huguet, A., Olthuis, J., McGrath, P., Tougas, M., Hayden, J., Stinson, J., & Chambers, C. (2017). Systematic review of childhood and adolescent risk and prognostic factors for persistent abdominal pain. *Acta Paediatrica*, *106*, 545–553. https://doi.org/10.1111/apa.13736

Huguet, A., Tougas, M. E., Hayden, J., McGrath, P. J., Stinson, J. N., & Chambers, C. T. (2016). Systematic review with meta-analysis of childhood and adolescent risk and prognostic factors for musculoskeletal pain. *Pain*, *157*, 2640–2656. https://doi.org/10.1097/j.pain.0000000000000685

Imai, K. (2018). MatchIt. R package version 3.0.2..

Kashikar-Zuck, S., Cunningham, N., Peugh, J., Black, W. R., Nelson, S., Lynch-Jordan, A. M., Pfeiffer, M., Tran, S. T., Ting, T. V., Arnold, L. M., Carle, A., Noll, J., Powers, S. W., & Lovell, D. J. (2019). Long-term outcomes of adolescents with juvenile-onset fibromyalgia into adulthood and impact of depressive symptoms on functioning over time. *Pain*, *160*, 433–441. https://doi.org/10.1097/j.pain.0000000000001415

Kaufman, E. L., Tress, J., & Sherry, D. D. (2016). Trends in medicalization of children with amplified musculoskeletal pain syndrome. *Pain Medicine*, pnw188. https://doi.org/10.1093/pm/pnw188

Klasen, B. W., Hallner, D., Schaub, C., Willburger, R., & Hasenbring, M. (2004). Validation and reliability of the German version of the Chronic Pain Grade questionnaire in primary care back pain patients. *Psychosoc Medicine*, *1*, 1–12.

Kliem, S., Lohmann, A., Klatt, T., Mößle, T., Rehbein, F., Hinz, A., Beutel, M., & Brähler, E. (2017). Brief assessment of subjective health complaints: Development, validation and population norms of a brief form of the Giessen Subjective Complaints List (GBB-8). *Journal of Psychosomatic Research*, *95*, 33–43. https://doi.org/10.1016/j.jpsychores.2017.02.003

Kocalevent, R. D., Zenger, M., Hinz, A., Klapp, B., & Brahler, E. (2017). Resilient coping in the general population: Standardization of the brief resilient coping scale (BRCS). *Health and Quality of Life Outcomes*, *15*, 251., https://doi.org/10.1186/s12955-017-0822-6

Könning, A., Rosenthal, N., Brown, D., Stahlschmidt, L., & Wager, J. (2021). Severity of chronic pain in german adolescent school students. *The Clinical Journal of Pain*, *37*(2), 118–125. https://doi.org/10.1097/ajp.0000000000000898

Könning, A., Rosenthal, N., Friese, M., Hirschfeld, G., Brown, D., & Wager, J. (2021). Factors associated with physician consultation and medication use in children and adolescents with chronic pain: A scoping review and original data. *European Journal of Pain*, *25*(1), 88–106. https://doi.org/10.1002/ejp.1661

Löwe, B., Wahl, I., Rose, M., Spitzer, C., Glaesmer, H., Wingenfeld, K., Schneider, A., & Brähler, E. (2010). A 4-item measure of depression and anxiety: Validation and standardization of the Patient Health Questionnaire-4 (PHQ-4) in the general population. *Journal of Affective Disorders*, *122*, 86–95. https://doi.org/10.1016/j.jad.2009.06.019

Mair, P., & Wilcox, R. R. (2019). Robust statistical methods in R using the WRS2 package. *Behavior Research Methods*, *52*(2), 464–488. https://doi.org/10.3758/s13428-019-01246-w

R Core Team.R (2019). *A language and environment for statistical computing*. : R Foundation for Statistical Computing (Version 3.6.1).

Randall, E. T., Smith, K. R., Conroy, C., Smith, A. M., Sethna, N., & Logan, D. E. (2018). Back to living: Long-term functional status of pediatric patients who completed intensive interdisciplinary pain treatment. *Clinical Journal of Pain*, *34*, 890–899. https://doi.org/10.1097/AJP.0000000000000616

Ruhe, A., Wager, J., Schmidt, P., & Zernikow, B. (2013). Economic effects of chronic pain in childhood and adolescence: self-assessment of health care costs for affected families before and after a multidisciplinary inpatient pain therapy. *Schmerz*, *27*, 577–587.

Sangha, O., Stucki, G., Liang, M. H., Fossel, A. H., & Katz, J. N. (2003). The Self-Administered Comorbidity Questionnaire: A new method to assess comorbidity for clinical and health services research. *Arthritis and Rheumatism*, *49*, 156–163. https://doi.org/10.1002/art.10993

Shelby, G. D., Shirkey, K. C., Sherman, A. L., Beck, J. E., Haman, K., Shears, A. R., Horst, S. N., Smith, C. A., Garber, J., & Walker, L. S. (2013). Functional abdominal pain in childhood and long-term vulnerability to anxiety disorders. *Pediatrics*, *132*, 475–482. https://doi.org/10.1542/peds.2012-2191

Simons, L. E., Sieberg, C. B., Pielech, M., Conroy, C., & Logan, D. E. (2013). What does it take? Comparing intensive rehabilitation to outpatient treatment for children with significant pain-related disability. *Journal of Pediatric Psychology*, *38*, 213–223. https://doi.org/10.1093/jpepsy/jss109

Sinclair, V. G., & Wallston, K. A. (2004). The development and psychometric evaluation of the Brief Resilient Coping Scale. *Assessment*, *11*, 94–101. https://doi.org/10.1177/1073191103258144

Soltani, S., Kopala-Sibley, D. C., & Noel, M. (2019). The Co-occurrence of Pediatric Chronic Pain and Depression. *Clinical Journal of Pain*, *35*, 633–643. https://doi.org/10.1097/AJP.0000000000000723

Stahlschmidt, L., Zernikow, B., & Wager, J. (2016). Specialized rehabilitation programs for children and adolescents with severe disabling chronic pain: Indications, treatment and outcomes. *Children*, *3*, 33.– https://doi.org/10.3390/children3040033

Steinhausen, H. C. (2013). What happens to children and adolescents with mental disorders? Findings from long-term outcome research. *Zeitschrift Fur Kinder- Und Jugendpsychiatrie Und Psychotherapie*, *41*, 419–431.

Streibelt, M., Schmidt, C., Brunger, M., Spyra, K. (2012). Comorbidity from the patient perspective - does it work? Validity of a questionnaire on self-estimation of comorbidity (SCQ-D). *Orthopäde*, *41*, 303–310.

Toliver-Sokol, M., Murray, C. B., Wilson, A. C., Lewandowski, A., & Palermo, T. M. (2011). Patterns and predictors of health service utilization in adolescents with pain: Comparison between a community and a clinical pain sample. *The Journal of Pain*, *12*, 747–755. https://doi.org/10.1016/j.jpain.2010.12.011

Wager, J., Hechler, T., Darlington, A., Hirschfeld, G., Vocks, S., & Zernikow, B. (2013). Classifying the severity of paediatric chronic pain–an application of the chronic pain grading. *European Journal of Pain*, *17*, 1393–1402. https://doi.org/10.1002/j.1532-2149.2013.00314.x

Wager, J., Szybalski, K., Schenk, S., Frosch, M., & Zernikow, B. (2019). Predictors of treatment outcome in children with medically unexplained pain seeking primary care: A prospective cohort study. *European Journal of Pain*, *23*, 1507–1518. https://doi.org/10.1002/ejp.1426

Walker, L. S., Dengler-Crish, C. M., Rippel, S., & Bruehl, S. (2010). Functional abdominal pain in childhood and adolescence increases risk for chronic pain in adulthood. *Pain*, *150*, 568–572. https://doi.org/10.1016/j.pain.2010.06.018

Warner, L. M., Ziegelmann, J. P., Schuz, B., Wurm, S., Tesch-Romer, C., & Schwarzer, R. (2011). Maintaining autonomy despite multimorbidity: Self-efficacy and the two faces of social support. *European Journal of Ageing*, *8*, 3–12. https://doi.org/10.1007/s10433-011-0176-6

Wilcox, R. R. (2012). Introduction to robust estimation and hypothesis testing Elsvier.

Yuen, K. K. (1974). The two-sample trimmed t for unequal population variances. *Biometrika*, *61*, 165–170.

Zernikow, B., Ruhe, A., Stahlschmidt, L., Schmidt, P., Staratzke, T., Frosch, M., & Wager, J. (2018). Clinical and economic long-term treatment outcome of children and adolescents with disabling chronic pain. *Pain Medicine*, *19*, 16–28. https://doi.org/10.1093/pm/pnx067

Zernikow, B., Wager, J., Hechler, T., Hasan, C., Rohr, U., Dobe, M., Meyer, A., Hübner-Möhler, B., Wamsler, C., & Blankenburg, M. (2012). Characteristics of highly impaired children with severe chronic pain: A 5-year retrospective study on 2249 pediatric pain patients. *BMC Pediatrics*, *16*, 54–65. https://doi.org/10.1186/1471-2431-12-54

How to cite this article: Wager J, Ruhe A-K, Stahlschmidt L, et al. Long-term outcomes of children with severe chronic pain: Comparison of former patients with a community sample. *Eur J Pain*. 2021;25:1329–1341. https://doi.org/10.1002/ejp.1754