Reliability Study

Reporting the Location and Extent of Pain in Adolescents: A Test-Retest Reliability Study

Anna Folli, BSc[1,2], Deborah Falla, PhD[1], Corrado Cescon, PhD[2], Federica Vanoni, MD, PhD[3,4], Emiliano Soldini, MSc[5], and Marco Barbero, PhD[2]

From: [1]Centre of Precision Rehabilitation for Spinal Pain (CPR Spine), School of Sport, Exercise and Rehabilitation Sciences, College of Life and Environmental Sciences, University of Birmingham, Birmingham, United Kingdom; [2]Rehabilitation Research Laboratory 2rLab, Department of Business Economics, Health and Social Care, University of Applied Sciences and Arts of Southern Switzerland, Manno, Switzerland; [3]Institute of Pediatrics of Southern Switzerland, EOC, Bellinzona, Switzerland; [4]Faculty of Biomedical Sciences, Università della Svizzera italiana, Lugano, Switzerland; [5]Competence Centre for Healthcare Practices and Policies, Department of Business Economics, Health and Social Care, University of Applied Sciences and Arts of Southern Switzerland, Manno, Switzerland

Address Correspondence:
Anna Folli ,BSc
Rehabilitation Research Laboratory 2rLab, Department of Business Economics, Health and Social Care, University of Applied Sciences and Arts of Southern Switzerland
Via Violino 11
6928 Manno, Switzerland
E-mail: anna.folli@supsi.ch

Disclaimer: This study was supported by the Thim van der Laan Foundation (Landquart, Switzerland) and by the University of Applied Sciences and Arts of Southern Switzerland (Manno, Switzerland).

Conflict of interest: Each author certifies that he or she, or a member of his or her immediate family, has no commercial association (i.e., consultancies, stock ownership, equity interest, patent/licensing arrangements, etc.) that might pose a conflict of interest in connection with the submitted article.

Article received: 12-01-2023
Revised article received: 01-03-2024
Accepted for publication: 03-12-2024

Free full article: www.painphysicianjournal.com

Background: A pain drawing is a self-administered assessment that requires the patient to shade in on a body chart the areas in which he or she experiences pain, regardless of the intensity. Pain drawings have already been validated in several adult populations.

Objectives: The aim of this study is to establish adolescents' test-retest reliability in reporting the extent and location of their pain using a paper-based pain drawing.

Study Design: A one-day test-retest reliability study was set up.

Setting: The study took place in 2 separate locations—a pediatric hospital and a private physiotherapy practice in Ticino, in the southern part of Switzerland. This reliability study was approved by the local ethics committee of Ticino (2021-00492 CE 3832).

Methods: Adolescents with musculoskeletal pain (aged 11-16 years) were included. All participants were asked to shade the areas in which they experienced pain over the previous week. After the administration of a questionnaire and the acquisition of further personal data, the pain drawing was administered again. The pain drawings were then scanned and analyzed using a digital platform, which allowed the extraction of pain extent and location values. The test-retest reliability was evaluated on these data. The intraclass correlation coefficient and Bland-Altman analysis were used to assess the reliability of the reporting of the pain extent, whereas the Jaccard similarity coefficient was used to calculate the reliability of the reporting of the pain location.

Results: The reporting of the pain extent was observed to have excellent test-retest reliability: $ICC_{2,1}$: 0.959 (95% CI: 0.925-0.978). The Bland-Altman analysis showed a mean difference close to 0: -0.010% (limits of agreements -0.962 to 0.942). The reliability of the reporting of pain location was also supported by the Jaccard index mean score of 0.82 (± 0.19).

Limitations: Reliability of reporting may vary depending on the nature of the pain, its duration, or the type of disorder and body areas involved.

Conclusions: Adolescents complaining musculoskeletal pain showed reliability in reporting pain extent and location using pain drawings.

Key words: Pain drawing, reliability, adolescents, pain, evaluation, body chart, pain extent, pain location

Pain Physician 2024: 27:E637-E643

Chronic and recurrent pain is common in childhood and adolescence. The prevalence of back pain in children has been estimated to be between 9% and 25%, and musculoskeletal and/or limb pain is present in 9% to 56% of children (1). According to some authors, chronicity rates in adolescents, like those found in adulthood, suggest that some of these disorders may be related to the development of chronic symptoms as early as adolescence (1).

The pain drawing (PD) is a self-administered assessment tool initially designed by Harold Palmer to distinguish "psychogenic pain" from "organic pain" (2). This measure consists of coloring in on a body chart (BC) (i.e., a stylized representation of the human body) all the areas in which the patient has experienced pain during a specific recall period, regardless of the pain's intensity. The PD can be administered using a pen-on-paper approach or digital tools (e.g., a tablet). Subsequently, the PD can be analyzed either qualitatively through visual inspection or quantitatively by extracting metrics such as pain extent (size) and pain location (somatic distribution). Clinicians can use these data to assess the clinical course of patients' symptoms or to evaluate their responses to provocative tests (3).

Patients' reliability in reporting their pain on a BC remains an underexplored aspect of the PD method, especially in specific populations, such as in adolescents. Studies that have been published on this topic emphasize that healthy people (4), patients with different pain conditions, such as low back pain, neck pain, nononcological pain (5-9), whiplash-associated disorders (10), temporomandibular disorders (11), neuropathic pain (12), and primary dysmenorrhea (13), and institutionalised elderly people (14) are reliable in reporting their pain. To the best of our knowledge, only Foxen-Craft et al (6) have focused their study on young patients, confirming the reliability and validity of body maps for assessing pain location and widespread pain in people aged 10 to 17 years. The method proposed by these authors involves analyzing pain maps by extracting the total number of pain sites without considering the pain extent or location. Thus, although BCs are included in several questionnaires for the assessment of pain experienced by children and adolescents (15), no one has investigated adolescents' reliability in reporting the extent or location of their pain.

Objectives

The purpose of this study is to determine whether adolescents who are given a PD to report the extent and location of their pain are reliable in doing so. Ensuring that adolescents are reliable in reporting their pain with this method would support the use of PDs for assessing pain location and extent in this population.

Methods

Study Design

This reliability study was approved by the local ethics committee of Ticino (2021-00492 CE 3832), and all patients and parents were informed and signed the informed consent form. The information and signing of the informed consent followed the Swissethics guidelines for research on children and adolescents (16). The reporting in this study adheres to the Guidelines for Reporting Reliability and Agreement Studies (17).

Participants

Adolescents between 11 and 16 years of age who had musculoskeletal pain, were assessed by a pediatrician, and, if necessary, referred for rehabilitation, were included in the study. Exclusion criteria were disturbances in visuospatial perception, problems with the function of the dominant hand, and inability to give consent for the study.

Procedures

Data collection was carried out in 2 centers: a regional children's hospital immediately after the orthopedic doctor's visit and a physiotherapy practice before one of the planned treatment sessions. In a separate room that provided an adequate setting in terms of privacy and comfort, patients were presented with paper BCs tailored to their genders, featuring both frontal and dorsal views of the body. The BCs were generically adapted to adolescent bodies (less marked musculature and less developed sexual characteristics than adults). The type, color, and size of the marker tip used to shade the locations of the patients' pain were standardized (red, 0.8 mm). Standardized verbal explanations of what a PD was and how to complete it were provided. The instructions emphasized the importance of coloring all areas where pain was experienced, regardless of its intensity or type. A physiotherapist (AF) demonstrated on a test PD how to color the areas affected by pain and which markings to avoid (e.g., dots, X's, or circles). Following the explanation, the patients were allowed to practice on a test BC to become familiar with the process. After this familiarization, patients were asked to complete the

first PD (PD1). The verbal instructions were standardized as follows: "Please shade in on this body chart the pain you have experienced during the last week. Color all the areas in which you experienced pain, regardless of how strong it was. Try to be as precise as possible." Once the first PDs were completed, each patient's age, gender, diagnosis, dominant hand, weight, and height were acquired. Then, patients were presented with a questionnaire consisting of the VAS, the Italian version of the pain catastrophizing scale (18), and the "physical functioning" section of the Bath Adolescent Pain Questionnaire (BAPQ) (19). Data acquisition and the completion of the questionnaire took approximately 20 minutes. This time was considered to allow for a good balance between recall bias and the possibility of pain fluctuation. Immediately afterward, patients were asked to complete a second PD (PD2). The verbal instructions were the same, and the patients did not know until then that they would have to complete 2 PDs. After they completed each PD, the patients were asked whether they were satisfied with it. If they were unsatisfied, they were given the opportunity to finish their drawings.

The PDs were then scanned and analyzed using a digital platform, which allowed the extraction of pain extent and location.

Statistical Analysis

The sample size was calculated according to Walter et al (20) by setting the following parameters: 2 replicates, $\rho 0 = 0.6$, $\rho 1 = 0.8$, $\alpha = 0.05$, and $\beta = 0.2$. The minimum sample size was 39.1. Therefore, 40 participants were recruited.

Adolescents' test-retest reliability in reporting pain extent was examined using the intraclass correlation coefficient ($ICC^{2,1}$). The criteria used for interpretation of the ICCs were those proposed by Koo and Li (21): < 0.5: poor reliability; 0.5-0.75: moderate reliability; 0.75-0.9: good reliability; > 0.90: excellent reliability. Bland-Altman plots were provided to give a visual representation of systematic biases, outliers, and the overall level of agreement between 2 consecutive PDs completed by the same patient.

Pain location test–retest reliability was calculated using the Jaccard similarity coefficient, analyzing the overlap of pain areas (22). The index is represented by the ratio of the number of "anatomical regions" colored in both BCs (PD1 $\cap$ PD2) to the total number of colored-in "anatomical regions" (PD1 $\cup$ PD2), and that index ranges from 0 (the PDs' areas do not overlap) to one (the PDs' areas overlap completely). The supplementary material presents an illustration and description of the anatomical areas according to which the pain location analysis was carried out.

Results

The study was conducted over the course of 2 years. A total of 42 adolescents were included in the study. Two of the patients were excluded after data collection and before the data were analyzed, due to parental interference in the PDs' completion.

The patients completed 2 PDs, each PD consisting of a dorsal and a ventral part. In total, 160 images were analyzed. The mean (± SD) age, height, weight, and BMI of the patients were 13.4 (± 1.6) years old, 163 (± 9) cm, 57.1 (± 12.9) kg, and 21.4 (± 3.8) kg/m^2, respectively. Table 1 reports the demographic and clinical features of the sample.

The areas affected by pain included mainly the knees and the spine. Figure 1 represents the pain distribution divided by gender.

The comparisons between the colored areas in each patients' first and second tests are depicted in Fig. 2.

Test-retest reliability was excellent, with an $ICC^{2,1}$ of 0.959 (95% CI 0.925-0.978). The Bland-Altman analysis (Fig. 3) showed a mean difference close to 0: -0.010 (limits of agreements -0.962 to 0.942).

Table 1. *Demographic and clinical features of the total sample. Results are expressed as mean ± SD.*

	N	Age (years)	Height (cm)	Weight (kg)	BMI (kg/m^2)	Pain Intensity (VAS cm)	Pain Extent PD1 (%)	Pain Extent PD2 (%)	Pain Catastrophizing	Physical Functioning (BAPQ)
Girls	20	13.8 (± 1.7)	163 (± 8)	54.8 (± 9.2)	20.7 (± 2.8)	3.7 (± 2.1)	1.28 (± 1.75)	1.30 (± 1.79)	14 (± 7)	9 (± 5)
Boys	20	12.9 (± 1.4)	164 (± 10)	59.5 (± 15.7)	22.0 (± 4.5)	4.3 (± 2.1)	1.32 (± 1.69)	1.33 (± 1.44)	13 (± 7)	9 (± 5)
Total	40	13.4 (± 1.6)	163 (± 9)	57.1 (± 12.9)	21.4 (± 3.8)	4.0 (± 2.1)	1.30 (± 1.70)	1.31 (± 1.60)	14 (± 7)	9 (± 5)

VAS, visual analog scale; BMI, body mass index; PD, pain drawing; BAPQ, Bath Adolescent Pain Questionnaire.

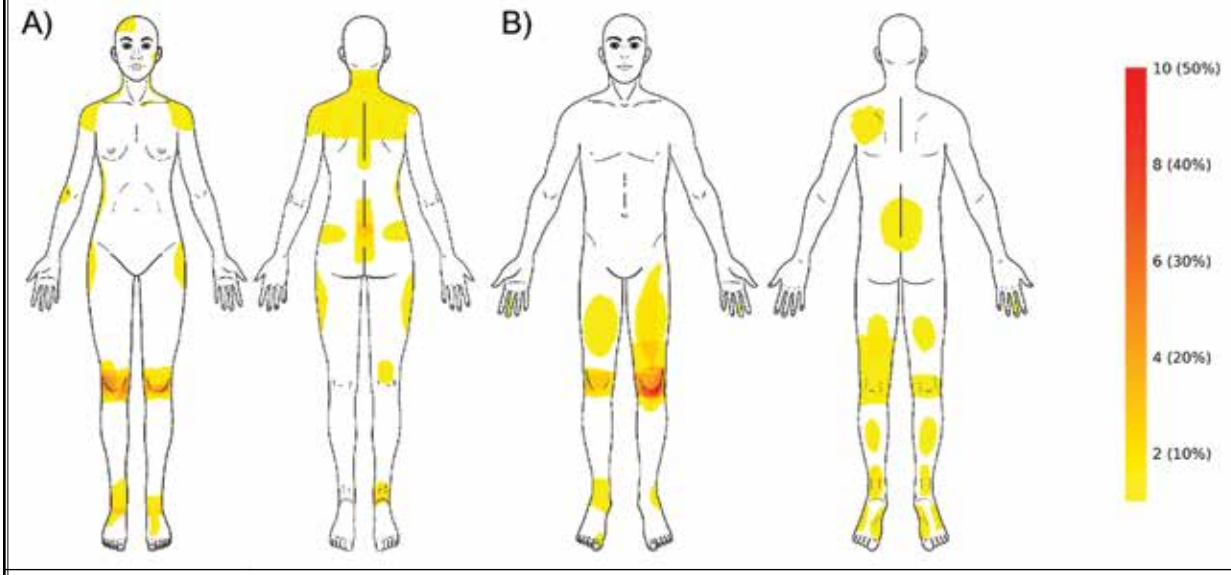

Fig. 1. *Pain frequency maps generated separately for girls (A) and boys (B). The color bar represents the frequency of the colored areas. Red represents the most frequently reported area of pain, whereas yellow represents the least.*

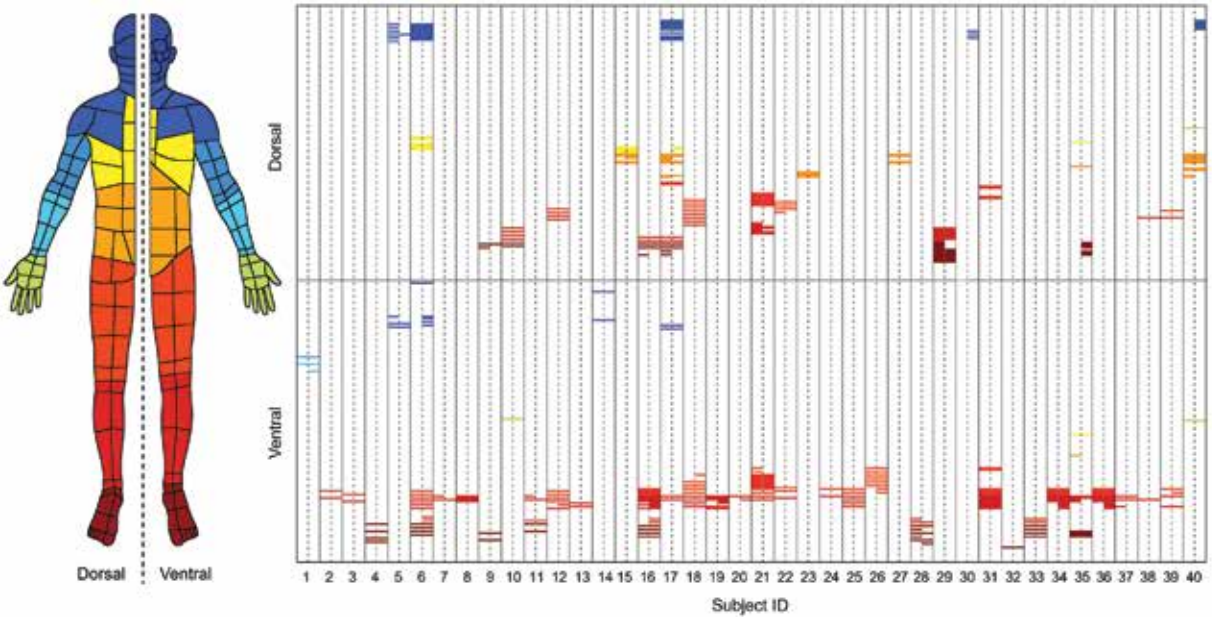

Fig. 2. *Representation of the areas colored by all participants in the 2 BCs. Each column represents one participant, subdivided into PD1 and PD2 (with dotted line). The areas are ordered from top to bottom, starting from the parietal area of the skull, down to the fifth toe. The colors in the graph refer to the areas represented in the BC on the left. The upper part of the graph represents the dorsal areas, the lower part the ventral areas.*

The reliability of pain location reporting was also supported by the Jaccard index mean score of 0.82 (± 0.19). The complete results of the reliability analysis are reported in Table 2.

Discussion

We assessed the reliability of adolescents, aged 11 to 16 years, who used PDs to report the extent and

location of their pain. The results for pain extent reporting show a reliability of the total sample ranging from good to excellent. Moreover, the mean value of the reliability of the pain location reporting exceeded the value of 0.82, meaning that, on average, 82% of the colored areas overlapped. The analysis of pain extent revealed a reliability among adolescents that was comparable to or even higher than that observed in adults with chronic neck (CNP) pain and chronic lower back pain (CLBP) (5). The adolescent patients also demonstrated a level of reliability in pain reporting that equaled or exceeded that of adults with temporomandibular disorders (11).

The population included in the current study was heterogeneous. Indeed, reliability may vary depending on the nature of the pain, its duration, or the type of disorder and body areas involved. Barbero et al (5) evaluated PDs by patients with CLBP or CNP that persisted for an average of 80.5 ± 98.8 months and 83.3 ± 102.8 months, respectively. Although the population observed by Barbero et al (5) was much less heterogeneous than the one included in the current study, the reliability of the 2 studies was comparable: the $ICC^{2,1}$s of the pain extents calculated on the test-retest of patients with CLBP and CNP in the previous study were 0.97 (95% CI: 0.95-0.98) and 0.92 (95% CI: 0.87-0.98), respectively, both similar to that of the current study (0.959, 95% CI: 0.925-0.978). The results obtained by Pitance et al (11) are also comparable to those of the current study, although the confidence intervals are wider, making its reliability less certain, particularly as the frontal-body BC is concerned. In the present study, only one patient, who had pain related to an ankle sprain, reported difficulty in drawing her pain, since it was very lateral. Although the patient reported this struggle, analysis of her data showed little variation in pain extent between PD1 and PD2 (-0.030%) and a Jaccard index only slightly below the sample mean (0.75).

Regarding pain location, the present study's reliability results are higher than those of the studies on adults by Barbero et al (5) and Pitance et al (11). The differences in the reliability of pain location between the current study and the previous studies could be due to the differences in the somatic presentation of pain and the psychological features of the populations included. Patients with CLBP and CNP (5) commonly report pain in the dorsal region of the body, which may be more difficult to represent. Indeed, the pain location analyzed with the Jaccard index in the current study was slightly lower in girls, who on average reported posterior pain more frequently than did boys. Additionally, many patients included in this study reported anterior pain, often in the knee. This type of pain may be represented more easily than the pain considered in the populations of previous studies, both because the knee area may be more easily identifiable on the BC and because the pain affects a more circumscribed area. Furthermore, patients with CLBP and CNP (5), as well as those with temporomandibular disorders (11), often present with symptoms associated with central sensitization, pain catastrophizing, anxiety, depression, disability, and impaired quality of life, all of which may influence PDs. In contrast, the current sample of adolescents did not demonstrate pain catastrophizing or poor

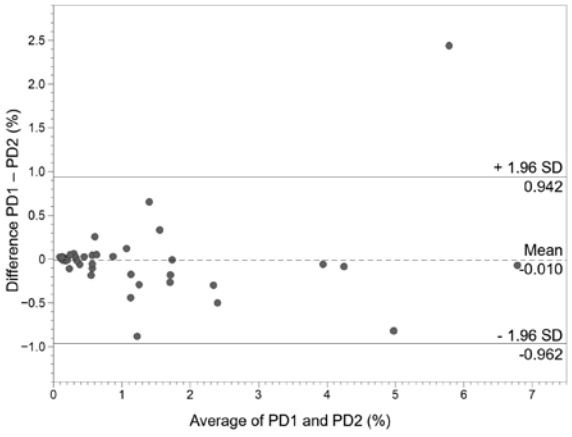

Fig. 3. *Bland-Altman plot showing the test-retest reliability of pain extent. PD, pain drawing; SD, standard deviation.*

Table 2. *Results of the reliability analysis.*

	Total (40)	Girls (20)	Boys (20)
$ICC_{2,1}$ (mean, 95% CI)	0.959 (0.925-0.978)	0.992 (0.980-0.997)	0.920 (0.810-0.968)
Mean of difference (%)	-0.010	-0.014	-0.006
Limits of agreement (%)	-0.962 to 0.942	-0.475 to 0.447	-1.290 to 1.278
Jaccard index (mean, SD)	0.82 (± 0.19)	0.79 (± 0.22)	0.85 (± 0.15)

ICC, intraclass correlation coefficient; CI, confidence interval.

physical functioning. It should be noted, however, that we did not collect data on pain duration in this study.

The time that elapsed between the completion of each of the 2 PDs ensured a good balance between the stability of symptoms and the risk of recalling the previous PD. This procedure is reasonable and has been proposed in earlier studies (5). In addition, the distribution on the Bland-Altman graph does not show any trends that could depend on a systematic bias.

Additionally, the researcher ensured that the patients had no doubts about the completion of the PD and the instructions they had received. The patients also demonstrated and confirmed that they could fill out the PD with ease. Moreover, the children all reported a sense of ease in approaching the instrument, probably also due to their familiarity with markers and coloring on paper, activities that for patients of that age group are commonplace.

As emphasized by the Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (IMMPACT) group, there is a need for standardized, versatile, and applicable pain outcome measures for diverse populations (23). The pediatric group of IMMPACT (PedIMMPACT) also emphasizes that pain in children and adolescents deserves special attention, and although attempts have already been made to adapt assessment instruments, this population needs more valid and applicable outcome measures (24). The PDs presented in this study followed a process of adaptation that led to the modification of the BCs for the adolescent population. The applicability to multiple populations, also confirmed by this study's results, is an added value of the PD, which is now a reliable outcome measure that can overcome not only the communication barriers that often affect consultations with adolescents but also linguistic barriers. Initiating proper pain communication with adolescents is, furthermore, considered extremely important by the patients themselves (25), and a PD allows both the patient's personal involvement and the initiation of conversation about broader aspects than pain location and pain extent. Lee et al (25) indicated that, in some cases, children had difficulty describing their pain. Bypassing verbal communication and simply asking the patient to color the areas where pain is present could facilitate the evaluative approach. Thus, a PD provides a valid tool for the assessment of pain extent and pain location as well as an anchor for ensuring an assessment and treatment process that centers the adolescent's empowerment, in line with Lee's recommendations (25).

Notably, a recent study published by Foxen-Craft et al (26) emphasized the importance of recognizing widespread pain in young people with chronic pain conditions, highlighting the association of widespread pain with aspects such as pain interference, pain catastrophizing, fatigue, anxiety, and depression. Identifying patients with widespread pain would enable physicians to detect the presence of a nociplastic pain mechanism and adapt the treatment to fit it. This possible development makes the use of body maps and PDs to assess adolescent patients and inform their treatment an even more relevant method.

All these considerations underline the validity of these results and provide an incentive to use the PD technique with adolescent patients. Simultaneously, the aforementioned considerations highlight the importance of conducting further studies that investigate the effectiveness of PDs for adolescent patients.

Conclusions

In conclusion, this study confirms adolescents' reliability in reporting the extent and location of their pain using paper-based PDs. Nevertheless, further studies are needed to verify PDs' other psychometric properties on adolescents.

Authors' Contributions

MB, AF, and DF were the major contributors to the conception and design of the study. CC, ES, and FV contributed to the conception and design of the study. AF was the main responsible for data collection. AF, MB, and CC analyzed the data. AF, MB, and DF interpreted the data. AF and MB were responsible for drafting the manuscript. DF and CC, FV, and ES revised the manuscript and contributed substantially. All authors approved the final version.

Acknowledgments

The authors would like to thank Fisioterapia Kinesis SA and the physiotherapy and administrative team, as well as the research secretariat and the secretariat of the Istituto Pediatrico della Svizzera Italiana, for their support during data collection.

References

1. King S, Chambers CT, Huguet A, et al. The epidemiology of chronic pain in children and adolescents revisited: A systematic review. *Pain* 2011; 152:2729-2738.
2. Palmer H. Pain charts; a description of a technique whereby functional pain may be diagnosed from organic pain. *N Z Med J* 1949; 48:187-213.
3. Barbero M, Navarro-Santana MJ, Palacios-Ceña M, et al. Clinical significance and diagnostic value of pain extent extracted from pain drawings: A scoping review. *Diagnostics (Basel)* 2020; 10:604.
4. Leoni D, Falla D, Heitz C, et al. Test-retest reliability in reporting the pain induced by a pain provocation test: Further validation of a novel approach for pain drawing acquisition and analysis. *Pain Pract* 2017; 17:176-184.
5. Barbero M, Moresi F, Leoni D, Gatti R, Egloff M, Falla D. Test-retest reliability of pain extent and pain location using a novel method for pain drawing analysis. *Eur J Pain* 2015; 19:1129-1138.
6. Foxen-Craft E, Scott EL, Kullgren KA, et al. Pain location and widespread pain in youth with orthopaedic conditions: Exploration of the reliability and validity of a body map. *Eur J Pain* 2019; 23:57-65.
7. Jamison RN, Washington TA, Gulur P, et al. Reliability of a preliminary 3-D pain mapping program. *Pain Med* 2011; 12:344-351.
8. Margolis RB, Chibnall JT, Tait RC. Test-retest reliability of the pain drawing instrument. *Pain* 1988; 33:49-51.
9. Roach KE, Brown MD, Dunigan KM, Kusek CL, Walas M. Test-retest reliability of patient reports of low back pain. *J Orthop Sports Phys Ther* 1997; 26:253-259.
10. Southerst D, Stupar M, Côté P, Mior S, Stern P. The reliability of measuring pain distribution and location using body pain diagrams in patients with acute whiplash-associated disorders. *J Manipulative Physiol Ther* 2013; 36:395-402.
11. Pitance L, De Longhi B, Gerard E, et al. Digital pain drawings are a useful and reliable tool for assessing patients with temporomandibular disorders. *J Oral Rehabil* 2021; 48:798-808.
12. Rosner J, Lütolf R, Hostettler P, et al. Assessment of neuropathic pain after spinal cord injury using quantitative pain drawings. *Spinal Cord* 2021; 59:529-537.
13. Rodrigues JC, Avila MA, Dos Reis FJJ, et al. 'Painting my pain': The use of pain drawings to assess multisite pain in women with primary dysmenorrhea. *BMC Women's Health* 2022; 22:370.
14. Silva AG, De Francesco S, Rodrigues M, Queirós A, Cerqueira M. Test-retest reliability of pain measures in institutionalized older adults: Number of painful body sites, pain intensity, and pain extent. *Pain Pract* 2021; 21:270-276.
15. von Baeyer CL, Lin V, Seidman LC, Tsao JC, Zeltzer LK. Pain charts (body maps or manikins) in assessment of the location of pediatric pain. *Pain Manag* 2011; 1:61-68.
16. Swissethics. Checklist: Research on and with children and adolescents under the age of 18. 2018. Retrieved from https://swissethics.ch/en/templates/forschung-mit-kindern
17. Kottner J, Audigé L, Brorson S, et al. Guidelines for reporting reliability and agreement studies (GRRAS) were proposed. *J Clin Epidemiol* 2011; 64:96-106.
18. Monticone M, Baiardi P, Ferrari S, et al. Development of the Italian version of the Pain Catastrophising Scale (PCS-I): Cross-cultural adaptation, factor analysis, reliability, validity and sensitivity to change. *Qual Life Res* 2012; 21:1045-1050.
19. Eccleston C, Jordan A, McCracken LM, Sleed M, Connell H, Clinch J. The Bath Adolescent Pain Questionnaire (BAPQ): Development and preliminary psychometric evaluation of an instrument to assess the impact of chronic pain on adolescents. *Pain* 2005; 118:263-270.
20. Walter SD, Eliasziw M, Donner A. Sample size and optimal designs for reliability studies. *Stat Med* 1998; 17:101-110.
21. Koo TK, Li MY. A guideline of selecting and reporting intraclass correlation coefficients for reliability research. *J Chiropr Med* 2016; 15:155-163.
22. Jaccard P. Nouvelles recherches sur la distribution florale. *Bull la Société vaudoise des Sci Nat* 1908; 44:223-270.
23. Turk DC, Dworkin RH, Burke LB, et al. Developing patient-reported outcome measures for pain clinical trials: IMMPACT recommendations. *Pain* 2006; 125:208-215.
24. McGrath PJ, Walco GA, Turk DC, et al. Core outcome domains and measures for pediatric acute and chronic/recurrent pain clinical trials: PedIMMPACT recommendations. *J Pain* 2008; 9:771-783.
25. Lee RR, Mountain D, Connelly M, et al. 'That's what makes me better': Investigating children and adolescents' experiences of pain communication with healthcare professionals in paediatric rheumatology. *Eur J Pain* 2023; 27:111-128.
26. Foxen-Craft E, Bourchtein E, Kaplan C, Clauw DJ, Scott E. Pain widespreadedness, and not primary pain location, is associated with comorbid symptoms in children with chronic pain. *Clin J Pain* 2023; 39:1-7.