BrightHearts: A pilot study of biofeedback assisted relaxation training for the management of chronic pain in children with cerebral palsy

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Abstract
Background: Chronic pain is estimated to impact one-in-three children with cerebral palsy (CP). Psychological interventions including behavioral and cognitive strategies play a key role in chronic pain management, but there is a paucity of research exploring their use in children with CP.
Aim: To investigate the acceptability and feasibility of biofeedback assisted relaxation training (BART) for chronic pain management in children with CP using a mixed-methods study design.
Methods: Biofeedback assisted relaxation training was delivered via BrightHearts, an iOS application. Inclusion criteria were as follows: CP; self-reported chronic pain; age 9-18 years; and fluent English speaker. Children used BrightHearts for ten minutes daily, over four weeks. Qualitative post-intervention interviews were undertaken (child, parent) and quantitative pre-post measures (child) were gathered including pain intensity (numerical rating scale), and anxiety intensity (numerical rating scale). Content analysis was conducted for qualitative data. Descriptive statistics and exploratory analyses were performed for quantitative data.
Results: Ten children participated (n = 3 male, mean age = 13.1 years SD = 2.5 years, GMFCS level I = 4, II = 2, III = 3, IV = 1). Predominant movement disorder was spasticity (n = 7) and dyskinesia, mainly dystonia (n = 3). Content analysis suggested an overarching theme “BrightHearts is a good thing to put in my toolbox” providing an overall representation of participants’ experiences. For many, BrightHearts was a valuable supplement to children’s pain management strategies: “The source of the pain is still there, but the actual effect of the pain isn’t so relevant.” Four sub-themes were identified: “Managing my pain;” “Managing my anxiety and stress,” “Helping me do what I need to do;” and “Fitting it into my life.” Some participants reported improvements in their anxiety management, and others described benefits in sleep and school following improved pain/anxiety management. A range of practical and personal factors within this heterogeneous group presented barriers to using BrightHearts including limited time, attention, and boredom. Seven-out-of-eight children would recommend...
1 | INTRODUCTION

Chronic pain has been identified as an alarmingly common comorbidity in cerebral palsy (CP), impacting one-in-three children with the condition.1 Chronic pain can intensify disability associated with CP, interfering with physical function, participation in recreational activities, and quality of life.2 It is also associated with increased risk of depression, insomnia, and anxiety.3 For many children with CP, the negative consequences of chronic pain may be long-lasting, with improvements in pain over time seen in only a minority (21%).4 There is a paucity of high-quality evidence to support current clinical practice, thus limiting pain management in this group.5

A multimodal treatment approach is considered the gold-standard in chronic pain management, corresponding with greater recognition of the biopsychosocial model of pain.6 Psychological interventions are recognized to have a critical role in the management of chronic pain.7,8 In the general pediatric population, there is evidence to support psychological interventions (including remote-delivered interventions) for improvement in pain intensity and pain-related disability in young people with chronic pain.7,8 These interventions include behavioral strategies, cognitive strategies, or a combination of both.7,8

A recent review emphasized the lack of multimodal and psychological interventions for chronic pain management in children with CP, in contrast with other pediatric diagnostic groups.5,7 Delivering appropriate psychological treatment suitable for children with CP and chronic pain may be complicated by the frequent co-occurrence of intellectual disability and non-verbal communication.5 Chaleat-Valayer et al9 reported that children with CP engage in fewer cognitive and behavioral strategies to manage their pain experience (ie, pain coping strategies) compared to typically developing children.

Biofeedback assisted relaxation training (BART) is a psychological intervention, which can be used for pain management.10 It is a behavioral therapy whereby an individual learns to voluntarily modulate a biological process via electromechanical devices that measure physiological parameters and deliver feedback (eg, auditory, visual).10 Heart rate is one of many physiological indicators of pain and anxiety. It may be used to teach a user to engage in purposeful relaxation.11 BrightHearts is a heart-rate-controlled application (app) for remote delivery of BART.11 Designed specifically for pediatric use, BrightHearts displays an interactive geometric artwork, which changes in response to heart-rate variability.11 It has demonstrated feasibility and acceptability for the management of procedure-related pain and anxiety in children, including children with CP.12

This mixed-methods pilot study aimed to evaluate the feasibility and acceptability of BART via BrightHearts for the management of chronic pain in children aged 9-18 years with CP. We explored children’s experience of using BrightHearts daily, over a four-week period. A qualitative understanding of participant and parent experiences was sought and quantitative outcomes were measured.

The detailed research questions addressed using qualitative methodology were:

1. Do child-parent dyads perceive BrightHearts helps the child manage their chronic pain, and if so, how?
2. Do child-parent dyads perceive an impact of using BrightHearts on activities and participation, and if so, what is the impact?
3. Do child-parent dyads perceive the recommended frequency and duration of BrightHearts use (10 minutes daily, over one month) acceptable for the management of chronic pain, and why/why not?

Quantitative analysis was used to explore the effect of BrightHearts use on pain, anxiety, and quality of life.

2 | METHODS

2.1 | Study design

A prospective pilot study was conducted between September 2017 and February 2019. The study followed an embedded mixed-methods design: experimental model, with qualitative methods following completion of the main quantitative experiment.13 Child-parent dyads were recruited via the Rehabilitation Departments, Sydney Children’s Hospitals Network and the New South Wales/Australian Capital Territory CP Registers, Cerebral Palsy Alliance. Informed written consent was obtained from the child and their parent. Ethics approval was granted by the Sydney Children’s Hospitals Network and Cerebral Palsy Alliance Human Research Ethics Committees.
2.2 | Participants

Children with a diagnosis of CP, aged between 9 and 18 years, and their parent were included in the study. Inclusion criteria were as follows: (1) Child self-reported chronic pain, defined as recurrent pain over a period of at least three months; and (2) Fluent English speaker. Exclusion criteria were (1) Child has previous biofeedback training; (2) Child currently receiving intensive or multimodal chronic pain treatment by specialist pain teams; (3) Child diagnosis of moderate intellectual impairment or greater; (4) Child diagnosis of severe auditory or vision impairment; and (5) Child or their parent require an interpreter. The first 10 participants to meet inclusion criteria and provide informed consent were included. This sample size was decided in advance as it was seen as appropriate size for the primary qualitative data analysis and would provide insight regarding estimated time for sample recruitment, study design (particularly participant burden of outcome measures), and whether further investigation is justified.

2.3 | Intervention

Child and parent participants were provided education and training on the BrightHearts intervention at the beginning of the study by the first author. Child participants were instructed to use the BrightHearts app once-a-day for 10 minutes, over a four-week period. There were no specific instructions regarding the time of day to use the app. BrightHearts is a BART app designed for Apple iOS devices (iPhone, iPad, etc.). BrightHearts was developed for pediatric populations through an iterative design process and has demonstrated feasibility for use with children experiencing procedure-related pain. The app displays an interactive visual and auditory artwork, responding to changes in heart rate measured using a wireless, Bluetooth heart-rate sensor. This pilot study utilized the Mio Link device, a Bluetooth 4.0 transmitter with wrist band design, to send real-time heart-rate data. The app displays layers of concentric circles that change in color and contract or expand with changes in baseline heart rate, calculated an averaging of the first thirty-two inter-beat-intervals measured at the start of each session. In accordance with temperature mapping, the artwork changes colors from orange (baseline) shifting to yellow, green, cyan, and eventually blue as average heart rate (moving window, average of the last thirty-two beats) decreases further from baseline average—to a maximum decrease of 10%. Additional bursts of blue rings appear when the lowered heart rate is sustained. A deviation toward baseline heart rate alters the artwork color back to the initial orange. Auditory feedback is also provided with relaxing chime sounds initiated each time there is a decrease in heart rate (moving average of the previous four inter-beat-intervals measured in milliseconds), and with a shimmering “mark tree” wind chime sound effect that is only revealed when average heart rate of the last thirty-two interbeat intervals has decreased sufficiently from the baseline (Figure 1).

2.4 | Measures

2.4.1 | Demographics and medical record review

Information was gathered via a child-parent dyad completed demographics form and review of medical records. Data were collected relating to movement disorder/s, topography of movement disorder/s, Gross Motor Function Classification System, Manual...
Ability Classification System, and comorbid diagnoses including anxiety (documentation of generalized anxiety by clinician) and intellectual impairment (mild impairment: IQ 50-69 or as assessed by clinical judgement).

2.4.2 | Semi-structured interviews

Following completion of the intervention period, semi-structured interviews were conducted with the child participant (n = 10) and the parent participant (n = 9; Appendix S1). Questions explored the individual experience of using BrightHearts, the implementation of BrightHearts for the child’s management of pain, perceived barriers for use, and how the intervention did/did not help the child participant manage their pain. All interviews were conducted by a single interviewer (KO), a researcher trained in interview techniques and familiar with participants having been the point of contact throughout the study. Child interviews were conducted alone (n = 5) or with the parent present (n = 5), based on child-parent dyad preference. All child interviews were conducted face-to-face. All but one parent interview was conducted face-to-face, with one parent interview conducted via telephone for parent convenience. Interviews ranged from 19 to 36 minutes. Interviews explored the child and the parent perspective of using BrightHearts and focused on the impact of BrightHearts on their pain and/or anxiety, influence on their participation, the acceptability of the intervention including frequency and duration of use of the app, and overall experience. All interviews were recorded digitally and transcribed verbatim for analysis.

2.4.3 | Participant satisfaction survey

A brief paper-based survey for completion by child participants was utilized to supplement the qualitative interview (Appendix S2). The survey included a combination of open-inquiry and dichotomized (yes/no) questions concerning participants’ satisfaction with the intervention, likes/dislikes, and recommendations.

2.4.4 | Pain, anxiety, and impact of pain weekly questionnaire

A questionnaire was conducted via telephone/farce-to-face with the child participant at baseline, weekly during the intervention period, and at conclusion of the pilot. Participants were asked to rate their pain intensity (0-10) and anxiety intensity (0-10) in the previous week using single integer numerical rating scales. Due to the frequent co-occurrence of chronic pain and anxiety disorder, as well as previous application of BART for anxiety treatment in the general population, secondary outcome measures of anxiety were evaluated.

2.4.5 | Pre- and post- quantitative surveys (pain, anxiety, and quality of life)

A comprehensive range of quantitative paper-based surveys were conducted at baseline and follow-up, as a means of choosing appropriate outcome measures for potential future randomized controlled trial. All outcome measures were completed as child self-report. A pain survey was administered including chronic pain presence (yes/no), pain intensity (Faces Pain Scale - Revised), pain location on body chart, and pain interference (Patient Reporting Outcomes Measurement Information System Pediatric Pain Interference Scale). The State-Trait Anxiety Inventory and Cerebral Palsy Quality of Life questionnaire were also administered.

2.5 | Data analysis

Qualitative data gathered during semi-structured interviews with child-parent dyads were analyzed using a content analysis approach, “a research method for making replicable and valid inferences from data to their context, with the purpose of providing knowledge, new insights” (p.108). The aim of the content analysis was to gather a “condensed and broad description of the phenomenon” being explored (p.108). The transcribed interviews formed the unit of analysis, which were initially reviewed in their entirety via an inductive approach to classify “meaning units” applicable to the research question. Meaning units included “words, sentences or paragraphs containing aspects related to each other through their content and context” (p.106), which were labeled with codes. These codes were then reviewed, comparing similarities/differences and classifying codes as “belonging” to higher order categories. Through a process of abstraction, categories were grouped under four sub-themes with one overarching theme. Methods including continuous dialogue among research investigators during analysis, and utilization of multiple direct quotations within the results were applied to improve trustworthiness of the qualitative findings. Qualitative data analysis was conducted by the first and second authors using NVivo software.

Quantitative data analysis was conducted using SPSS Version 25 software. Normality was initially assessed using the Shapiro-Wilk analysis and inspection of histogram graphs. As data was normally distributed, paired t tests were conducted with mean and standard deviation scores reported. Additionally, nonparametric Wilcoxon rank sum analysis was conducted due to small sample size. Findings remained unchanged following nonparametric analysis and paired t-tests results are reported for ease of clarification.

3 | RESULTS

Ten children completed the pilot study between September 2017 and February 2019 (n = 3 males; mean age = 13.1 years SD = 2.5 years). Demographic characteristics of all ten child participants are detailed
in Table 1. One further child-parent dyad was recruited and consented to participate but withdrew during the first week of the study, choosing not to provide a reason. Semi-structured interviews were conducted with 10 child participants and a parent of 9/10 of these children. One parent could not complete an interview due to scheduling difficulties.

### 3.1 Qualitative findings

Content analysis of the qualitative data led to the identification of an overarching theme "BrightHearts is a good thing to put in my toolbox," along with four sub-themes: "Managing my pain," "Managing my anxiety and stress," "Helping me do what I need to do," and "Fitting it into my life." Each of the four sub-themes is comprised of categories, as demonstrated in Figure 2. These categories represent the experiences and views of child participants and their parents, which contribute to each sub-theme.

#### 3.1.1 Overarching theme: "BrightHearts is a good thing to put in my toolbox"

The child participant quote "BrightHearts is a good thing to put in my toolbox" (Child E) was identified as the overarching theme, providing an overall representation of child-parent dyads’ experiences and perceptions of using BrightHearts for the management of chronic pain. A few parents acknowledged that although BrightHearts was unlikely to be a first-line pain treatment, it equipped young people with enhanced skills to potentially better manage their pain experience: “The source of the pain is still there for sure, but I think the actual effects of the pain isn’t so relevant” (Parent C). Parent E described BrightHearts as "a brilliant alternative or a supplement to things that people are already doing."

Three child participants reported equal benefit in managing both pain and anxiety as comorbid issues, while six children saw greater benefit in managing either anxiety (n = 4) or pain (n = 2). One participant did not report improvements in pain or anxiety management.
All four child participants who reported greater benefit in managing anxiety had a comorbid history of anxiety. A range of practical and personal factors within this heterogeneous group also presented barriers to using BrightHearts. Despite these challenges, BrightHearts was viewed as a worthwhile platform to develop relaxation skills, which could be adapted to multiple scenarios to enhance function and participation. Each of the four sub-themes will be discussed below, incorporating both child and parent quotes to highlight the themes.

3.1.2 | Sub-theme: Managing my pain

BrightHearts allowed children to better manage their pain in various ways. Although most child participants reported being “in the same amount of pain,” they reported that “dealing with the pain now is different.” Several participants found BrightHearts helpful in implementing breathing techniques and relaxation skills to manage pain: “I can now sit and breathe when I am in pain, and my muscles relax” (Child D) and “to stay more relaxed when in pain” (Child F). These relaxation skills were also beneficial in managing the emotional/cognitive response to pain, particularly pain-related anxiety. For example, one child said “it can keep you calm and relaxed and keep you from stressing about the pain” (Child J).

For others, BrightHearts utilized distraction mechanisms to manage pain and pain-related anxiety: “the app takes your focus away from the pain. If you focus on [the pain], it just gets worse instead of getting better” (Child H) and “It calmed me down and distracted me from pain” (Child F). One child participant, however, did not find the geometric artwork to be a suitable form of distraction, suggesting alternative positive reinforcement mechanisms tailored to their age and/or personal preference (eg, a game component to learn BART). Another child participant reported no benefit for their pain symptoms, but rather a positive impact in managing their general anxiety. Despite these individual differences in experience, all child-parent dyads would recommend the BrightHearts intervention to others for chronic pain management as there was “no harm in trying” (Parent B) and “There’s so many parents that, they’re so desperate to help their kids in any way, why not try it?” (Parent I). Non-pharmacological strategies for pain management were perceived as a priority for both children and parents, especially if they offered an “alternative to medication” and associated side-effects: “the medications can just make you so tired that you can’t stay up” (Child E).

3.1.3 | Sub-theme: Managing my anxiety and stress

Child-parent dyads found the BrightHearts intervention helpful in managing the young person’s general anxiety and stress. In this complex clinical cohort, where chronic pain and general anxiety often present concurrently, many child-parent dyads reported a widespread benefit from using BrightHearts. Others reported that BrightHearts “helped the anxiety side more than the pain side” (Child I). Parent E found BrightHearts useful for their child to “to work through the anxiety attack or offset an anxiety attack.” Children and parents also spoke of BrightHearts being a useful way to “learn to regulate minor stressors” (Parent E), for example, “when I got frustrated it helped me to relax” (Child H). Child-parent dyads emphasized the importance of active involvement and having a “sense of control” in the management of anxiety. BrightHearts was a valuable resource to one dyad because “the things that she can control help reduce the anxiety far more than things that are external” (Parent H). Another parent emphasized the significance of the feedback component of the intervention in facilitating relaxation and stress management “because just telling him to control his breathing, as a parent, it never works” (Parent A). Child participants reported feeling “calm” and “relaxed” when using the app, with some individuals preferring the geometric artwork while others found the music “more soothing” and “the music is quite calming” (Child C). However, using the app could be challenging at times and counter-intuitive to managing anxiety/stressors, with a small number of participants reporting at times feeling “stressed” and “frustrated” when they were unable to transition the colors of the geometric artwork from red to blue- “it was stressful... it kept going orange” (Child G).

3.1.4 | Sub-theme: Helping me do what I need to do

Many child participants reflected upon other ways in which improved pain and/or anxiety management using BrightHearts impacted positively in their day-to-day lives. Some child-parent dyads benefited from BrightHearts as a global relaxation tool, which improved the young person’s sleep: “you’re more relaxed and you’re able to fall asleep quickly” (Child I) and “a good thing to really calm him down before bed” (Parent C). A few reported previous sleep interference due to pain, which improved with BrightHearts use. One parent participant said: “using the app, disrupted sleep was rarer. It [sore legs] still happened but not as frequent” (Parent C).

Some child participants reported being able to participate better in their school life as a result of enhanced management of pain and/or anxiety. One child-parent dyad reported “a lot of anxiety from school and schoolwork” (Parent A) and using BrightHearts was “good to just stop thinking about stressful things and just focus on that [the app]” (Child A). This parent further commented that the child “would never forget on a school day to use it [BrightHearts].” For another child, painful muscle spasms previously interfered with school life: “I just would ignore it [painful spasms] until lunchtime, but now I can deal with it effectively” (Child H). BrightHearts allowed this participant to “use the breathing techniques when I had it [painful spasms] at school.”

One parent participant reported improvements in their child’s physical functioning as a result of better pain management: “His mobility’s a lot better because there’s—I don’t know whether it’s the fact there’s not such a fear of pain.” (Parent D).

3.1.5 | Sub-theme: Fitting it into my life

Participants were positive about the accessibility of the BrightHearts app and its ease of use. Some child participants flourished with the
TABLE 2  Change in pain, anxiety, and quality of life outcomes over time

| Outcomes                        | Baseline | Postintervention | 95% CI       | P    |
|---------------------------------|----------|------------------|--------------|------|
| Pain intensity (NRS)            | 4.4 ± 2.9| 4.4 ± 2.3        | −1.97 to 1.97| 1.000|
| Anxiety intensity (NRS)         | 4.4 ± 3.4| 4.0 ± 2.0        | −2.41 to 3.21| .754 |
| State anxiety (STAI)            | 33.4 ± 11.1| 31.3 ± 7.6      | −1.86 to 6.11| .248 |
| Trait anxiety (STAI)            | 39.6 ± 11.7| 37.6 ± 11.5      | −1.34 to 5.34| .200 |
| CPQoL-feelings about functioning| 73.0 ± 19.6| 71.6 ± 21.1      | −6.27 to 9.08| .670 |

Note: Data are reported as means and standard deviations. Abbreviations: CPQoL, cerebral palsy quality of life questionnaire (score range: 0-100); NRS, numerical rating scale (score range: 0-10); STAI, state trait anxiety inventory (score range: 0-60).

The “feelings about functioning” domain of the Cerebral Palsy Quality of Life questionnaire is reported as it is the only common domain across both 9-12 y version and 13-18 y version.

3.2 | Quantitative results

3.2.1 | Participant satisfaction survey

Findings from dichotomized (yes/no) items on the participant satisfaction survey (n = 2 missing child data) illustrated that 7/8 child participants would recommend the BrightHearts app to other people who experience pain similar to their own, and 6/8 child participants noticed a positive change in their pain since using BrightHearts.

3.2.2 | Pre- and post- quantitative surveys (pain, anxiety, and quality of life)

All paper-based questionnaires were completed by n = 7 child participants. Certain follow-up questionnaires were not completed by some child participants due to conflicting schedules and study burden including: Cerebral Palsy Quality of Life questionnaire (n = 3), State-Trait Anxiety Inventory (n = 2), pain experience survey (n = 2), and participant satisfaction survey (n = 2).

We found no evidence of a difference in participants’ self-reported pain intensity or anxiety intensity scores, as assessed via numerical rating scales (0-10), between baseline and conclusion of the intervention (see Table 2). Across the cohort, a trend of decreasing average pain intensity and anxiety intensity ratings was observed during the intervention period, though this was not statistically significant. No significant differences were detected in participants’ self-reported quality of life, trait anxiety, and state anxiety post-intervention, when compared to baseline (see Table 2).

4 | DISCUSSION

Chronic pain is a common problem impacting young people with CP, yet research has not explored psychological interventions for the management of chronic pain in this cohort. Our study explored the use of BrightHearts, a biofeedback assisted relaxation training (BART) application, for the management of chronic pain in children aged 9-18 years with a diagnosis of CP. This pilot study examined the outcome of daily, ten-minute use of the BrightHearts app over a four-week period. The main result of our study was the qualitative findings that child and parent participants reported a generally positive experience engaging with the intervention and suggested beneficial impact on managing pain, anxiety, and the potential to help increase participation in daily life. Although the intervention was sometimes difficult to fit into one’s lifestyle,
regular engagement with the BrightHearts app was perceived as acceptable and feasible. The quantitative results showed no statistically significant reduction in pain or anxiety intensity scores following the intervention, though the study was not powered to detect such differences.

Chronic pain is often complex with numerous contributing factors, requiring a multimodal and interdisciplinary intervention for effective treatment. In our study, child-parent dyads acknowledged the importance of this multimodal approach and appreciated the role of non-pharmacological strategies. This aligns with Gross et al’s (2018) Delphi study where exploration of innovative pain treatments and alternative therapies were identified as leading research priorities by individuals with CP. Our key qualitative finding that BrightHearts is “a good thing to put in my toolbox” is a promising result suggesting that BART may be useful non-pharmacological therapy as part of a holistic treatment approach.

Our study emphasized the complex interplay between pain and psychosocial symptoms, particularly anxiety. Many study participants reported a therapeutic benefit to both chronic pain and general anxiety, with some reporting greater effect in one domain while others had difficulty distinguishing between the two. This is likely influenced by the co-occurrence of a history of anxiety in half of our study cohort, consistent with the frequent rates reported in pain literature. These findings suggest that improvements in symptoms may vary depending on individual factors and the suitability of BART may potentially be personalized. Our study design and small sample size did not permit an investigation of mental health conditions on treatment effect. Future research should consider psychological symptoms commonly co-occurring with chronic pain, particularly depression and anxiety disorder, and their impact on treatment outcomes.

In addition to generalized mental health conditions, it is imperative to consider pain-related emotions and cognitions. Participants in our study acknowledged the key role of affective mechanisms in the perpetuation of pain, such as pain catastrophizing (eg, “If you focus on [the pain], it just gets worse instead of getting better” (Child H)). Some child-parent dyads suggested that BrightHearts was helpful in improving maladaptive pain behaviors to gain greater control of their pain experience (eg, “It can keep you calm and relaxed and keep you from stressing about the pain” (Child J)). Cherkin et al recently demonstrated that mindfulness-based stress reduction programs, involving awareness of breathing strategies similar to BART, yielded equivalent improvements in pain catastrophizing and pain-related functional disability when compared to cognitive behavioral therapy. These optimistic findings suggest a need to further explore psychological pain treatment options in CP, with emphasis on targeting mechanisms associated with increased pain disability such as pain catastrophising.

Within this heterogeneous clinical cohort, it is acknowledged that BrightHearts may be an appropriate tool for some children with CP but not others. We hypothesized that this intervention may not be suitable for all intellectual abilities within the CP diagnostic spectrum. Thus, this pilot study was limited to children with no greater than a mild intellectual impairment. It is important to investigate how psychological interventions (including BrightHearts) may be modified to suit the needs of children with CP and moderate-to-severe intellectual disability. It is also imperative to consider individual factors (eg, personal preference, attention difficulties) that may discourage children from engaging with BrightHearts, such as finding it “boring” or not a suitable form of distraction. For others, engagement may need to be modified to be shorter in duration or less frequent based on individual factors, including attention difficulties and scheduling demands, respectively.

Overall participants emphasized the importance of a flexible intervention, which could be adapted to suit their individual needs. The remote delivery of BART was perceived as advantageous, whereby individuals were able to engage in the intervention at a time/setting of their choice. Purposeful and adaptive engagement in BART allowed some participants to make improvements in their participation and function (eg, evening use with reported improvements in sleep). Future development of the BrightHearts app with popular (but proprietary) heart-rate sensing devices (eg, Fit Bit, Apple watch) would allow for greater accessibility to this intervention, further increasing flexibility.

### 4.1 Strengths and limitations

This is the first study investigating the use of BART for chronic pain management in children with CP. The study was strengthened by its mixed-methods design, which allowed for more detailed insight than qualitative or quantitative analysis alone. As per the gold-standard in pain assessment, child self-report was utilized across both quantitative and qualitative data exploration. The addition of parent qualitative interviews offered further insight, which was valuable in understanding the acceptability of an intervention to be implemented in everyday life.

Our study has certain limitations to be considered when interpreting the findings. Our sample size was small and our study did not include a control arm. The sample size of ten was decided in advance, as opposed to ceasing recruitment when saturation of qualitative data was reached (the ideal for qualitative analysis). Nonetheless, the collection of data from 19 interviews (n = 10 child, n = 9 parent) was deemed saturated by authors. Missing data imposed restrictions for the quantitative data analysis and generalizability of these findings. Potential sources of selection bias include the recruitment of study participants via tertiary hospitals and CP Register, as well as the inclusion criteria being restricted to children with no greater than a mild intellectual impairment. It was hypothesized that BrightHearts may not be suitable for children with greater than a mild intellectual impairment; however, this has not been investigated to date.

1Not all devices that measure heart rate are suitable for BART. At the time of writing, the Apple Watch does not provide 3rd party developers with direct access to “real-time” heart-rate data, requiring developers to instead collect this information retrospectively via Apple Health Kit application.
Improvements in pain symptomology may have been confounded by participants receiving common medical treatments during the study period, which are known to alleviate pain in CP (eg, botulinum toxin-A injections, baclofen). Receiving such interventions was permitted within our study design based on the premise that the BrightHearts app would be clinically applicable as an intervention that compliments routine pharmacological treatments, as per the biopsychosocial model of pain.

4.2 | Future directions

There is a need for large randomized controlled trials to evaluate the efficacy of BART for chronic pain in children with CP, as well as other psychological interventions such as cognitive behavioral therapy. Multi-site trial design may assist in the recruitment of a specific sub-groups of children with CP and help avoid delays in recruitment. Missing data indicated that pre/post measures presented a participant burden, which may be avoided in future studies by co-designing outcome measures with individuals with a lived experience of CP. Studies should investigate the benefit of therapies over time (eg, 12-24 months) and include core outcome measures as recommended by clinical and research guidelines.33 Existing psychological interventions for chronic pain in the general pediatric population should be trialed for children with CP, after being adapted to various intellectual abilities and gross motor functional abilities. Mixed-methods research should investigate the suitability and acceptability of adapted interventions with larger sample sizes that cease recruitment when saturation is reached. Technology-delivered interventions are seen as advantageous by children with CP and their parents to improve accessibility. We encourage future research considering the use of technology (eg, smart phone application, internet website) to provide chronic pain interventions.

5 | CONCLUSION

This pilot study demonstrated the acceptability of biofeedback-assisted relaxation training (BART) for the management of chronic pain in children and adolescents with CP. The BrightHearts app was easy to use and flexible. Overall, young people and their parents had a positive perception of the BrightHearts app reporting benefit in their ability to manage chronic pain, general anxiety, and enhancing participation and functioning. Despite some challenges in fitting the intervention into everyday life, many child-parent dyads viewed the BrightHearts app as worthwhile and a valuable psychological intervention within a holistic model of chronic pain management. Future research with larger sample size would be beneficial to distinguish the effects on chronic pain and anxiety.

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CONFLICTS OF INTEREST

Authors KO, NS, SP, and AM have no conflicts of interest. GK declares the interest of being the co-investigator (artist and designer) responsible for the development of the interactive artwork, and has released a version of BrightHearts that is available free of charge from the Apple app store, via his individual developer account.

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SUPPORTING INFORMATION
Additional supporting information may be found in the online version of the article at the publisher’s website.

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