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Pain burden in children with cerebral palsy (CPPain) survey: Study protocol

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Abstract

Pain is a significant health concern for children living with cerebral palsy (CP). There are no population-level or large-scale multi-national datasets using common measures characterizing pain experience and interference (ie, pain burden) and management practices for children with CP. The aim of the CPPain survey is to generate a comprehensive understanding of pain burden and current management of pain to change clinical practice in CP. The CPPain survey is a comprehensive cross-sectional study. Researchers plan to recruit approximately 1400 children with CP (primary participants) across several countries over 6-12 months using multimodal recruitment strategies. Data will be collected from parents or guardians of children with CP (0-17 years) and

from children with CP (8-17 years) who are able to self-report. Siblings (12-17 years) will be invited to participate as controls. The CPPain survey consists of previously validated and study-specific questionnaires addressing demographic and diagnostic information, pain experience, pain management, pain interference, pain coping, activity and participation in everyday life, nutritional status, mental health, health-related quality of life, and the effect of the COVID-19 pandemic on pain and access to pain care. The survey will be distributed primarily online. Data will be analyzed using appropriate statistical methods for comparing groups. Stratification will be used to investigate subgroups, and analyses will be adjusted for appropriate sociodemographic variables. The Norwegian Regional Committee for Medical and Health Research Ethics and the Research Ethics Board at the University of Minnesota in USA have approved the study. Ethics approval in Canada, Sweden, and Finland is pending. In addition to dissemination through peer-reviewed journals and conferences, findings will be communicated through the CPPain Web site (www.sthf.no/cppain), Web sites directed toward users or clinicians, social media, special interest groups, stakeholder engagement activities, articles in user organization journals, and presentations in public media.

KEYWORDS
cerebral palsy, disability, pain, survey

1 | INTRODUCTION

Cerebral palsy (CP) is the most common physical disability in childhood. It describes a group of permanent developmental disorders caused by early injury to the immature brain and affects both movement and posture. Disturbances of sensation, perception, cognition, communication, and behavior, as well as a wide range of co-morbidities and secondary conditions, often accompany the motor disorder.[1] The degree of motor disability and associated difficulties vary widely,[1] with approximately 25% having severe CP, meaning that they are non-ambulatory due to their motor impairment and/or have cognitive impairment.[2]

Pain is the most prevalent secondary condition[3] and is a serious health concern[5] for children (0-17 years) with CP. Pain is an experience with sensory, emotional, cognitive, and social components.[4] Persons with CP often experience pain from several sources,[5,6] including the movement disorder itself, co-morbidities, musculoskeletal problems, and frequent exposure to painful procedures, including surgery.[2,5-7] As such, pain is frequently experienced as a combination of different types of pain, each with its own etiology, characteristics, intensity, location, duration, frequency, and interference with activities and function. Children with CP and their parents consider pain a significant burden.[8] Pain negatively influences daily activities,[9] and its interference with daily functioning (physical, mental, cognitive, and social) and sleep is a more important determinant of the perceived pain burden than the experience (eg, intensity) itself.[8,10] Despite this, pain interference and functional disability from unmanaged pain have received relatively little attention in the literature compared with pain prevalence based on intensity ratings.[11] Existing guidelines for assessment and management of CP in this age group, such as those from the National Institute for Health and Care Excellence (NICE), acknowledge the pain problem and provide a template for pain queries.[12] Yet, assessments of perceived intensity or behavioral signs associated with pain as suggested by the guidelines only provide partial representation of the complex pain experience.[13] Sensory aspects other than intensity (ie, frequency, duration, and quality), cause of the pain, the influence of emotions, thoughts, or social factors on pain and its burden, and management of pain are also rarely addressed and lacking in the literature.[11] Existing studies are not designed to be comprehensive in scope, and measurement and key features are emphasized and measured differently in different studies across different samples. Longitudinal studies on pain in children with cerebral palsy are also minimal. One Canadian study of 148 outpatient children with CP found that pain trajectories differed between participants. Pain frequently changed over time, and these changes were influenced by the etiology of the pain.[9] A more complete understanding of childhood pain in CP remains lacking, making it difficult to get a conclusive picture of overall burden and practice guidance to reduce this significant burden.

Although multiple studies have addressed it, pain prevalence remains undetermined in children with CP. Reports vary from 14% to 76% due to the heterogeneity in this group, differences in samples, recall periods, and the operationalization of pain. In one cross-sectional study (N = 280) of children with CP, 67.1% reported acute pain (ie, in the past week) and 31.4% reported chronic pain (ie, lasting more than 3 months).[14] In comparison, the 2007/2008 Canadian Community Health Survey found the prevalence of chronic pain in typically developing adolescents (12-17 years) to be 2% in males

and 6% in females.[15] In the adolescent part of the Trøndelag Health Study (Young HUNT),[16] researchers found that 19% of girls and 11% of boys reported having pain more than once a week.[17] Although the results from these studies are not directly comparable, they indicate children with CP are at increased risk for pain compared with their typically developing peers.

Pain is more prevalent with increased age,[18,19] severity of motor impairment,[20,21] in females,[22] and individuals with spastic-dyskinetic CP.[14] While age and sex have been identified as risk factors for pain across different pediatric populations,[23] risk factors associated with the CP diagnosis and the associated motor disability are unique for children with CP. Although some risk factors have been identified, replication of these findings in larger samples is needed. A larger and representative sample will also provide the opportunity to identify other risk factors as well as protective factors not previously identified. For example, an association between nutritional status and pain has been suggested, but not systematically evaluated.[24] In other pediatric populations, parental psychosocial factors have been shown to affect how parents assess and cope with their child's pain,[25] but data from children with CP are missing. In children with CP, unrelieved pain has been associated with decreased quality of life [26,27] and health-related quality of life,[28,29] decreased social participation,[30-32] mental health problems,[28,33] sleep problems,[18,34] and limitations in performing normal daily activities.[18,34,35] In a recent systematic review, McKinnon [11] concluded that a majority of these associations remain inconclusive. Consequently, they need to be further evaluated in a larger and representative sample that will enable detailed analyses on a sub-group level.

Compared to their typically developing peers, children with severe CP, in particular, frequently display indicators of pain that are ambiguous, idiosyncratic, and diminished.[36] In addition, ongoing unrelieved pain may diminish observable signs of pain [37] and impede the situation further by making the pain even more difficult to detect. Parental assessment is considered a cornerstone in the assessment of pain, as the parents know their child and his or her unique expressions of pain best. This viewpoint is also present in existing research, as studies primarily rely on parental proxy report,[11] although children and their parents may experience the child's pain situation differently.[8] There appears to be a consistent bias toward underestimation of pain intensity in children in general[38,39] and in children with CP specifically[7] compared with the children's own rating. Other aspects of the pain burden, such as pain interference, have not been addressed. There are some studies comparing child and parent report on mental health, health-related quality of life, and their association with the child's pain.[28,40] Still, a broader understanding of differences between parent proxy and child self-report across all aspects of the pain burden and the influence of child, parent, and contextual factors on the accuracy of parental assessment is needed.[11]

Pain is undermanaged in children with CP.[5,41] Clinicians report uncertainty in how to identify and manage pain [5,42] and evidence on effective pain management strategies is limited.[43,44] Knowledge on the strategies children and their parents use to manage pain and the helpfulness of those approaches as well as social and professional support in managing the child's pain is lacking. Parents' experiences with pain treatment in specialist health services have only been briefly described,[45,46] and no studies have been identified addressing primary care, although these children live their life and receive a substantial proportion of the help they need from community-based healthcare services.

More recently, the COVID-19 pandemic has brought substantial changes to our everyday life and to the healthcare services. Treatments for chronic pain have been interrupted.[47] These interruptions in health care in general may negatively affect pain, disability, and mental health for individuals living with chronic conditions.[48] In turn, it may also negatively affect parents or other family members. In the face of social distancing and its limitations concerning face-to-face consultations, the use of telemedicine and eHealth interventions has increased. However, the effect of this change in treatment approaches is unknown.[47] As such, an exploration of the impact of the current pandemic on pain and access to health care for children with CP is needed.

In response to the many challenges described above, this study will represent the most comprehensive and largest pain survey in the CP pediatric population to date. The CPPain study will provide a broad evaluation of different dimensions of pain, and individual and social factors associated with pain. First, it will provide a comprehensive description of the pain experience (ie, intensity, duration, location, quality, frequency of pain episodes), pain interference with activities of daily living and participation, and current pain management approaches for children with CP. Second, the study will describe and compare pain experience and interference from both children's and parents' perspectives whenever possible and between children with CP and siblings. The planned repetition of the survey will provide longitudinal data, which are currently limited. These findings are also intended to serve as the foundation for a planned intervention to address the pain burden of children with CP. As such, this survey will facilitate a transition from the current situation toward our vision of every child with CP experiencing his or her pain as sufficiently managed and able to enjoy life.

2 | METHODS

2.1 | Study design

The CPPain survey employs a cross-sectional survey design that spans across different levels of care (home, municipality, local, and specialty hospitals) and countries (Canada, Finland, Norway, Sweden, and the United States). It is conducted as part of the multidisciplinary CPPain research program (2019-2025) (Figure 1), acting as a baseline evaluation. It will also serve as part of a later outcome evaluation. The main idea behind the CPPain program is that children with CP and their parents have the best knowledge of the child's pain burden and need to have a key role in describing and defining the problem, devising and implementing solutions, and evaluating their effect in close collaboration with researchers and clinicians.

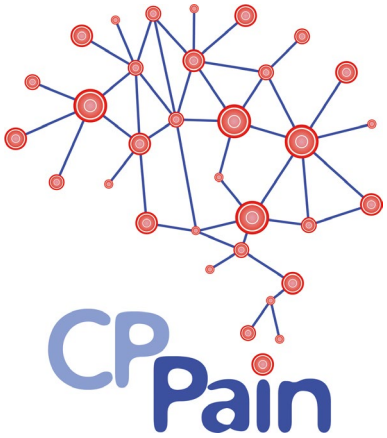

FIGURE 1 CPPain Logo

2.2 | Aims and objectives

The aim of the CPPain survey is to examine pain burden in children with CP. The primary objectives and hypotheses of the study are to:

1. Describe and compare pain burden in children with CP across different levels of motor disability and CP sub-types. The large international sample will enable stratification by Gross Motor Function Classification System (GMFCS) (levels I-V)[49,50] and CP subtype (spastic hemiplegic CP, spastic diplegic CP, spastic quadriplegic CP, dyskinetic CP, ataxic CP, and other/unspecified CP).[51] It is hypothesized that a) these children are living with a considerable pain burden, and b) pain characteristics and interference (eg, the magnitude of the burden) differ significantly between different levels of motor disability and CP sub-types.
2. Explore how the different aspects of the pain burden correlate. It is hypothesized that a) there will be a moderate correlation between different aspects of the experience (eg, intensity, frequency, number of painful locations, duration) and b) between aspects of the experience and pain interference.
3. Explore associations between child and parent individual and social factors and different aspects of pain burden in children 0-17 years with CP. It is hypothesized that previously identified risk factors (eg, age, gender, CP subtype, degree of motor disability) and other factors not previously explored in this population (eg, additional difficulties, co-morbidities, history of painful procedures and surgeries, spasticity treatment, access to and satisfaction with pain care, socioeconomic factors, and parental pain status and coping skills) will significantly correlate with aspects of pain burden.
4. Compare pain burden between adolescents with CP (12-17 years) and an age- and context-matched control group (typically developing siblings). There are three comparison groups: self-reporting adolescents with CP, parent report for adolescents unable to self-report, and self-report from siblings (controls). It is hypothesized that (a) pain burden will be significantly higher in children with CP compared to the control group, and (b) significantly higher in adolescents unable to self-report compared with self-reporting adolescents with CP and controls.
5. Compare self- and proxy-reported pain burden in verbal adolescents 12-17 years with CP compared with parent proxy report. It is hypothesized that (a) parents will underestimate pain burden compared with children's own ratings, (b) the difference in child and parent scores will increase with increasing severity of the pain burden, and (c) parent pain status and child/parent psychosocial factors will predict some of these differences.
6. Explore current management of pain and access to pain care. Current management of pain includes the use and perceived effectiveness of self-management strategies, medications, and treatments (eg, botulinum toxin injections, surgery), and complementary and alternative methods (CAM). Access to pain care was defined as participants' personal and professional network and the perceived helpfulness of these connections. Effects of the COVID-19 pandemic on children with CP's situation and access to care will be considered. It is hypothesized that (a) higher perceived effectiveness of self-management strategies, medications, or treatments will be positively correlated with a lower pain burden, (b) the use of CAM will be negatively correlated with perceived effectiveness of medications and treatments, (c) a stronger personal and professional network (number of actors and strengths of connections) will be positively correlated with a lower pain burden, and (d) the COVID-19 pandemic will have disrupted access to pain care and resulted in an increased pain burden. All analyses will control for age, sex, and degree of motor disability.

2.3 | Sample and setting

The primary study population includes children younger than 18 years (no lower age limit) with a CP diagnosis from Canada, Finland, Norway, Sweden, and the upper mid-western USA. Participants will be identified and recruited through national registries (if available), patient lists from participating institutions, user organizations, and social media. Each primary participant (ie, the child with CP) can generate between one and five respondents, including themselves, 1-2 parents or guardians, and 1-2 siblings (12-17 years). The survey will be distributed to approximately 4000 children and their families. Based on the average survey response rate, a 35% response rate is expected,[52] meaning that about 1400 primary respondents will be included in the study. Parents of all included children with CP will provide proxy report, regardless of whether the children themselves are able to provide self-report. Children with CP 12 years or older and cognitively able (approx. 50%) and siblings will be invited to provide full self-report. Children with CP 8-11 years of age and cognitively able, as well as older children with CP unable to complete the full survey, will provide an abbreviated self-report.

2.3.1 | Sample size estimation

There is no existing measure of pain burden. Children find pain interference the most important aspect of pain burden;[8] therefore, pain interference measured as a sum score (0-120 points) of the modified short form of the Brief Pain Inventory (mBPI-SF)[53] will be our main outcome variable. Based on two previous studies,[20,54] a standard deviation of approximately 25 points is expected, and a 10 point difference in score will be considered clinically important. Using 80% power and a type I error rate of 5%, 131 participants will be needed in each group when comparing three groups (eg, high, medium, and low pain interference) and 98 participants will be needed in each group when comparing two groups (eg, cases and controls).[55] With an expected sample size of 1400 primary respondents, the study will have enough power to detect differences in pain interference scores between groups and between self- and proxy-reported pain burden on a subgroup level (gross motor function levels, nationality, age, and gender).

2.4 | Survey development

The authors first identified the different dimensions to include in the survey from a scoping search of existing literature. From that literature and the collective experience in the research group, we decided that the survey needed to address the following dimensions: demographic and diagnostic information, the experience and expression of pain (location, duration, frequency, quality, intensity, behavioral signs), sources of pain, pain interference, pain coping, and pain management. We also decided to include dimensions known interfere with pain such as activities of daily life, nutritional status, participation, mental health, and health-related quality of life. These key aspects of the pain burden and interfering factors are described in the introduction.

To help ensure a child-centered perspective, we decided early to collect responses from both the children themselves and their parents whenever possible. At the same time, we wanted child and parental answers to be comparable. Consequently, only questionnaires that were available in both a proxy report version and a self-report version were eligible for inclusion in the survey. We can provide an overview of the questionnaires that were evaluated for inclusion in the survey upon request. With this initial framework as a starting point, the research group met in January 2019 and reached consensus on survey constructs. A set of published and previously validated measures to survey each dimension was decided upon, except for: (a) the dimensions "Demographic and diagnostic information" and "Pain management," where to the research group could not identify suitable measures, and (b) the dimension "Pain experience" where the measure selected by the research group needed to be further refined and adapted. Due to the COVID-19 outbreak, a fourth study-specific questionnaire was added later, which will aim to address the impact of COVID-19 infection and the pandemic on pain and access to pain care. For an overview of the dimensions addressed in the CPPain survey and the associated questionnaires, see Table 1.

2.4.1 | Translation and initial validation of translated versions of existing measures

Most of the selected measures were only available in English. After securing the necessary permissions from the scale developers or copyright holders, these measures were translated into Norwegian, Swedish, and Finnish using a standardized back-translation approach.[56] The translated versions of previously validated measures were further evaluated in cognitive interviews with members of the target population to determine content validity,[57] using the approach described in Andersen et al.[58]

2.4.2 | Development of study-specific measures

All four study-specific measures were developed through an iterative process, where drafts were circulated in the research group and discussed with user panels to reach consensus on preliminary versions. Whenever possible, existing measures were used as a starting point. The extended demographic and diagnostic information questionnaire is based on the standardized measures and variables included in the Norwegian Quality and Surveillance Registry for Cerebral Palsy (NorCP).

To measure the different aspects of the pain experience, the Dalhousie Pain Interview (DPI) initially developed by Breau and colleagues was further refined.[6] The DPI was originally developed as a researcher-administered interview to be conducted with a parent or caregiver of an individual with intellectual and/or developmental disability. The DPI has been used previously in studies in this population.[20,54] Specifically, it has been used to (a) study parent-reported pain in a small sample (n = 34) of children with CP to determine the relationship between motor function and musculoskeletal pain[20] and (b) assess concurrent validity of the modified Brief Pain Inventory (mBPI) as a proxy-reported tool in a heterogeneous sample of children with CP (n = 167).[54] Within this project, the DPI was adapted to an electronic questionnaire format rather than an oral interview. A section on self-management of pain was added, based on a questionnaire developed for use in the It Doesn't Have to Hurt Campaign.[59] From this revised proxy version of the DPI, the child self-report version was developed and additional questions assessing quality of pain (eg, shooting, burning) previously identified in an adult CP study [60] were added to the self-report version.

The questionnaire on pharmacological management of pain used a questionnaire developed by Tutelman and colleagues for use in a pediatric cancer population as a starting point.[61] To this, questions addressing common treatments for CP (eg, botulinum toxin injections), the influence of these treatments on pain, the personal and professional network surrounding the child and family, network members' role in

TABLE 1 Dimensions, associated measures, and their content

Dimension	Questionnaire/source	Content
Demographic and diagnostic information	Diagnostic and demographic (SSQ) Register data*	Age, gender, motor and cognitive function, associated difficulties, communication skills, socioeconomic factors
Pain experience/Self-management of pain	mDPI[6] (adapted)	Pain location(s), cause(s), duration, frequency, and intensity, quality** and self-management of pain (strategies/effectiveness)
Expression of pain	PPP[68]***	Behavior associated with pain
Pain management /pain care	Pain Management (SSQ)	Pharmacological pain management, professions involved, and the use of complementary and alternative approaches
Pain coping	PCS[69,70]	Negative attitudes toward pain (pain catastrophizing)
Pain interference	mBPI-SF[53]	Pain interference with activities and affect
Activities of daily life	CPCHILD[71]	Difficulties associated with personal care and mobility
Nutritional status	Screening tool[72]	Feeding/swallowing difficulties and undernutrition
Participation	CASP[73,74]	Participation in activities at home, at school and in the community. Supportive strategies, assistive devices, or modifications used
Mental health	RCADS[75]	Mental health, specifically anxiety, and depression
HQoL	KidScreen-10[76]	Generic health-related quality of life index and overall health
COVID-19	COVID-19 and pain (SSQ)	COVID-19 infection and pain. Effect of the pandemic on pain and access to care

Note: HQoL = Health-related Quality of Life, *from NorCP (Norway). For other countries (USA/Canada), comparable information is collected in an extended demographics form. **Self-report only. ***Proxy report only. SSQ = Study-Specific Questionnaire; mDPI = Modified version of Dalhousie Pain Interview; PPP = Paediatric Pain Profile; PCS = Pain Catastrophizing Scale; mBPI-SF = Modified Brief Pain Inventory-Short Form; CPCHILD = The Caregiver Priorities and Child Health Index of Life with Disabilities; CASP = Child and Adolescent Scale of Participation; RCADS = Revised Children's Anxiety and Depression Scale.

pain management and care, use of complementary and alternative approaches, and access to pain education were added.

The questionnaire on COVID-19 and pain was designed to address the impact of the pandemic on key aspects of the pain constructs explored in the survey (experience, interference, management, and access to care).

2.4.3 | Survey versions

The survey will be available in all study languages in three different versions: (1) parent proxy, (2) self-report—full (age 12-17), and (3) self-report—abbreviated (age 8-11 and older children with cognitive limitations). The content of the proxy and full self-report versions of the survey is described in Table 1. The abbreviated version includes a small selection of key questions addressing their most troublesome pain (intensity, frequency, and location), pain interference, management of pain, and health-related quality of life.

2.4.4 | Data collection tool and study database

The common study database for all participating countries is located in Norway. The Clinical Trial Unit (CTU) at Oslo University Hospital has set up the online survey in Viedoc™ (www.viedoc.com) in accordance with their procedures. Identical electronic Case Report Forms (eCRFs) were first developed and piloted in English, before they were translated into Norwegian, Swedish, and Finnish. The eCRF system is General Data Protection Regulation (GDPR) and U.S Food and Drug Administration (FDA) Code of Federal Regulations 21 Part 11-compliant. Each eCRF has been extensively tested both by members of the research group and end-users to eliminate errors and ensure its utility. Corresponding paper-based forms have been developed for those participants who request a paper-and-pencil-version.

2.5 | Study procedure

2.5.1 | Recruitment

In Norway, participants will primarily be recruited from the Norwegian Quality and Surveillance Registry of Cerebral Palsy (NorCP). NorCP is a national registry monitoring the prevalence and severity of pediatric CP and has coverage of >90%. In the upper Midwest USA, participants will primarily be recruited from the patient lists of Gillette Children's Specialty Healthcare, in St. Paul, Minnesota. In Canada, participants will be recruited through the Canadian CP-registry and pediatric habilitation centers. In Sweden and Finland, recruitment will be done through pediatric habilitation centers and other relevant organizations (eg, neuropediatric professionals). In addition, the study

will be advertised through patient organizations (eg, the Norwegian CP Association, CP Canada Network, the Finnish CP Association, CP-Sweden) and social media. For each primary participant (eg, the child with CP), up to four secondary participants may be recruited (1-2 parents and 1-2 siblings). The data collection is estimated to take between 6 and 12 months. Each country will have a designated contact person for interested parties. Each national investigator will manage recruitment, consent, and data collection in his/her country.

Parents/guardians will receive an information letter about the study and provide consent for participation. In the Nordic countries (Finland, Norway, and Sweden), both parents/guardians will have to consent to the child's participation and adolescents >15/16 years have to provide their own consent to participate. In addition, adolescents with CP >15/16 years will also have to consent to their parents' participation, unless they are unable to give informed consent. In North America (Canada and USA), consent from one parent/guardian is sufficient for all children <18 years. Non-responders will receive two reminders 3-4 and 7-8 weeks after the initial information letter.

2.5.2 | Data collection

Pseudo-anonymized survey data will primarily be collected online using Viedoc™ (www.viedoc.com). Participating countries will also have the option to offer a paper-based alternative. Data from paper-based forms will be manually entered into Viedoc by the national collaborator or a designate. All participants within a family will receive their individual login information to the corresponding version of the survey. When first logging into the survey, the participant confirms participation before commencing on the survey. It is estimated that the surveys will take between 20 and 50 minutes to complete, depending on the child's pain situation. Importantly, it is estimated that unlike the other survey versions, the child abbreviated form will take <10 minutes. Participants will be able to take breaks, but will be encouraged to complete the survey within one week. Family members will also be encouraged to complete their versions of the survey within a week to strengthen comparisons. If the child requires assistance to fill in the survey, it will be noted that the assistant should preferably be someone other than the parent providing the proxy report. The survey will be open for 6 weeks after the first login. Both the modified version of DPI and translated measures will be further validated by re-administering these parts of the survey to respondents who consent to participate in a validation study nested into the CPPain survey. We will use a staggered approach to data collection, starting in Norway Q4-2021. We estimate that the data collection period in each country will be 6-8 months and that the entire data collection will be completed Q4-2021.

2.5.3 | Reminders

Viedoc will send two automated reminders, 14 and 28 days after the respondent first logged into the survey. If the respondent has not provided a mobile phone number or an email address or has requested a paper-based survey, reminders will be handled manually and sent by regular mail. If the respondent fails to login to the survey, a manual reminder will be sent to inquire whether they need more time to respond or want to withdraw from the survey.

2.6 | Analysis plan

Data will be exported from Viedoc as Excel files, imported into SPSS, and combined into a master file for analysis. Data transfer and collaboration agreements are already in place that regulate collaboration on data analysis and publishing of results. Data transfer will be in accordance with GDPR regulations. Data will be analyzed using appropriate statistical methods for comparing groups (self-reporting children with CP vs parents or siblings, different degrees of disability or pain burden, etc) and to study the personal and diagnostic predictors associated with low, medium, and high pain burden. Stratification will be used to investigate subgroups (gender, age groups, degree of motor disability, subtype of CP, etc), and analysis will be adjusted for appropriate sociodemographic variables. SPSS will be the main tool for statistical analysis. Other specialized software, such as R, will be applied as needed. The master file will be stored for 5 years after publication of results. Each national group will retain full access to and control over their own data. In addition to the publications planned based on the entire database, national investigators will be able to publish sub-studies based on national data as long as it does not interfere with any main publications.

3 | DISCUSSION

Pain is a significant burden[8] and a serious health concern[62] for children and adolescents with CP, and there is an urgent need for better pain management in pediatric CP. In response to this challenge, the aim of this study is to gain a comprehensive picture of the current situation by surveying a large and partly population-based sample across several countries. The comprehensive data collection will provide a broader knowledge base concerning their pain burden than is currently available, and as such, increase our understanding of their pain. More importantly, it will lay the foundation for systematic practice change as the first component of a multi-step process toward the development and implementation of patient-centered interventions to reduce the pain burden in young people with CP. The planned intervention will move the current situation toward our vision of every child with CP experiencing his or her pain as sufficiently managed, and able to enjoy life.

3.1 | Strengths

The major strengths of the CPPain survey will be its scope and size, enabled by the multi-national approach, the diverse research group, and extensive user involvement.

Pain is always a complex experience, but even more so in this population.[43] The CPPain survey will broadly address pain burden and how it is managed, while existing studies primarily have taken a more narrow view,[11] focusing on pain intensity as a representation of this complex experience. Moreover, a child-centered perspective[63] will be taken in this study by including child self-report whenever possible. When addressing pain in this population, children's views have often been overlooked, but to comply with the rights of the child, we need to include their perspectives and preferences.[64] The research group has attempted to select and design survey versions that are appropriate for children themselves and that still mirror the caregiver questionnaires. Further, an abbreviated version of the child survey will be available to ensure that children who cannot answer the full version due to age or cognitive difficulties still get the chance to express their views. Such a tiered approach to obtaining the self-reported pain experience of children with CP has not previously been accomplished.

The multi-national collaboration and estimated large study sample size will also be unique. Collaboration across several countries is needed to obtain a sample of this size, and the large sample size and comprehensive data we collect will allow for more broad information and detailed sub-group analyses, as well as identifying trends in pain by different CP status (GMFCS level and CP subtype), age, and gender. The importance of a large enough sample cannot be overstated. Children with CP are a highly heterogeneous group and by averaging responses across smaller samples, important differences remain undetected, and these differences may be of vital importance for determining treatments and managing pain.

Understanding and managing pain require a multi-professional approach where the patient voice is a key part of the team.[65,66] In addition to including the lived experience, our research group is diverse concerning professional backgrounds and clinical and research experience. The group has engaged in an extensive process to compile the survey, including the development of study-specific questionnaires where no suitable measures were identified, the systematic translation and cultural adaptation of all measures into all study languages, and the comprehensive testing of the online version of the survey to confirm its feasibility.

To ensure further that the survey is representative of both lived experiences and the different contexts in which pain is experienced, including available treatment services, two advisory panels, one user panel and one clinician panel, were consulted regularly during the development of the survey. While the user panel consists of individuals with CP and parents of children with CP, the clinician panel consists of clinicians with different backgrounds from both the hospital and the community setting. The research group believes this will help ensure that respondents perceive the survey as timely and relevant and that it reflects the reality concerning systems and services.

3.2 | Limitations and biases

Despite the strengths described above, the CPPain survey has limitations, including susceptibility to selection bias, attrition bias, and recall bias. In addition, the survey will be launched during the COVID-19 pandemic.

While part of the study sample will be population based (Norway), it is not possible to eliminate selection bias due to the inclusion of a non-probability sample in the other participating countries. The selection of participating countries is a practical choice, based on where key members of the research group are located. However, the study has put into place measures to help address this and other biases that may arise from the study, including diverse recruitment strategies and alternative survey administration methods. The average response rate for surveys is 33%.[52] To boost the response rate, a comprehensive system of information and reminders has been set up. Users involved in the survey development have expressed that this survey addresses an important issue for them and one they have a stake in illuminating, meaning participants will be more likely to both consent to participate and actually complete the survey compared with the average survey.

Including siblings as a control group may be considered a limitation. On the one hand, it can be argued that being the sibling of a child living with a disability, they are not representative of children in general. On the other hand, by including siblings as the control group, we have ensured that both cases and controls live in a similar social and socioeconomic environment, and we know that social factors have a large influence on the experience and expression of pain.[67]

Finally, the COVID-19 pandemic has brought about major changes. Not only have parents experienced an increased care burden due to closing schools and childcare, there have been major changes in the provision of health care, where services have been closed or less accessible than before.[47] Little is known about whether and how thiese changes has affected pain experience or pain care for children overall and for children with CP in particular, but it is reasonable to assume that they have had an impact. Consequently, it is reasonable to believe that the ongoing pandemic will influence the answers to this survey. To address this issue and to learn more about the effects of the pandemic on pain care in this population, we have added a questionnaire specifically addressing this issue.

In summary, it is expected the CPPain survey will represent the largest and most comprehensive survey of pain conducted in children and adolescents with CP to date. The study will further our understanding of the pain burden children and adolescents currently live with, how the pain interferes with their life, how it is managed and the pain care these children and their families utilize. These findings will be the crucial first step toward the co-creation of an intervention together with children with CP and their parents to address their pain situation.

4 | ETHICS AND DISSEMINATION

This project complies with children's right to express their views and be taken seriously in matters concerning themselves. Participation is voluntary and both children and parents will be given enough information tailored to their individual level of understanding, to be able

to give informed consent/assent. Users will be closely involved in all phases of the project. The CPPain program and this survey were developed in collaboration with the Telemark chapter of the Norwegian CP association and a patient partner from their board participates actively in both the steering group and the research group.

The Norwegian Regional Committee for Medical and Health Research Ethics (REC South-East) (2019/618 and 11907) and the Research Ethics Board at the University of Minnesota in USA (STUDY00011274) have approved the study. Canadian, Finnish, and Swedish approvals are underway.

Findings will be presented in scientific publications and conferences, and the researchers will specifically target colleagues in the field through the Pain and Intellectual Developmental Disabilities Special Interest Group (PIDDSIG) within the International Association for the Study of Pain (IASP). In addition, findings will be communicated to users, clinicians, and managers in both primary care and secondary care throughout the project period. Dissemination channels will include the CPPain Web site (www.sthf.no/cppain), social and public media, meetings within the user or provider organizations, educational events, articles in user organization journals, and Web sites directed toward users and/or clinicians, such as Solutions for Kids in Pain (SKIP) knowledge mobilization network (www.kidsinpain.ca).

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