BMJ Open

Effectiveness and scalability of an electronic patient-reported outcome measure and decision support tool for family-centred and participation-focused early intervention: PROSPECT hybrid type 1 trial protocol

Vera Kaelin,[1,2] Vivian Villegas,[1] Yi-Fan Chen,[3] Natalie Murphy,[4] Elizabeth Papautsky [ID] ,[5] Jodi Litfin,[6] Natalie Leland,[7] Varun Maheshwari,[1] Beth McManus,[4] Mary Khetani [ID] ,[1,2,8] High Value Early Intervention Research Group[6]

For numbered affiliations see end of article.

Correspondence to
Dr Mary Khetani;
mkhetani@uic.edu

ABSTRACT

Introduction Early intervention (EI) endorses family-centred and participation-focused services, but there remain insufficient options for systematically enacting this service approach. The Young Children's Participation and Environment Measure electronic patient-reported outcome (YC-PEM e-PRO) is an evidence-based measure for caregivers that enables family-centred services in EI. The Parent-Reported Outcomes for Strengthening Partnership within the Early Intervention Care Team (PROSPECT) is a community-based pragmatic trial examining the effectiveness of implementing the YC-PEM e-PRO measure and decision support tool as an option for use within routine EI care, on service quality and child outcomes (aim 1). Following trial completion, we will characterise stakeholder perspectives of facilitators and barriers to its implementation across multiple EI programmes (aim 2).

Methods and analysis This study employs a hybrid type 1 effectiveness-implementation study design. For aim 1, we aim to enrol 223 caregivers of children with or at risk for developmental disabilities or delays aged 0–3 years old that have accessed EI services for three or more months from one EI programme in the Denver Metro catchment of Colorado. Participants will be invited to enrol for 12 months, beginning at the time of their child's annual evaluation of progress. Participants will be randomised using a cluster-randomised design at the EI service coordinator level. Both groups will complete baseline testing and follow-up assessment at 1, 6 and 12 months. A generalised linear mixed model will be fitted for each outcome of interest, with group, time and their interactions as primary fixed effects, and adjusting for child age and condition severity as secondary fixed effects. For aim 2, we will conduct focus groups with EI stakeholders (families in the intervention group, service coordinators and other service providers in the EI programme, and programme leadership) which will be analysed thematically to explain aim 1 results and identify supports and remaining barriers to its broader implementation in multiple EI programmes.

Strengths and limitations of this study

► The broad range of activities the Young Children's Participation and Environment Measure covers might support relevance of the tool across a variety of early intervention (EI) eligible families.

► Continuous stakeholder engagement and site collaboration may strengthen intervention quality, protocol adherence and completion of the planned research.

► Due to the COVID-19 pandemic, caregivers might be better able to appraise their child's participation at home as compared with community.

► Due to the COVID-19 pandemic, there might be more challenges recruiting families due to increased challenges with accessing EI services.

► Limiting the inclusion criteria to English-speaking families may limit generalisability of the results to a subset of families that are enrolled in EI.

Ethics and dissemination This study has been approved by the institutional review boards at the University of Illinois at Chicago (2020-0555) and University of Colorado (20-2380). An active dissemination plan will ensure that findings have maximum reach for research and practice.
Trial registration number NCT04562038.

INTRODUCTION

Early intervention (EI) endorses a family-centred approach to designing and enacting the individualised family service plan (IFSP). According to a family-centred care approach, families are formal members of their child's EI team and are expected to engage in shared decision making about the design and enactment of their child's EI service plan.[1 2] Family

engagement in shared decision-making can be achieved by intentionally leveraging family expertise about their care priorities and potential supports, barriers and strategies for goal attainment.

Electronic patient-reported outcome (e-PRO) measures are relevant tools to engage families in the EI service context, as they offer a means to systematically collect and integrate the family perspective about service quality and outcomes to supplement routine EI practitioner assessments.[3] Additionally, EI programmes with electronic data capture systems can use e-PROs to: (1) expedite and expand provider reach in assessing the needs of individual families and (2) aggregate data across families to examine trends in EI service quality and outcomes over time for continuous quality improvement of EI services.[4–6]

The development of the IFSP at the child's annual meeting is the earliest and one of the most meaningful opportunities to foster family engagement using patient-reported measures.[7 8] The IFSP gets developed following EI eligibility determination. The service coordinator first conducts a family assessment via interview; then, the service coordinator, family, and EI provider use the results of the eligibility evaluation and family assessment to develop the IFSP, a written document listing the EI services (eg, occupational therapy, physical therapy) that will be provided and the target functional outcomes of interest. This IFSP is reviewed and revised on at least an annual basis during the entirety of the time the child is active in EI services. This process requires collaboration between families and providers to visualise a preferred future, develop mutually agreed on goals accordingly, and engage in shared decision-making to implement and evaluate intervention(s) for goal attainment.[7 8] The use of e-PRO measures can scale to multiple programmes within the broader EI service system, giving families an online venue to organise and communicate their expertise and share in decisions as a full member of their child's EI team.

The Young Children's Participation and Environment Measure (YC-PEM) is an evidence-based e-PRO that was developed with provider and family input.[9–11] It provides individual caregivers with a valid, reliable and feasible way to comprehensively communicate information about their child's current participation in valued activities and areas of unmet participation need.[12–15] The YC-PEM electronic patient-reported outcome (e-PRO) yields a brief summary of family responses in the form of a report that can be shared with EI team members ahead of the child's annual IFSP meeting. The information in this report can integrate with standardised processes used to facilitate IFSP meetings, thereby helping to further structure provider–parent communication around intervention priorities and goal attainment strategies.[16–19]

The YC-PEM e-PRO holds promise in improving EI service quality in two ways: (1) expediting and expanding EI provider reach for family-centred and participation-focused service design in partnership with individual families; and (2) aggregating the YC-PEM e-PRO data to examine trends in participation over time as a function of EI service use for groups of families.[13 20 21] It is therefore a promising electronic health systems intervention to strengthen the parent–practitioner relationship and equip programmes with a common data element for conducting robust patient-centred outcomes research to drive quality improvement efforts.[22 23] Yet, it is not known, whether the YC-PEM e-PRO is a valuable option for implementation within the context of a routine EI workflow.

The Consolidated Framework for Implementation Research therefore guided our foundational efforts to explore the trialability of implementing the YC-PEM e-PRO in an EI service context.[24] We conducted two single-arm pilot studies to examine the tool's feasibility, acceptability, and value when implemented into an EI clinical workflow.[13 14 21] We first tested the feasibility of the YC-PEM e-PRO within a small university-affiliated EI programme. Results indicated that the YC-PEM e-PRO could detect families' desire for change in their child's participation, and aggregate data could be used to show a significant association between EI service intensity and the child's level of participation in valued home activities.[13] However, feasibility was mixed, and EI service providers suggested that the YC-PEM e-PRO be embedded in a child's annual IFSP meeting to improve its acceptability.[13] We then transitioned to a larger clinical context (ie, a large non-university affiliated EI programme) to examine the feasibility of the revised use of the YC-PEM e-PRO, integrating feedback from the prior study. For this second pilot study, caregivers completed the YC-PEM e-PRO in preparation for their child's IFSP meeting. Preliminary evidence supported the feasibility (80% completion rate; mean completion time of 21.3 min vs 45–120 min for usual care), acceptability (>50% of families found it helpful, regardless of caregiver education or family income), and value of the YC-PEM e-PRO option (64% of families viewed their responses to share with other members of their child's EI team).[14]

Results of these prior two pilot research phases suggested the need to further tailor this intervention, strengthen stakeholder engagement, and plan for more rigorous scientific methods to build its evidence base.[13 14 25 26] Therefore, for this current protocol, we adapted the YC-PEM e-PRO to the EI service context in two ways. First, the YC-PEM e-PRO was paired with a program-specific decision support tool to facilitate integration of results during the IFSP meeting. Second, the YC-PEM e-PRO instructions were enhanced to introduce users to the concept of participation and the importance of personal norms when appraising it, which differs from more routinely used norm-based assessments of developmental and functional skills.[27 28] Increasing stakeholder engagement was a result of lower feasibility rates and stakeholder feedback in prior research.[25 26] We, therefore, identified and sponsored a group of EI providers, specifically service coordinators, to champion the work by engaging them in research protocol development and YC-PEM e-PRO implementation.[26] To enhance scientific

rigour, this protocol introduces a control group and longitudinal data to interpret the effectiveness of the intervention, and a mixed-methods component to characterise salient supports and barriers to its longer-term implementation as a health systems intervention across multiple EI programmes.[28]

Objectives

Our overarching objective of this project is to ascertain the value of implementing the YC-PEM e-PRO within a routine EI context. This will be achieved with the successful execution of two aims. Aim 1 will evaluate the effectiveness of the YC-PEM e-PRO option, when paired with a decision support tool, on valued outcomes of EI service quality (parent perceptions of family-centredness of services, parent activation for shared decision-making, and parent engagement in service plan implementation), service focus, and child developmental and functional skill gains. Aim 2 will characterise EI stakeholder perspectives of facilitators and barriers to implementing the YC-PEM e-PRO as a valued option across multiple EI sites.
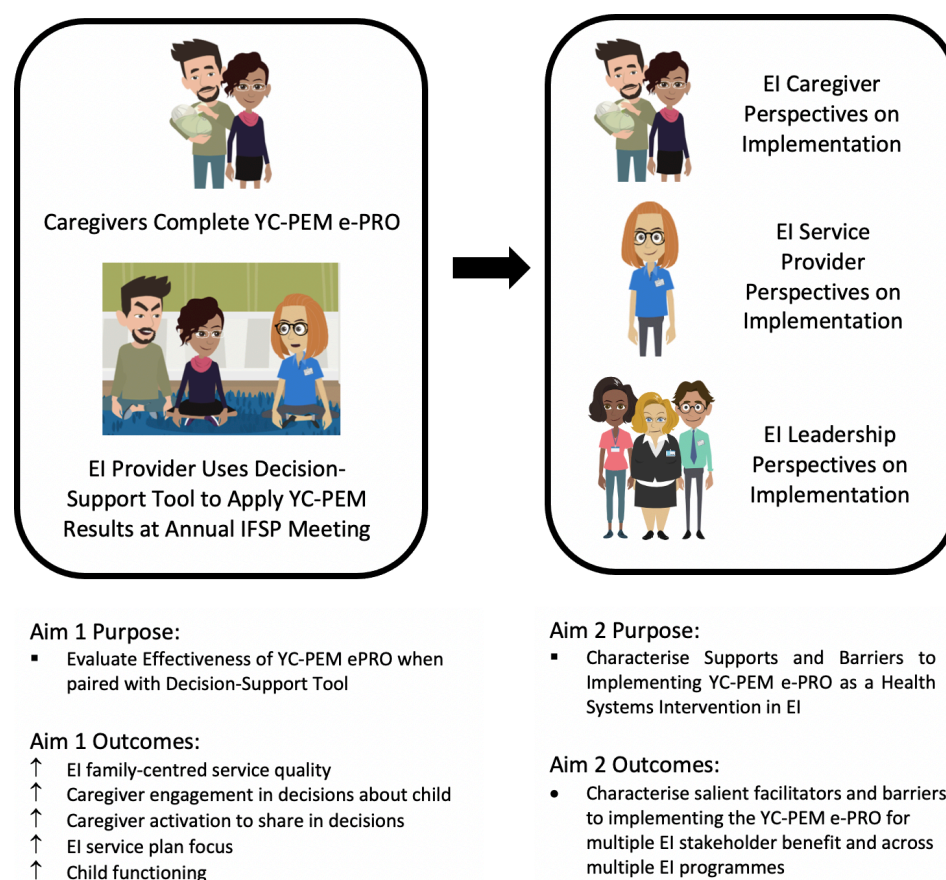
For aim 1, we hypothesise that as compared with usual care, intervention participants (ie, caregivers) will (1) report greater satisfaction with enabling and partnership (primary outcome; H1); (2) report greater general and specific information exchange within the EI team for shared decision-making; coordinated and comprehensive EI care; and respectful and supportive EI care (secondary outcome; H2); (3) report higher engagement and activation for shared decision making to design the EI service plan (secondary outcome; H3); (4) receive a greater breadth and intensity of participation-focused EI services (secondary outcome; H4) and (5) their children will demonstrate greater gains in developmental and functional skills (secondary outcome; H5).

METHODS AND ANALYSIS
Study design and setting

We will use a hybrid type 1 effectiveness-implementation study design to achieve two specific aims (see figure 1). In aim 1, we will conduct the Parent-Reported Outcomes for Strengthening Partnership within the Early Intervention Care Team (PROSPECT) community-based pragmatic trial, to test the effects of the YC-PEM e-PRO and a decision support tool intervention. The study protocol for aim 1 adheres to the Standard Protocol Items: Recommendations for Interventional Trials guidelines, as recommended for reporting of clinical trial protocols.[29] EI stakeholder perspectives guided the design of a PRagmatic Explanatory Continuum Indicator Summary 2 (PRECIS-2) Wheel describing the pragmatic trial design (see figure 2).[30 31] In aim 2, we will gather stakeholder

Aim 1 Purpose:
- Evaluate Effectiveness of YC-PEM ePRO when paired with Decision-Support Tool

Aim 1 Outcomes:
- ↑ EI family-centred service quality
- ↑ Caregiver engagement in decisions about child
- ↑ Caregiver activation to share in decisions
- ↑ EI service plan focus
- ↑ Child functioning

Aim 2 Purpose:
- Characterise Supports and Barriers to Implementing YC-PEM e-PRO as a Health Systems Intervention in EI

Aim 2 Outcomes:
- Characterise salient facilitators and barriers to implementing the YC-PEM e-PRO for multiple EI stakeholder benefit and across multiple EI programmes

Figure 1 Hybrid type-1 effectiveness-implementation study design. EI, Early Intervention; IFSP, Individualised Family Service Plan; YC-PEM e-PRO, Young Children's Participation and Environment Measure electronic Patient-Reported Outcome.
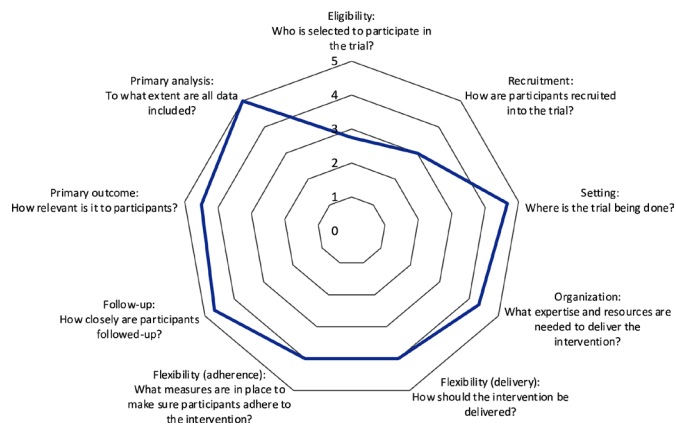
Figure 2 PRECIS-2 wheel for PROSPECT trial. PRECIS-2, PRagmatic Explanatory Continuum Indicator Summary 2; PROSPECT, Parent-Reported Outcomes for Strengthening Partnership within the Early Intervention Care Team.

perspectives on supports and barriers to implementation across multiple EI programmes. Aim 2 will use an explanatory sequential (quan >QUAL) mixed-methods design.[32] This hybrid effectiveness-implementation study design has the internal validity advantages of a randomised control trial, yet study results can be more readily implemented into 'real-world' programming.[33]

Note. PRECIS-2 has nine domains, each scored on a 5-point Likert scale (from 1=very explanatory to 5=very pragmatic).[30][31]

Eligibility criteria and recruitment

For aim 1 (see figure 3), we aim to recruit caregivers from a large, urban, and non-university affiliated EI programme. We started recruitment in October 2020 and plan to end
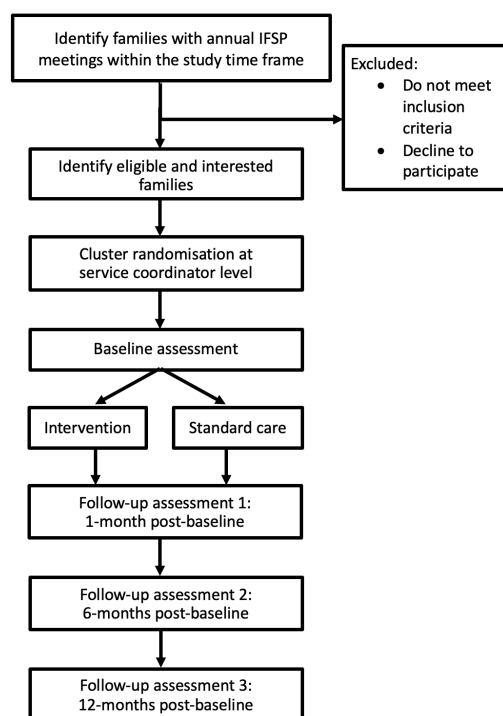
Figure 3 Aim 1 flow chart.

recruitment in December 2021, or when we reach target enrolment. Caregivers are included if they: (1) are at least 18 years old; (2) identify as the parent or legal guardian of a child enrolled in EI; (3) read, write and speak English; (4) have internet and telephone access; and (5) have a child 0–3 years old who has received EI for 3 or more months.

A designated EI staff member identifies and calls potential caregivers 4 weeks before their child's annual IFSP meeting. During this call, eligible and interested caregivers are directed to the project website where they identify their service coordinator, view a recruitment flyer, and create a user account to enrol in the project. After account creation, each caregiver confirms their eligibility, provides consent and HIPAA authorisation, and completes the baseline measures online via REDCap electronic data capture tools hosted at the University of Illinois at Chicago.[34][35]

To support participant recruitment, participants who verbally consent to participate are reminded via email on a weekly basis, with up to three reminders. Three strategies are used to support participant retention during follow-up phases: (1) automated weekly email reminders; (2) participant compensation increases to US$20 for completing surveys at 6 months and 12 months post-baseline; and (3) through the EI programme, families can apply for financial childcare support for their time needed for complete the surveys.

In aim 2, we will recruit three types of EI stakeholder groups: (1) families assigned to the intervention group during aim 1 and who indicated interest to participate in aim 2; (2) EI service coordinators involved in this trial (n=6) and EI service coordinators and practitioners that are not involved in this trial (n=6); and (3) EI programme leadership, including programme director, supervisors, and data and programme managers (n=10). EI service coordinators and leadership involved in this trial will be recruited by email, and we will use snowball sampling to expand enrolment to those employed by the EI programme but not involved in the PROSPECT trial.

Sample size

The aim 1 power analysis intends to maintain 80% power and total 5% type I error. We hypothesise that the change score of the primary outcome, the Measure of Processes of Care (MPOC) subdomain: enabling and partnership (H1), from pre-baseline to post-baseline at 1 month is approximately a medium Cohen's d effect size, that is, 0.5,[36] between the intervention and control groups. This effect size is close to an empirically large effect size for rehabilitation studies in a category of skills and habits.[37] Although a medium effect size is observed at 26 weeks by Fonvig et al,[38] our hypothesised effect size is based on comparing the intervention scope (targeting improvements in family-centred care during IFSP development) to prior work evaluating interventions that are broader in scope, and therefore, expecting to detect a larger effect in a shorter time frame. Considering the cluster

randomisation at the level of the service coordinator, a design effect according to Eldridge *et al*[89] is used to take account of the cluster dependence, various cluster sizes, and the small number of clusters by assuming an intraclass correlation coefficient of 0.02, a range of cluster sizes as 15–25, an average cluster size of 20, and 6 clusters. Using two sample t-test with the design effect, total 178 families, that is, 89 families per group and 30 families per service coordinator, would be required to detect the targeted effect size. Given an attrition rate of 20%, we plan to recruit total 223 families, that is, 111–112 families per group and 37 families per service coordinator.

Randomisation

To avoid contamination, aim 1 will employ a cluster-randomised design at the EI service coordinator level.[40] Six EI service coordinators who are members of the High Value Early Intervention Research Group and expressed interest in the project were initially randomised into intervention group (n=3) and control group (n=3), using computer generated randomisation. Participating caregivers are therefore directed to either the intervention or control group, based on their assigned EI service coordinator's group assignment.

To reduce cross contamination among service providers, only service coordinators allocated to the intervention group were included in co-creating the decision support tool to be paired with the YC-PEM e-PRO. They were asked to not share this information with service providers assigned to the control group. Additionally, the research team meets with intervention and control group service coordinators separately during aim 1 completion to discuss experiences and potential concerns.

Blinding

EI service coordinators fulfil the role of a case manager and coordinate evaluations, care planning and outcomes monitoring for families. EI service coordinators cannot be blinded to the intervention as it involves modifying their clinical practice, nor can researchers be blinded to participants' group allocation. However, the statistician will be blinded to participants' group assignment while conducting all analyses. EI service providers are not trained or provided knowledge about the intervention. Since EI services are provided in the child's natural environment (eg, home), there are fewer interactions across service providers and service coordinators during a routine workday as compared with a clinic environment.

Intervention
Technical specifications

The intervention is administered via a project website that pairs with REDCap. For this website, HTML5, CSS3 and AngularJs were used to create the frontend, and NodeJS was used to create the back-end of the intervention. On entering the project website, participants select their assigned EI service coordinator, create a user account and log in. From the main menu on login, assignment into the intervention or control group is automated based on EI service coordinator, and participants are routed to the applicable survey to be completed via REDCap.[34 35]

Both the intervention and control group workflows are enabled with the following functionality: (1) confirmation logics in REDCap when caregivers are reviewing inclusion criteria and providing informed consent and HIPAA authorisation and (2) a back-end portal that is password protected and accessed by research staff who are granted administrative access to monitor enrolment and completion rates, completion time, email and phone queries, and confirmation of PDF reports when sent to the EI team. Research staff access a dashboard that was created using NodeJs, Express server and EJS with MySQL(postgress) and used to store these data into a database. Parts external to REDcap are hosted by the Amazon EC2 server for hosting the Linux Ubuntu server. For security, server alerts are configured and port blocking is allowed based on IP address.

The intervention group REDCap project includes additional functionality, including: (1) skip logic functionality for YC-PEM e-PRO completion and (2) a progress bar indicating the number of pages left to complete, beginning with the demographics page. On completing the REDCap surveys, intervention group participants are directed back to the main menu to access an online summary report of their YC-PEM e-PRO responses. The summary report is populated via REDCap application programming interface using Angularis and Kendo libraries. The summary report uses bar graphs to represent the data, provides options to customise it with the child's name and photo, and can be downloaded and saved as a PDF. This report is automatically sent to the EI team.

User experience

The YC-PEM e-PRO contains 13 types of home activities in four categories, and 11 types of community activities across four categories (eg, personal care management is a type of home activity within the category of basic care routines).[12 15 41] For each activity, caregivers are asked about their 1) child's participation frequency (from never=0 to daily=7), (2) child's level of involvement (from not very involved=1 to very involved=5) and (3) desire for their child's participation to change (yes, no). When caregivers indicate 'yes, desire change' for one or more activities in a category, they are prompted to describe up to three strategies that they have used to support their child's participation in that category. After completing the participation items for a YC-PEM e-PRO section (eg, home), caregivers are asked to rate their perception of how environmental features and resources (eg, sensory qualities, equipment and supplies) impact their child's participation in that setting (from usually makes it harder=1 to usually helps=3).[12 15 41] The information in this YC-PEM e-PRO summary report is systematically referenced by the EI service coordinator when administering a companion guide to support shared decision-making during the child's annual IFSP meeting.

This shared decision support tool, consisting of the IFSP template form with added prompts and placeholders for the service coordinator to transfer and build on information contained in the YC-PEM e-PRO summary report, was co-created with service coordinators assigned to the intervention group. The prompts and placeholders provide service coordinators with structure for consistently integrating YC-PEM e-PRO results into the IFSP meeting guide to elicit and focus family priorities in the context of EI service provision.

Intervention fidelity

Participating EI service coordinators have either a bachelor's degree in early childhood education and/or at least 5 years of related experience. They joined the High Value Early Intervention Research Group by completing an online research training programme (95–135 min), consisting of online learning modules delivered asynchronously, followed by a simulation experience whereby service coordinators review and contribute to a research product.[26] For this project, all EI service coordinators in the research group were invited to a 1-hour training for orientation to the project, with those allocated to the intervention group taking part in an additional workshop to co-create the shared decision support tool. To further promote intervention fidelity, monthly meetings are scheduled with EI service coordinators allocated to the intervention group. During meetings, case example(s) of using the YC-PEM e-PRO report and the shared decision support tool are shared and discussed. This fosters protocol adherence across service coordinators and provides opportunity for peer mentorship.[25] Additionally, the meetings will inform the development of qualitative questions for the aim 2 interview guide regarding supports and barriers to longer-term implementation.

Strategies for implementation

Two implementation strategies (see table 1) have been integrated into the project to support: (1) user navigation through the YC-PEM e-PRO and (2) EI coordinator engagement.

Standard care

Control group participants will receive standard care consisting of an annual IFSP meeting where families and service coordinator discuss the developmental assessment results and other service provider's (eg, occupational therapists, physical therapists) observations and conduct a family assessment (ie, semi-structured family interview) to elicit families' concerns, priorities and resources for IFSP development. The YC-PEM e-PRO is not part of standard care and will therefore not be completed by the control group.

Outcomes assessment

Information about select child and family demographics will be gathered at baseline, including the child's age, gender (ie, male, female, self-described), and caregiver race (ie, American Indian/Alaskan Native, Asian, Black or African American, Caucasian, Hawaiian or other Pacific Islander, self-described).[42] Table 2 summarises primary and secondary outcome measures.

Qualitative data

Following trial completion, data will be collected through focus groups with three types of EI stakeholders using a semi-structured interview guide. Questions will centre on explaining quantitative results on the usefulness of the implementation strategies employed (eg, how helpful was the video for completing the YC-PEM e-PRO; how helpful the YC-PEM e-PRO report was to guide discussion at the annual IFSP meeting).

Focus groups with EI service coordinators and leadership will also revisit benefits and challenges with implementing YC-PEM e-PRO, as previously described during aim 1 monthly meetings with service coordinators and project coordinators. Additional questions will be posed about remaining barriers and facilitators to its broader and sustained deployment across multiple EI programmes, with an eye towards provider efficiency as now captured in the Quadruple Aim of healthcare.[43] This may include inquiring about expanding YC-PEM e-PRO report functionality to include programme dashboards that are tailored to the distinct informational needs of families, providers and EI leadership. Whereas families may benefit and be restricted to viewing their child's item-level e-PRO results, providers may need access to item-level results for multiple families on their caseload, and EI leadership may benefit from viewing e-PRO results in aggregate form instead (eg, average percent desired change across home activities for groups of EI families). Research staff without prior EI experience will facilitate focus groups, to ensure participant comfort in providing feedback on facilitators and barriers to implementation.

Patient and public involvement

The YC-PEM e-PRO has a 13-year history of EI programme leadership, service provider, and caregiver engagement to guide its design and use,[9–11] including: (1) caregiver engagement to design, validate and begin to identify supports and barriers to its implementation in a single EI programme[9 13 25 28 44]; (2) co-creation of an online knowledge hub with resources (eg, user guide, tip sheet) to increase its uptake by families, service providers and organisations[45]; (3) caregiver and service provider engagement to upgrade and reduce the burden of delivering this intervention during the aim 1 trial (eg, designing animated instructional videos, creating activity illustrations and co-creating summary reports),[46 47] and (4) caregiver and service provider engagement and capacity building to deliver the YC-PEM e-PRO with productive, sustainable community-academic collaboration.[25] The resulting research group infrastructure enables interested EI service coordinators to share in decisions about protocol design (research question and outcome measure selection), management of participant recruitment/retention, the use YC-PEM e-PRO information in

Table 1 Implementation strategies integrated during project preparation phase

Strategy	Description	Rationale
Strategies to Support YC-PEM e-PRO User Navigation		
Expand and enhance YC-PEM e-PRO instructions in REDCap using an embedded animated introductory video	► New embedded YC-PEM animated instructional video script was written at a fifth grade reading level and introduced caregivers to key participation concepts	Families indicated need for simplifying original instructions to optimise navigation and clarity on how an assessment of child's participation is distinct from assessment of developmental and functional skills.[14 27]
Embed activity illustrations within the YC-PEM e-PRO to augment text descriptions	► A biomedical illustrator created 27 illustrations to represent multiple ways that children and caregivers might participate in a YC-PEM e-PRO activity (ie, for meal preparation, illustrations depict children participating in setting the table, unloading groceries and scooping ingredients) ► Illustrations were intentionally diversified according to child and caregiver age, gender and race/ethnicity	This strategy aligns with current evidence that regardless of health literacy levels, individuals find it easier to imagine website content that includes visuals or animations.[63]

Improve layout of YC-PEM e-PRO report structure	► Co-design a report with providers that includes upgrades to:
	  – Content/Typography: personalised titles, introductions and conclusions
	  – Colour: intentional use of calming and alerting colours
	  – Structure/space: open text boxes strategically placed to offer caregivers opportunities to write down notes
	  – Style/tone: report elicits specific emotions/tones (eg, sense of caregiver pride, professionalism)
	  – Graphics/symbols: tables and bar graphs to display results
		Stakeholder engagement in designing ways to optimise the reporting of information from participation-focused tools will increase the likelihood of their adoption into existing organisational workflows.[25 64]

Strategies to Support EI Provider Engagement		
Launch and maintain a new research group at the EI study site	► EI service coordinators co-designed a High Value Early Intervention Research Group to earn credit for their contributions to study design, implementation, interpretation and/or dissemination of results ► Ten members have so far: (1) shared in decisions about protocol design and strategies for participant recruitment and retention; (2) co-created the trial name and logo, video options for online consent and HIPAA authorisation, and the structure and schedule for EI service coordinator check-ins during trial completion	Service coordinator workload, competing priorities and/or lack of familiarity with research may contribute to disengagement.[26] This research group may be a key driver of protocol implementation, as most EI programmes are not university-affiliated and vary in research capacity

EI, early intervention; HIPAA, Health Insurance Portability and Accountability Act; REDCap, Research Electronic Data Capture; YC-PEM e-PRO, Young Children's Participation and Environment Measure Electronic Patient-Reported Outcome.

the IFSP planning meeting, and the training and retention of EI service coordinators during the aim 1 trial. An EI engagement specialist also joined the research group and provided input on caregiver recruitment and created video options for caregivers to provide informed consent and HIPAA authorisation. The research group, inclusive of the EI engagement specialist, will disseminate results to study participants via non-refereed internal newsletters and an external podcast.

Statistical analysis

Data will be exported from REDCap and into SAS V.9.4 for analyses.[48] For aim 1, the sample will be described at pre-intervention and analysed to accomplish the following tasks before the main analyses. We will depict univariate and bivariate data distributions using frequency with percentage for categorical variables and mean with SD or median with IQR for continuous variables. Baseline imbalances between intervention and control groups will be identified using univariate generalised linear mixed model[49] with details provided in the main analysis below. Moreover, data anomalies, such as outliers and statistical assumptions for planned analyses, such as non-normality, will be checked and remedied with data transformations or more statistically robust approaches. Finally, the amount and patterns of missing data will be evaluated and handled by each method's assumption or by using multiple imputation with fully conditional specification approach[50] as appropriate.

Main data analyses will be based on intent-to-treat principle in this cluster-randomised clinical trial. To estimate the intervention effect on score changes over time compared with the control group, generalised linear mixed model will be fitted for each of the outcomes of interest, including MPOC subdomain enabling and

Table 2 Outcome measures

Outcome (Construct)	Outcome measure	Description	Primary outcome	Secondary outcome	Time point (Data Collection)
Service quality	MPOC-20; 20 Items	Assesses caregiver perceptions of family-centred care in four domains: (1) enabling and partnership; (2) providing general and specific information; (3) coordinated and comprehensive care and (4) respectful and supportive care.[65] The MPOC-20 has acceptable to good reliability and validity.[65 66]	X	X	Baseline, 1, 6 and 12 months
Caregiver engagement for shared decision making	PPEM; 5 Items	Caregiver-rated questionnaire measuring perceived engagement in services		X	Baseline, 1, 6 and 12 months
Caregiver activation for shared decision-making	P-PAM-13; 13 Items	Measure that assesses caregiver knowledge, skills and confidence to manage their child's care.[67 68]		X	Baseline, 1, 6 and 12 months
Service plan focus	Retrieved via EI service record abstraction	Record abstraction will be used to collect and code data on the individual family service plan as finalised following the IFSP meeting, using established quality criteria, to derive an estimate of the proportion of participation-focused goals in the service plan.[16]		X	Post-intervention
Child functioning	Retrieved via EI service record abstraction	Record abstraction will be used to collect the child's percent developmental delay in five developmental domains as derived from standardised developmental assessments (ie, adaptive behaviour, cognitive, communication, physical, social emotional) and COS scores. COS scores are a consensus rating by the EI team based on developmental scores and practitioner and parent perceptions of child function.[69]		X	Baseline and 12 months

COS, Child Outcomes Summary; EI, Early Intervention; IFSP, Individualised Family Service Plan; MPOC-20, Measure of Processes of Care; P-PAM, Parent Patient Activation Measure; PPEM, Parent Participation Engagement Measure.

partnership (H1) as the primary outcome, and the remaining MPOC subdomains (H2), partnership P-PAM and PPEM (H3), DS and COS (H5) as the secondary outcomes. Each model will include group (intervention vs control), time (1, 6 and 12 months post-baseline vs baseline), and their interactions as the primary fixed effects, while adjusting for appropriate factors, such as child age, and condition severity, as secondary fixed effects along with coordinator-level and subject-level random effects, if possible. This model controls for cluster dependence at three levels (coordinator, participant and repeated measures) through the random effects and permits the calculation of intercepts for each individual participant. Moreover, the model will estimate average slopes and intercepts in terms of coefficients, which provide the magnitude of the intervention effect between two groups especially via the interaction terms. Considering the small number of coordinator-level cluster as 6, the between-within approach[51] will be applied for small-sample correction as suggested by Leyrat *et al*.[52] Missing data can be handled by this model under the missing at random assumption with the maximum use of available data. The estimated intracluster correlation coefficient for the primary outcome and estimated coefficients in each model for every outcome will be presented along with the coefficients' SE, p values. Reports for aim 1 will follow the guidance by Campbell *et al*.[53] A two-sided $p<0.05$ would be considered as statistical significance. The statistician will be blinded as to group assignment while conducting all analyses.

We will rate EI service plans using quality rating criteria,[16 54] to derive estimates of the proportion of participation-focused goals before and after intervention. We will control for history of family interview completion (yes, no) to examine the effect of the upgraded YC-PEM e-PRO and decision support tool on the proportion of participation-focused goals in the EI service plan.

Qualitative analysis

Focus group interviews will be transcribed verbatim, and transcripts will imported into NVivo V.11.0 for analyses.[55] We will use deductive and inductive content analysis[56] to group data into categories of common facilitators and barriers to implementing the YC-PEM e-PRO. We will initially use the CFIR to develop a draft codebook based on known implementation factors[24] and refine it iteratively. The data coding process will be as follows. First, two study staff will independently code 20% of data. Second, the researchers will meet to resolve coding discrepancies and collaboratively review and update the codebook. The remaining data will be split between the two coders and analysed separately using the updated codebook.

Categories will be labelled using participants' own words and reviewed by co-investigators to ensure homogeneity (ie, each theme is distinct and has multiple instances of supporting text). Illustrative quotes will also be identified and included. We will develop three sets of summaries (one per EI stakeholder type) to be shared with members of a national advisory group comprised of EI practitioners who are employed across multiple states to review findings. Advisory group members will provide input on the relevance (a rating of impact and ease of implementation) of each support and barrier to the YC-PEM e-PRO in their EI programme. These inputs will inform systematic prioritisation of factors to implementation across multiple EI programmes nationally.

DISCUSSION

The YC-PEM e-PRO, when paired with a decision support tool, holds promise for providing an alternative option for family-centred and participation-focused EI care as compared with the standard face-to-face semistructured interview.[13 28] However, there is need to further optimise the YC-PEM e-PRO and examine its effectiveness and implementation within the EI service context, by including a control group for comparison, examining effects across multiple endpoints and over time, and collecting diverse stakeholder perspectives to appraise supports and barriers to its scalability across multiple EI programmes.[28] This protocol paper describes a hybrid type 1 effectiveness-implementation study consisting of two parts: (1) a pragmatic cluster randomised control trial to examine effectiveness on select child and family

outcomes (aim 1); and (2) an explanatory sequential mixed-methods approach to examining supports and barriers to longer-term implementation within an EI context (Aim 2).

While results on effectiveness will inform estimates about the value of this intervention, results on barriers and facilitators for intervention implementation are needed to support intervention uptake across multiple EI programmes.[58 59] For the latter, we aim to gain diverse EI stakeholder perspectives to guide the design of implementation strategies that can support longer-term implementation as a health systems intervention. EI families may provide insights about potential upgrades to the YC-PEM e-PRO and decision support tool for family-centred and participation-focused care with individual families.[60–62] In contrast, EI providers and leadership may provide greater insight about how to sustain this intervention in ways that reduce provider burden and increase leadership capacity for monitoring service trends and identifying targets for continuous quality improvement. Both perspectives are needed to advance its implementation as a health systems intervention in EI.

It is expected that the results of this research will provide evidence to guide hybrid type 2 and/or type 3 implementation research on the YC-PEM e-PRO across multiple EI programmes, to systematically support its implementation as a health systems intervention for advancing family-centred and participation-focused services in EI.

There are three anticipated limitations to the proposed study. First, limiting the inclusion criteria to English-speaking families for both parts may limit generalisability of the results from this phase of work to a subset of families that are enrolled in EI. Additionally, conducting this study during the COVID-19 pandemic poses limitations to timely participant recruitment and data collection. There might be more challenges recruiting families due to lower EI enrolment in the single EI programme. Similarly, caregivers who do access EI and choose to enrol might be better able to appraise their child's participation at home as compared with the community due, in part, to COVID-19 restrictions.

Author affiliations
[1]Children's Participation in Environment Research Lab, University of Illinois at Chicago, Chicago, Illinois, USA
[2]Rehabilitation Sciences, University of Illinois at Chicago, Chicago, Illinois, USA
[3]Center for Clinical and Translational Science, University of Illinois at Chicago, Chicago, Illinois, USA
[4]Health Systems, Management & Policy, University of Colorado - Anschutz Medical Campus, Aurora, Colorado, USA
[5]Biomedical & Health Information Sciences, University of Illinois at Chicago, Chicago, Illinois, USA
[6]Rocky Mountain Human Services, Denver, Colorado, USA
[7]Occupational Therapy, University of Pittsburgh, Pittsburgh, Pennsylvania, USA
[8]CanChild Centre for Childhood Disability Research, McMaster University, Hamilton, Ontario, Canada

Twitter Elizabeth Papautsky @elpPhD

ORCID iDs
Elizabeth Papautsky http://orcid.org/0000-0003-2771-9579
Mary Khetani http://orcid.org/0000-0003-2628-3371

REFERENCES

1 Hanna K, Rodger S. Towards family-centred practice in paediatric occupational therapy: A review of the literature on parent-therapist collaboration. *Aust Occup Ther J* 2002;49:14–24.
2 King G, Kertoy M, King S, *et al*. A measure of parents' and service providers' beliefs about participation in family-centered services. *Children's Health Care* 2003;32:191–214.
3 Rosenbaum P. How do we know if interventions in developmental disability are effective? *Dev Med Child Neurol* 2020;62:1344.
4 Lowes LP, Noritz GH, Newmeyer A, *et al*. 'Learn from every patient': Implementation and early results of a learning health system. *Dev Med Child Neurol* 2017;59:183–91.
5 The DaSy Center. *DaSy Data System Framework*. Menlo Park, CA: SRI International, 2014.
6 Nelson R, Bull B. *Building a better data system: What are process and data models?* Menlo Park, CA: SRI International, 2016.
7 An M, Palisano RJ. Family-professional collaboration in pediatric rehabilitation: A practice model. *Disabil Rehabil* 2014;36:434–40.
8 Brewer K, Pollock N, Wright FV. Addressing the challenges of collaborative goal setting with children and their families. *Phys Occup Ther Pediatr* 2014;34:138–52. doi:10.3109/01942638.2013.794187
9 Khetani MA, Cohn ES, Orsmond GI, *et al*. Parent perspectives of participation in home and community activities when receiving part C early intervention services. *Topics Early Child Spec Educ* 2013;32:234–45.

10 KhetaniMA, Graham JE, Alvord C. Community participation patterns among preschool-aged children who have received part C early intervention services. *Child Care Health Dev* 2013;39:490–9.

11 Khetani MA, Orsmond G, Cohn E, *et al*. Correlates of community participation among families transitioning from part C early intervention services. *OTJR* 2012;32:61–9.

12 Khetani MA, Graham JE, Davies PL, *et al*. Psychometric properties of the young children's participation and environment measure. *Arch Phys Med Rehabil* 2015;96:307–16.

13 Khetani MA, McManus BM, Arestad K, *et al*. Technology-Based functional assessment in early childhood intervention: A pilot study. *Pilot Feasibility Stud* 2018;4:65. doi:10.1186/s40814-018-0260-1

14 AlbrechtEC, Kaelin VC, Rigau BL, *et al*. Pilot implementation of an electronic patient-reported outcome measure for planning and monitoring participation-focused care in early intervention. *BMC Med Inform Decis Mak* 2020;20:199.

15 KhetaniMA. Validation of environmental content in the Young Children's Participation and Environment Measure. *Arch Phys Med Rehabil* 2015;96:317–22.

16 Bosak DL, Jarvis JM, Khetani MA. Caregiver creation of participation-focused care plans using Participation and Environment Measure Plus (PEM+), an electronic health tool for family-centred care. *Child Care Health Dev* 2019;45:791–8. doi:10.1111/cch.12709

17 Jarvis JM, Gurga A, Choong K, *et al*. Caregiver strategy use to promote children's home participation following pediatric critical illness. *Arch Phys Med Rehabil* 2019;99:e193–4.

18 Kaelin VC, BosakDL, Villegas VC, *et al*. Participation-focused strategy use among caregivers of children receiving early intervention. *Am J Occup Ther* 2021;75:7501205090p1–7501205090. doi:10.5014/ajot.2021.041962

19 Khetani MA, Lim HK, Corden ME. Caregiver input to optimize the design of a pediatric care planning guide for rehabilitation: Descriptive study. *JMIR Rehabil Assist Technol* 2017;4. doi:10.2196/rehab.7566

20 Williams U, Law M, Hanna S, *et al*. Personal, environmental, and family factors of participation among young children. *Child Care Health Dev* 2019;45:448–56.

21 Khetani MA, McManus BM, Albrecht EC, *et al*. Early intervention service intensity and young children's home participation. *BMC Pediatr* 2020;20:330.

22 Wang S, Blazer D, Hoenig H. Can eHealth technology enhance the patient-provider relationship in rehabilitation? *Arch Phys Med Rehabil* 2016;97:1403–6.

23 Schiariti V, Fowler E, Brandenburg JE, *et al*. A common data language for clinical research studies: the National Institute of Neurological Disorders and Stroke and American Academy for Cerebral Palsy and Developmental Medicine Cerebral Palsy Common Data Elements Version 1.0 recommendations. *Dev Med Child Neurol* 2018;60:976–86.

24 Damschroder LJ, Aron DC, Keith RE, *et al*. Fostering implementation of health services research findings into practice: A consolidated framework for advancing implementation science. *Implement Sci* 2009;4:1–15.

25 Rigau BL, Scully EA, Dooling-Litfin JK, *et al*. Community engagement to pilot electronic patient-reported outcomes (e-PROs) in early intervention: Lessons learned. *J Clin Transl Sci* 2018;2:20–6. doi:10.1017/cts.2018.15

26 Rigau, BL, Dooling-Litfin, J, Scully, E, *et al*. Building organizational capacity for research in early intervention. *Zero To Three J* 2019;39:54–61.

27 Imms C. Participation in diverse life situations for people with disability: A vision for the future. *Dev Med Child Neurol* 2020;62:5.

28 Albrecht EC, Kaelin VC, Rigau BL, *et al*. Pilot implementation of an electronic patient-reported outcome measure for planning and monitoring participation-focused care in early intervention. *BMC Med Inform Decis Mak* 2020;20:199. doi:10.1186/s12911-020-01189-9

29 Chan AW, Tetzlaff JM, Altman DG, *et al*. Spirit 2013 statement: Defining standard protocol items for clinical trials. *Ann Intern Med* 2013;158:200–7.

30 Thorpe KE, Zwarenstein M, Oxman AD, *et al*. A pragmatic-explanatory continuum indicator summary (PRECIS): A tool to help trial designers. *J Clin Epidemiol* 2009;62:464–75.

31 Loudon K, Treweek S, Sullivan F, *et al*. The PRECIS-2 tool: Designing trials that are fit for purpose. *BMJ* 2015;350:h2147.

32 Creswell J, Clark V. *Designing and conducting mixed methods research*. 2nd edn. Thousand Oaks, CA: SAGE Publications, Inc, 2011.

33 Curran GM, Bauer M, Mittman B, *et al*. Effectiveness-implementation hybrid designs. *Med Care* 2012;50:217–26.

34 Harris PA, Taylor R, Thielke R, *et al*. Research electronic data capture (REDCap)- A metadata-driven methodology and workflow process

35 Harris PA, Taylor R, Minor BL, *et al*. The REDCap consortium: Building an international community of software platform partners. *J Biomed Inform* 2019;95:103208.

36 Cohen J. *Statistical power analysis for the behavioral sciences*. 2nd edn. Hillsdale, NJ: Lawrence Earlbaum Associates, 1988.

37 Kinney AR, Eakman AM, Graham JE. Novel effect size interpretation guidelines and an evaluation of statistical power in rehabilitation research. *Arch Phys Med Rehabil* 2020;101:2219–26. doi:10.1016/j.apmr.2020.02.017

38 Fonvig CE, Rasmussen HM, Overgaard S, *et al*. Effectiveness of instrumented gait analysis in interdisciplinary interventions on parents' perception of family-centered service and on gross motor function in children with cerebral palsy: a randomized controlled trial. *BMC Pediatr* 2020;20:411.

39 Eldridge SM, Ashby D, Kerry S. Sample size for cluster randomized trials: Effect of coefficient of variation of cluster size and analysis method. *Int J Epidemiol* 2006;35:1292–300.

40 Schliep ME, Alonzo CN, Morris MA. Beyond RCTs: Innovations in research design and methods to advance implementation science. *Evid Based Commun Assess Interv* 2017;11:82–98.

41 Khetani MA, Coster W, Law MC. *Young Children's Participation and Environment Measure*. Fort Collins, CO: Colorado State University (copyright to authors), 2013.

42 Charles and Lynn Schusterman Family Foundation. *More than numbers. A guide toward diversity, equity, and inclusion (DEI) in data collection*, 2020: 1–55.

43 Bodenheimer T, Sinsky C. From triple to quadruple aim: Care of the patient requires care of the provider. *Ann Fam Med* 2014;12:573–6.

44 Benjamin TE, Lucas-Thompson RG, Little LM, *et al*. Participation in early childhood educational environments for young children with and without developmental disabilities and delays: A mixed methods study. *Phys Occup Ther Pediatr* 2017;37:87–107.

45 Villegas V, Salgado Z, Grief A. Stakeholder engagement to design products that can accelerate the uptake of the Young Children's Participation and Environment Measure (YC-PEM) in pediatric practice, 2021. CanChild centre for childhood disability participation knowledge hub. Available: https://www.canchild.ca/en/resources/223-young-children-s-participation-and-environment-measure-ycpem [Accessed 17 Dec 2021].

46 Villegas VC, Stoffel A, Greif A. Caregiver engagement to optimize participation-focused assessment and care planning in pediatric practice. In: *1.5-hour short course presented as a pre-recorded session for the Illinois Occupational Therapy Association annual conference*. Lisle, IL, 2020.

47 Gurga A, Greif A, Sidorowych C. Success story: Designing a participation-focused report yield, 2019. CanChild centre for childhood disability participation knowledge hub. Available: https://www.canchild.ca/en/resources/223-young-children-s-participation-and-environment-measure-ycpem [Accessed 08 Feb 2020].

48 SAS Institute Inc. *SAS/ACCESS® 9.4 Interface to ADABAS [Internet*. Cary, NC, 2013. https://www.sas.com/es_es/software/viya.html

49 Brown H, Prescott R. *Applied mixed models in medicine*. . John Wiley & Sons, 2014: Vol 42. 442.

50 Van Buuren S, Brand JPL, Groothuis-Oudshoorn CGM, *et al*. Fully conditional specification in multivariate imputation. *J Stat Comput Simul* 2006;76:1049–64. doi:10.1080/10629360600810434

51 Li P, Redden DT. Comparing denominator degrees of freedom approximations for the generalized linear mixed model in analyzing binary outcome in small sample cluster-randomized trials. *BMC Med Res Methodol* 2015;15:38. doi:10.1186/s12874-015-0026-x

52 Leyrat C, Morgan KE, Leurent B. Cluster randomized trials with a small number of clusters: Which analyses should be used? *Int J Epidemiol* 2018;47. doi:10.1093/ije/dyx169

53 Campbell MK, Piaggio G, Elbourne DR, *et al*. Consort 2010 statement: Extension to cluster randomised trials. *BMJ* 2012;345:e5661. doi:10.1136/bmj.e5661

54 Kolehmainen N, Marshall J, Hislop J, *et al*. Implementing participation-focused services: A study to develop the method for using audit and feedback in participation implementation (MAPi). *Child Care Health Dev* 2020;46:37–45.

55 QSR international Pty LTD NVivo, 2020. Available: https://www.qsrinternational.com/nvivo-qualitative-data-analysis-software/home

56 Mayring P. *Qualitative content analysis: Theoretical foundation, basic procedures and software solution*, 2014. https://www.researchgate.net/publication/266859800_Qualitative_content_analysis_-_theoretical_foundation_basic_procedures_and_software_solution

57 ICMJE. Defining the role of authors and contributors, 2021. Available: http://www.icmje.org/recommendations/browse/roles-and-

for providing translational research informatics support. *J Biomed Inform* 2009;42:377–81.

responsibilities/defining-the-role-of-authors-and-contributors.html [Accessed 12 Mar 2021].

58 Spiker D, Hebbeler K, Wagner M, *et al*. A framework for describing variations in state early intervention systems. *Topics Early Child Spec Educ* 2000;20:195–207.

59 Kasprzak C, Hebbeler K, Spiker D, *et al*. A state system framework for high-quality early intervention and early childhood special education. *Topics Early Child Spec Educ* 2020;40:97–109.

60 Jarvis JM, Kaelin VC, Anaby D, *et al*. Electronic participation-focused care planning support for families: A pilot study. *Dev Med Child Neurol* 2020;62:954–61. doi:10.1111/dmcn.14535

61 Jarvis JM, Gurga A, Greif A, *et al*. Usability of the participation and environment measure plus (PEM+) for client-centered and participation-focused care planning. *Am J Occup Ther* 2019;73:7304205130p1–7304205130.

62 Khetani MA, Cliff AB, Schelly C, *et al*. Decisional support algorithm for collaborative care planning using the participation and environment measure for children and youth (PEM-CY): A mixed methods study. *Phys Occup Ther Pediatr* 2015;35:231–52.

63 Meppelink CS, Smit EG, Diviani N, *et al*. Health literacy and online health information processing: Unraveling the underlying mechanisms. *J Health Commun* 2016;21:109–20. doi:10.1080/10810 730.2016.1193920

64 Moll S, Wyndham-West M, Mulvale G, *et al*. Are you really doing 'codesign'? Critical reflections when working with vulnerable populations. *BMJ Open* 2020;10:e038339. doi:10.1136/bmjopen-2020-038339

65 King S, King G, Rosenbaum P. Evaluating health service delivery to children with chronic conditions and their families: Development of a refined measure of processes of care (MPOC–20). *Children's Health Care* 2004;33:35–57.

66 King SM, Rosenbaum PL, King GA. Parents' perceptions of caregiving: Development and validation of a measure of processes. *Dev Med Child Neurol* 1996;38:757–72.

67 Hibbard JH, Mahoney ER, Stockard J, *et al*. Development and testing of a short form of the patient activation measure. *Health Serv Res* 2005;40:1918–30.

68 Pennarola BW, Rodday AM, Mayer DK, *et al*. Factors associated with parental activation in pediatric hematopoietic stem cell transplant. *Med Care Res Rev* 2012;69:194–214.

69 Early Childhood Technical Assistance Center. Child outcomes summary (COS) process. 2018.