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The Cellie Coping Kit for Kids with Craniofacial Conditions: Developing a Culturally Sensitive Psychosocial Support Tool and Coping Resource for Families with School Age Children with Craniofacial Conditions from Diverse Backgrounds

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The Cellie Coping Kit for Craniofacial Conditions: Developing and Pilot Testing a Psychosocial Support Tool and Coping Resource for Families with School-Age Children with Craniofacial Conditions

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Abstract

The current project aimed to develop and pilot test a psychosocial support tool and coping resource: the Cellie Coping Kit for Craniofacial Conditions (Cellie Kit). We adapted a cognitive behavioral theory-based tool that promotes resilience and coping, the Cellie Coping Kit, to be appropriate for families of children with craniofacial conditions. Adaptation was guided by an integration of the Cellie Kit adaptation process with heuristic and ecological validity frameworks to increase cultural sensitivity for Hispanic/Latinx families. Kit adaptation and current pilot testing are part of a larger project that includes kit translation, production, and pilot testing of a bilingual Spanish-English Cellie Kit, in addition to the English Cellie Kit development and pilot testing focused on within the current study. Presently, six English speaking families were enrolled and four completed the Cellie Kit Intervention Program, pre- and post-measures, and semi-structured satisfaction interviews and questionnaires at the end of their participation. A mixed-methods approach including descriptive statistics and codebook thematic analysis, were utilized. Study feasibility and acceptability aims were explored through examining researcher fidelity forms, and quantitative and qualitative review of verbal and written participant reports. We found evidence for the feasibility of the coping intervention based on intervention implementation metrics, and both feasibility and acceptability based on participant-reports. Findings will inform future modifications to enhance kit materials and intervention program design as well as guide future evaluation, and potential for integration into routine clinical care.

Keywords: craniofacial conditions, coping tool, resilience, family intervention

The Cellie Coping Kit for Craniofacial Conditions: Developing and Pilot Testing a Psychosocial Support Tool and Coping Resource for Families with School-Age Children with Craniofacial Conditions

Craniofacial conditions affect the soft tissue and bone in the face and skull, typically causing asymmetry or incomplete formation of facial features. The most common craniofacial condition is cleft lip and/or cleft palate which impacts more than 1 in 700 children (Mai et al., 2015). Incidence rates vary by racial and ethnic backgrounds with higher rates of cleft lip with cleft palate among Hispanic/Latinx, American Indians and Alaska Natives compared to White children (Canfield et al., 2014; Mai et al., 2015). Children with craniofacial conditions undergo extensive treatment that often includes multiple surgeries and numerous specialists (e.g., surgeon, audiologist, orthodontist, speech language pathologist) (ACPA-CPF, 2018) and many are at risk for developmental, speech and language delays, and difficulties with feeding, vision, hearing, learning, and behavior (ACPA-CPF, 2018). These craniofacial-related stressors put children with craniofacial conditions at risk for psychosocial difficulties including anxiety, depression, low self-concept, behavioral challenges, social stigmatization, and teasing (Crerand et al., 2017; Feragen & Stock, 2017). Caregivers play an important part in supporting their child's coping efforts and if they have a difficult time coping with their child's condition, their child is more likely to experience difficulties with adjustment too (Carmona et al., 2020; Sischo, Broder, Philips 2015). Therefore, family-centered approaches to addressing craniofacial-related stressors and promoting resiliency through developing child and caregiver coping tools that also support caregivers in helping their child cope are critical.

When conceptualizing the risks that children with craniofacial conditions face, it is important to consider salient and relevant aspects of their identities such as race, ethnicity, culture, language, family income, and insurance coverage, as these factors are crucial to understanding children and families' experiences within the health care system (Kazak, 2006). Access and quality of care, which often differ across these factors, are closely linked with children's health related quality of life (HRQoL) and family functioning. For example, language, cultural dissimilarities and cultural communication differences between patients and providers may act as barriers to receiving optimal care for Hispanic/Latinx families (Carmona et al., 2020; Jacquez et al., 2016). Black children may also experience disparate care including delays in surgical intervention, longer hospital stays following surgery, and higher hospital costs (Nidey & Wehby, 2019; Peck et al., 2020). Additionally, Latinx, Black, and publicly insured children with craniofacial conditions have reported lower HRQoL overall and across functional (publicly insured group only), social-emotional, and school domains, and lower family functioning compared to White and privately insured youth (Broder et al., 2012; Crerand et al., 2015). Collectively, this evidence points to racial, ethnic, and economic disparities in psychosocial risks and family coping resources and the need for linguistically and culturally appropriate care and tools to support families coping with craniofacial-related stressors (Carmona et al., 2020). Therefore, children with craniofacial conditions and their families need support coping in a way that increases access to quality care and addresses culturally relevant concerns. This can be done through connection to resources that empower youth and caregivers to advocate for their needs through access to psychoeducation about their conditions, culturally-acceptable coping and emotion-regulatory strategies, encouragement to connect to social supports and increased confidence and comfort communicating about the condition.

The current project seeks to take a step towards integrating evidence-based craniofacial-specific intervention tools into routine clinical care by developing a universal level coping resource. The psychosocial risk framework defines universal level supports as a broad-based support that is available to all families experiencing distress, such as challenges associated with a specific medical condition (Kazak, 2006). Universal level supports aim to prevent escalation of distress by promoting resilience through enhancing child and family coping by supporting use of existing resources and building upon them often through psychoeducation, teaching coping strategies, and connecting to additional social supports and resources (Kazak, 2006).

The Cellie Coping Kit is an evidence-based coping tool that promotes resilience and coping in families of school-aged children with chronic medical conditions from diverse backgrounds. This kit includes a stuffed toy (Cellie doll), children's coping cards outlining medical and emotional cognitive-behavioral coping strategies and a caregivers' booklet describing approaches for supporting their child's and their own coping. The Cellie Coping Kit was originally developed for children with cancer and has been successfully adapted for use with multiple specialized pediatric populations (e.g., sickle cell disease, food allergy) (Cole et al., 2017; Marsac et al., 2012; Marsac et al., 2014). Across pediatric conditions, caregivers and children have found the Cellie Coping Kit to be feasible (e.g., easy to use), relevant to their experiences, and effective in teaching coping strategies (e.g., emotional expression, communicating about condition, normalizing experiences, distraction during procedures; Cole et al., 2017; Marsac et al., 2012; Marsac et al., 2014; Marsac et al., 2018). Kit use has additionally been associated with improvement in pediatric HRQoL (Marsac et al., 2018).

The current project involved the adaptation of the Cellie Coping Kit for the craniofacial community, resulting in the development of the Cellie Coping Kit for Craniofacial Conditions (Cellie Kit). Given that the UI Health Craniofacial Clinic predominately serves families from diverse racial and ethnic backgrounds (i.e., 50% Hispanic/Latinx, 15% Black, 3% Asian and 2% Other races), those who are publicly insured (i.e., 70%) and a high proportion of Spanish-speaking Hispanic/Latinx caregivers and bilingual youth, we strove to adapt the Cellie Coping Kit to meet the needs of our specific population. The Cellie Coping Kit adaptation process for use with children with craniofacial conditions (Marsac et al., 2014; Marsac et al., 2018) was integrated with heuristic and ecological validity frameworks to increase cultural sensitivity and appropriateness for Hispanic/Latinx families (Barrera & Castro, 2006; Bernal et al., 1995; Marsac et al., 2014). The heuristic framework proposes a step-by-step process for conducting a cultural adaptation of an intervention to enhance participant engagement (Barrera & Castro, 2006). The ecological validity framework guides the adaptation process by assessing the cultural relevance and fit of intervention components, leading to adaptations that seek to increase the appropriateness of the intervention for a specific ethnic or cultural group and facilitate positive outcomes (Bernal et al., 1995). We focused on adapting the language used within the Cellie Kit and intervention to meet our patient populations’ linguistic and social identity needs and educational and acculturation experiences. Moreover, adaptation and intervention development took place during the course of a global pandemic and so, the intervention was designed to be implemented remotely to increase accessibility.

In line with previous versions of the Cellie Coping Kit, the aim for the Cellie Kit was to help to address condition-specific stressors and support families in building resilience through increasing psychological and social resources. The tool does this by providing psychoeducation, encouraging parent-child communication, teaching emotion regulatory skills, and helping families identify coping strategies to implement presently and in the future, to reduce the development of pediatric medical traumatic stress by increasing family preparedness to navigate and cope with condition-specific and healthcare-related challenges. The resource is framed as a universal support for families that can be utilized within a stepped approach to care.

Rationale

The purpose of our research is to support the psychosocial needs of craniofacial patients and their families, particularly those from backgrounds at greater risk for experiencing health disparities, through the provision of a low-cost, evidence-based tool that promotes resilience through teaching adaptive coping skills in a family-focused format. Specifically, we developed the Cellie Kit to fill an important need in the craniofacial community. While our bilingual Spanish-English version of the kit is in production, we moved forward with pilot testing the English kit with families who speak English. Therefore, the overarching aim of our pilot study is to explore the feasibility, acceptability, and relevance of the adapted coping kit for English-speaking families of school-aged children with craniofacial conditions. Within feasibility, we aim to explore (1) the feasibility of delivering the intervention program remotely with regard to implementation and usability. Additionally, we aim to explore (2) the acceptability and relevance of coping kit topics and intervention materials to the diverse life experiences of families with school-aged children with craniofacial conditions.

Method

Participants

Children were eligible to participate if they were between the ages of 6 and 12 years, had a craniofacial condition including cleft lip and/or palate, hemifacial microsomia or related condition (e.g., microtia), or a complex syndrome (e.g., Apert's Syndrome), had sufficient English verbal comprehension skills and the cognitive ability to participate in the intervention and complete questionnaires. Caregivers needed to be able to read written materials in English. Study procedures were approved by the (UI Health) Hospital Institutional Review Board (IRB) and an IRB Authorization Agreement was granted by the affiliated (DePaul) University.

Recruitment Procedures

Families were recruited during in-person and virtual clinic visits as well as through phone and email recruiting methods. Potential subjects were identified through study team member review of clinic schedule and clinic provider identification. Recruitment flyers were displayed within the Craniofacial Center, handed to eligible and interested families in clinic, and emailed to families. Families who expressed interest in participating were asked to share their contact information. A research team member followed-up with families during a clinic appointment, phone call and/or email to further assess interest and share additional information about the study. Clinical providers actively involved in a participant's care assessed initial family interest in participation but did not have an active role in recruitment. Research team members who were not involved in the patient's clinical care, provided additional information about the study, consented, and enrolled families.

Description of the Cellie Coping Kit Intervention

After providing consent and assent, in which confidentiality and its limitations were discussed, families completed baseline measures via Qualtrics. They imputed a study ID rather than their name to support privacy protection. Support completing measures was offered to all children and caregivers and two children completed their measures over Zoom with support from the master's level doctoral student who enrolled them and who would become their study interventionist. Afterwards, an initial coping kit training visit was scheduled over Zoom for the caregiver(s) and child to meet with a study interventionist (one of two previous clinical psychology externs at the UI Health Craniofacial Center including this writer). A Cellie Kit was mailed to families prior to the visit. The baseline visits were scheduled for one hour and included an introduction to the Cellie Kit components (see Figure 1.) and overview of their potential use (see Appendix for initial session components list on the Intervention Feasibility Measure and Intervention Manual). Collaboratively, the interventionist and families identified the family/child's top three craniofacial-related stressors or topics of interest, corresponding coping cards and caregiver booklet pages, and discussed and/or role-played potential family approaches to addressing stressors and reviewing topics utilizing the Cellie doll, coping cards, and caregiver booklet. Finally, interventionists engaged families in identification of and problem solving around anticipated barriers to independent family-directed kit use over approximately four weeks (see Table 1. for full additional study flow description).

Two and four weeks post initial visit, an interventionist contacted families for phone or Zoom booster sessions. During these visits, families were encouraged to continue using the kit and asked to describe how they have used the kit over the last two weeks. Additionally, barriers to kit use were problem-solved, the previously reported top three stressors/ topics were reviewed and if the family wished to shift topics, the interventionist assisted the family in determining how to use the kit to address new stressors/topics. The 4-week booster session was also the follow-up study visit, during which caregivers and children were asked to complete a study feedback and satisfaction questionnaire to inform Cellie Kit development improvements. Families were given the option to complete post-study measures during or after the study visit. The family's study interventionist administered the questionnaires in a semi-structured format for those who opted to do so. The interventionist encouraged openness and honesty and, in an effort to reduce social desirability responding, emphasized that the goal of receiving feedback is to learn how to improve the Cellie Kit. Additionally, after each study visit, the interventionist completed a study feasibility form to log intervention delivery mode and study intervention component completion.

Cellie Coping Kit for Craniofacial Conditions Development

The current project involved the adaptation of the Cellie Coping Kit for the craniofacial community to create the Cellie Kit. Previous adaptations of the Cellie Coping Kit have edited earlier Cellie Coping Kits' coping card and caregiver booklet content to align with relevant experiences of the new medical population. The Cellie

Coping Kit's adaptation process was merged with the **information gathering** and **preliminary adaptation design stages** of the ***heuristic framework*** (Barrera & Castro, 2006). As part of the **information gathering stage**, a literature review was conducted to identify healthcare and craniofacial-related stressors for children with craniofacial conditions and Hispanic/Latinx pediatric patients and their families, as well as effective coping strategies for managing these stressors and supporting psychosocial functioning. Appropriate coping constructs from previously developed Cellie Coping Kits were then identified and a bilingual Hispanic clinic patient and expert in the field of culturally-adapted interventions for Hispanic/Latinx youth and families were informally interviewed to gain feedback on previous Cellie Coping Kit pilot study design and identify gaps in craniofacial-related stressors. Editing of previous coping kit content and development of new content was collaboratively conducted with craniofacial experts (i.e., medical (surgeons, orthodontist, speech language pathologist, nurse practitioners, dental assistant) and mental health providers (pediatric psychologists, psychology graduate students) and another clinic family.

During the **preliminary adaptation design stage**, a draft of the adapted intervention materials and activities was developed, reviewed, and refined using qualitative feedback from informants in an iterative process (Barrera & Castro, 2006). Primary adaptations included: editing previously created Cellie Kit content and developing new card and page content to address relevant stressors and strategies for the craniofacial community (e.g., learning about craniofacial conditions and potential members of their craniofacial team, understanding and accepting gear, addressing discomfort talking to other kids, building confidence, responding to people's reactions and answering condition questions), meeting reading literacy and educational systems literacy needs (e.g., aiming for 6th grade reading level within caregiver booklet, addition of 504 Plan and Individualized Education Program process explanation and resources) and education and gender inclusive language (e.g., removal of gender pronouns in English kit; including statement of inclusivity and pronoun choice explanation in bilingual Spanish kit). Content feedback centered on relevance and comprehensiveness of addressed stressors and proposed coping strategies for the craniofacial community, and developmental appropriateness of language. The Cellie Coping Kit creator, a pediatric psychologist, also reviewed and refined content. Forty-four coping cards (10 topic sections), and a 49-page caregiver book (8 topic sections and 3 resource pages) were finalized. Subsequently, Cellie dolls, English coping cards, English caregiver books and kit bags were designed and produced.

As part of the ***ecological validity framework***, within the **preliminary adaptation design stage**, the principal investigator adapted intervention materials and activities by infusing cultural knowledge across factors, focusing on language (i.e., Spanish translation with Mexican dialect reflective of clinic population), and with attention towards promoting and supporting adaptive cultural values (e.g., encouraging caregivers to connect with their religious and spiritual networks if this is identified as a helpful source of support; using the phrase *trusted adult* instead of *parent* on children's coping cards to support the involvement of diverse family networks in caregiving), to increase Hispanic/Latinx cultural sensitivity (Bernal et al., 1995). Additionally, we provided a range of suggestions of coping strategies for each stressor in an effort to provide culturally-acceptable evidence-based strategies, that also reflect an appreciation of generational differences in cultural, socioemotional, and health-related beliefs that may exist between caregivers and children (Bernal et al., 1995). For example, we developed the Parenting Tips to Support My Child page to encourage caregivers to respond with warmth and show affection towards their child. Recognizing that depending on how closely Hispanic/Latinx caregivers identify with traditional ideas of marianiasmo (i.e., nurturing, self-sacrificing woman) and machismo (i.e., strong, dominant man), they may respond differently to suggestions to encourage their child's emotional expression and demonstrate affection through various methods, or believe that encouraging expression is a sign of weakness (Sue & Sue, 2016). As such we provided a range of options to increase the likelihood that some would be within a parent's level of comfort and agreement. For example, for touch, parents who support greater use of touch for comfort might like the hug suggestion, while parents who are reticent to show affection through touch may prefer a high five or pat on the back. Alternatively, these parents may prefer to show affection through actions or attention instead of touch.

The scope of the current project includes English and Spanish kit development and pilot testing of the English kit. For the Spanish-English bilingual kit, the principal investigator worked with a bilingual Hispanic team to translate materials into Spanish. Throughout the forward and backward translation process the team emphasized concepts over direct translation, and culturally appropriate and readable language. Based on available funding from the Face the Future foundation through the UI Health Craniofacial Center, we were able to produce 13 English Cellie Kits and will produce 12 bilingual kits (25 total research Cellie Kits). Therefore, recruitment goals were to enroll 13 English speaking families. Pilot testing of the bilingual kit, which will include bilingual coping cards (one side English, one side Spanish) and a Spanish caregiver book, is planned for the near future and is not included in the scope of the current project.

Measures

Participants completed psychosocial measures (e.g., anxiety, depression, health-related quality of life) not included in the scope of the current project prior to their initial visit and at their post-intervention follow up. These measures will not be described here.

Demographics

As part of the demographic form and Psychosocial Assessment Tool-Craniofacial Version described below, caregivers reported their and their child's race and ethnicity, languages spoken at home, insurance type, highest level of parent education, family income, child's age, sex, craniofacial, other medical and psychological diagnoses, history of past or planned surgeries, and therapies.

Family Psychosocial Risk and Resilience

Caregivers completed the 15-item Psychosocial Assessment Tool- Craniofacial Version (PAT-CV) to measure the family's level of psychosocial risk by identifying a family's areas of risk and resiliency across the domains of family structure/resources, social support, parent stress reactions, family beliefs, family problems, child problems, craniofacial-specific concerns, and caregiver confidence to manage and cope with craniofacial-related care. The sibling problems domain was not included in the developer provided measure version. Items vary by child's age and many include subitems. Items are scored as either a 1 (higher risk) or 0 depending on response and then weighted as determined by the PAT-CV scoring tool. Not all items are scored because those items psychometrically blend with other items or do not sufficiently load on a subscale.

Weighted scored items are totaled and map onto Universal (< 1.00), Targeted (1.00 to 1.99), and Clinical levels (2.00+) of risk (Kazak et al., 2018; Crerand et al., 2018). More psychosocial stressors and fewer supportive resources indicate increased risk (i.e., more challenges to illness adjustment and treatment adherence) and as families rise from Universal to Clinical levels of risk, they require more individualized and intensive supports to manage distress and promote effective coping. Of note, risk scores may be underestimated due to omission of sibling questions on the questionnaire version. The PAT-CV has been found to be reliable (i.e., test-retest reliability ($r = 0.77$, $P < .0001$) and valid (i.e., internal consistency Cronbach's $\alpha = 0.86$) for use with socioeconomically diverse families of children ages 1 month to 17 years old with craniofacial conditions (Crerand et al., 2018). The Psychosocial Assessment Tool from which the PAT-CV has been adapted has been utilized in racially, ethnically, socioeconomically diverse pediatric condition samples, translated into Spanish and used with Spanish speaking caregivers (Kazak et al., 2018; Pai et al., 2008; Pai et al., 2012).

Feasibility and Acceptability Measures

Child and Caregiver Intervention Satisfaction

Children and caregivers each completed complementary Intervention Satisfaction and Feedback Questionnaires based on previously developed and utilized Cellie Coping Kit feasibility and acceptability questionnaires at the post-intervention follow up visit (Marsac et al., 2012; Marsac et al., 2014; Marsac at el., 2018). The forms were delivered in a semi-structured interview format by the family's study interventionist or completed individually by participants as a Qualtrics survey depending on family preference and time availability at the visit. The child form included 27 items and the caregiver form included 29 items. Questionnaires asked families about feasibility (e.g., kit usage, knowledge gained, barriers to use) and acceptability (e.g., satisfaction with individual Cellie Coping Kit components, design, and content such that children were asked about the coping cards and Cellie and parents were asked about all components). Questions were asked in a mixed format of four multiple choice, 13 (child form) and 15 (caregiver form) yes/no, three Likert scale (I liked it … a lot, some or not at all) and seven open ended items. Some open-ended questions asked for suggestions for changes to make to the Cellie Kit and intervention program to improve feasibility and acceptability. Quantitative feasibility and acceptability responses were analyzed by percent of children and caregivers who selected a given response. Through thematic analysis feasibility and acceptability themes were identified within open-ended responses (Braun & Clarke, 2006; 2022).

Intervention Feasibility

After each study visit, interventionists completed a Craniofacial Cellie Coping Intervention Feasibility Measure based on previously developed and utilized Cellie Kit intervention feasibility forms (Marsac et al., 2019). This form tracked which implementation delivery mode (i.e., in-person, phone, video call) was utilized for visits, which session components (Baseline 14 items, 2-Week Booster 8 items, 4-Week Booster/Follow Up 10 items) were completed per visit, and the total/percentage of session components completed, to assess compliance with intervention implementation.

Missing Data

Regarding missing data, study interventionists followed up with families several times in order to maintain family engagement, reduce missing data on measures and reschedule missed session appointments. Regarding pre-intervention measures, we did not schedule initial sessions with families until pre-intervention measures were completed to support continued participation. Among analyzed pre-intervention measures, the only missing data was one child’s grade level. Participant grade levels were not reported. One caregiver provided duplicative responses, which were checked for discrepancies. For the two responses on the PAT-CV that impacted scoring, choices that indicated higher risk were used to maximally capture risk that families experienced during the study. Regarding post-intervention measures, one caregiver did not complete the satisfaction questionnaire due to confusion regarding the need to do so. Applicable satisfaction questionnaire responses were extrapolated from available interview data. In contrast, there were some satisfaction responses that were omitted or left blank because they were not applicable follow up questions based on previous responses. Families additionally did not receive their final remuneration gift card until post-intervention measures were completed. Data collected prior to participant drop out was used.

Data Analyses

This research was conducted within the epistemological paradigms of constructivism-interpretivism, with aspects of post-positivism (Dawadi et al., 2021). Key features guiding this work include, importance on the understanding and interpretation of participants’ experience, recognition that both the researchers’ and participants’ previous experiences, perspectives, and contexts interact to create knowledge and meaning, and subjectivity is inherent in research. To support the capture of these complex relationships, mixed-methods were applied.

To describe our sample, enrollment, feasibility, and acceptability, descriptive statistics were performed. Frequencies of descriptive statistics were calculated to describe our sample and can be found in Table 2. Families' level of psychosocial risk was reported based on PAT-CV measure scoring and calculation. Feasibility was measured by intervention implementation metrics and participant-reported feasibility and usability. Within implementation we described intervention delivery mode (i.e., video call vs. phone call) data and intervention delivery fidelity. Fidelity encompassed study visit attendance and completion rates, as well as individual intervention component completion ratios within each type of session. Among families who attended at least an initial session, we calculated intervention session attendance percentages (i.e., percentage of total visits attended --maximum 3 per family) as well as session-type (initial, 2-week booster, 4-week booster/follow-up-- attendance across families), and percentage of average intervention session-specific components completed across families --total intervention components (32); and session-specific intervention components: initial (14), 2-week booster (8), 4-week booster/follow up (10). Quantitative intervention feasibility and acceptability data were summarized using frequencies from parent and child responses on the Intervention Satisfaction and Feedback Questionnaire.

Thematic analyses were performed to identify additional qualitative feasibility and acceptability data within patient and caregiver written and oral open-ended and semi-structured interview responses as part of 2-week booster and 4-week booster/follow up appointments and verbal completion of the satisfaction and feedback questionnaires. Procedures used both direct participant statements as well as interpreted meanings to identify patterns across participant experiences using the Cellie Kit (Braun & Clarke, 2006). We utilized a codebook based thematic approach in which a codebook was used to provide a structure to support code organization, while still capturing developing analyses, and viewing research subjectivity as a resource and tool (Braun & Clarke, 2022).

Thematic analysis procedures included the following six phases: 1) familiarization with the data through reading open-ended responses and interview notes, 2) initial data coding within a combined transcript (i.e., generating inductive codes), 3) extrapolating potential themes from the data 4) reviewing and revising potential themes (i.e., creating and iteratively updating the codebook and themes during theme review and consolidation meetings), 5) naming and defining agreed upon themes, 6) presenting finalized themes and subthemes (Braun & Clarke, 2006)). In phase one, the principal investigator read through child and caregiver open-ended responses and family session interview notes for the four families to familiarize herself with the scope of the data. In phase two, data were coded by hand by the principal investigator, another doctoral--level graduate student, and the dissertation chair psychologist. Each family’s data was coded by two researchers. As part of stage three, through an initial review of the data, previous Cellie Kit manuscripts, and review of the feasibility and acceptability questionnaire, the principal investigator identified six a priori categories (later refined) to create an initial structure for the codebook: Usability, Approaches to Using Cellie Kit, Barriers to Use and Suggested Changes, Acceptability and Factors that Influence Acceptability Relevance and What Families Learned. Stages three through five blended into an iterative process. They consisted of data and theme review and consolidation meetings in which research team members met to read through transcripts together, review inductive code determinations, and develop consensus on emerging themes, subthemes, data labeling and codebook definitions. The researchers iteratively and collaboratively re-reviewed the data, developed the codebook and assigned data to representative themes and subthemes. Final themes/subthemes, codebook definitions, and participant response examples are listed in Table 6 and described in the results. Feasibility- related themes are presented after measure-based feasibility results and acceptability themes are presented after measure-based acceptability results. Qualitative themes were integrated with quantitative data to draw conclusions about initial Cellie Kit feasibility and acceptability. Missing quantitative data were omitted from analyses or abstracted from qualitative data if available.

Results

Participants and Enrollment

A total of 28 children and their caregivers were offered an opportunity to participate in the study: 6 children and either 1 or 2 of their caregivers were enrolled (21.4% participation rate) (see Figure 2. for Recruitment and Enrollment Flowsheet). Enrolled children ranged in age from 6 to 10 years old (M = 8.83 years old). Two families were lost to follow up; one after completing parent baseline measures and the other after attempts to reschedule their 2-week follow up. Four families (66.67%) completed all study intervention visits. Follow-up assessments were completed by 4 children (80% follow-up rate) and 3 caregiver sets (60% follow-up rate). We reached out to families often to maintain family engagement, reduce missing data on measures, and reschedule missed session appointments.

Our enrolled sample represented four unique craniofacial conditions (cleft lip and palate (50%), microtia (16.67%), Apert's Syndrome (16.67%), Osteogenesis Imperfecta (16.67%)) with a majority of youth diagnosed at birth (66.67%). Several children were also diagnosed with comorbid medical diagnoses and all children held at least one comorbid psychiatric diagnosis (see Table 2). Regarding treatment, all children participated in at least two therapies (e.g., speech, psychotherapy), two-thirds had at least three surgeries, and one third are scheduled for upcoming surgeries. Almost all youth (83.33%) receive school supports through a school 504 Plan or Individualized Education Program. Detailed child demographics can be found in Table 2.

Enrolled families included four mother-child dyads and two mother-father-child family units. A majority of enrolled parents were non-Hispanic White (62.5%) and English was the only language spoken in two thirds of homes; bilingual families also spoke Polish or Spanish. Families came from diverse socioeconomic backgrounds with family incomes ranging from <$15, 000 - $150,000 to $199,999 and half reported an income of $35 to $49,999. Children's insurance coverage was split between Medicaid and private insurance. With respect to psychosocial risk, half of study families fell within the Universal range and the other half within the Targeted psychosocial risk range. Additional caregiver and family details are found in Table 2.

Feasibility

Aim 1 sought to explore the feasibility of remotely delivering the psychosocial intervention program with regard to implementation and participant reported feasibility and usability. Regarding study implementation, all study intervention procedures were able to be delivered remotely by video call or over the phone and occurred at an about equal rate (54% vs 46% respectively) as shown in Table 3. All baseline appointments were video calls, but given a format choice for follow-up visits, families preferred phone call follow ups (75% vs 25%). Implementation fidelity targets (i.e., a majority of sessions completed at 80% or greater session fidelity) were met with 86.7% of intervention sessions (baseline, 2-week follow up, 4-week follow up) completed among families who attended the baseline visit. Additionally, over 95% of intervention session components were completed on average across each session type (See Table 3. for number of intervention components per visit).

Regarding feasibility and usability, on family-reported study measures, all caregivers utilized the caregiver book, all children played with the Cellie doll and most families engaged with the coping cards (75%) as presented in Table 4. Caregivers reported that the introduction to the Cellie Kit was helpful (100%) and all children reported understanding how to use it. Concerning specific kit components, caregivers reported that the caregiver book was easy to use and understand (100%). The coping cards received mixed feedback. All children who used the coping cards reported that they were easy to use; however, one child did not use the coping cards (parent did not introduce them to child), one child used the cards independently, and two utilized the cards alongside caregivers. Some parents felt that the coping cards were easy for their child to use and understand. All families indicated that they used the Cellie Kit at least 2 to 5 times throughout the study, all at home and half in medical settings. All caregivers and children reported that they plan to continue using the Cellie Kit in the future.

Similarly, several qualitative themes additionally supported quantitative findings suggesting that children and caregivers found the intervention feasible (see Table 6.).

Concerning *Usability,* within the subtheme of *individual participation and kit use*, specific kit component use varied by and within families. Some caregivers individually and some used it with their child; coping cards were used in all combinations – as a family, child individually, parent individually, and not at all – and the Cellie doll was always used by the child alone. Parents and children's comments about what made the kit easy to use were grouped into the subtheme of *ease of use.* They included implications that they received clear guidance and instruction around Kit use, they could pull from past therapy experiences, and the design of materials supported ease of use. One set of parents expressed feeling "confident" moving forward and another parent reported "using Cellie was easy for us" and felt that their past experience in pain therapy had primed them as they "knew how to use [therapy/coping] tools." Regarding design, one family liked the "portable" and separatable design so that "if there was like a specific topic that she was struggling with, that you needed to like share with grandparents or share with friends, that would be really easy to just like show them a couple of the cards, instead of give them the whole book." Families' *frequency and length of use* ranged considerably between and within families. Some families used coping cards and the caregiver book about once a week, while one used the caregiver book 7 to 8 times. Cellie doll use ranged from one time to "all the time." Time per each kit interaction varied from 15 minutes to an hour at a time. As for *locations of use*, while all families used the Cellie Kit at home, half of families brought it to medical settings including urgent care and the hospital and one parent reported applying the caregiver book to support her child at a doctor's appointment. Regarding *plans for future use*, all families reported that the Cellie Kit will be helpful for future surgeries as a preparation tool and at the surgery itself. Additional potential uses include at clinic appointments and as a future resource. For example, one father stated “We hope as [our daughter] grows older and has more concerns regarding her condition the kit will be helpful.”

The many ways that families approached using the kit as a whole and its components reflected feasibility and were captured within the broader theme of *Approaches to Using the Cellie Kit* (See Table 6.). The subtheme *flexible child-centered approaches* demonstrated that families interacted with the kit and its materials in ways that met their child’s learning style, needs, and parent and child engagement styles. For example, one participant (10-year old) reported "was very independent and was able to find answers on her own," while hands-on parent approaches were more common with younger children. With the caregiver booklet, some parents read parts prior to introducing materials to their child and some learned alongside their child. Moreover, families used kit components differently - children used the Cellie doll for comfort, play, or storage, while the coping cards and caregiver booklet were used for learning and teaching. Another subtheme was *relationship-centered approaches* in which using the kit was a family activity in which families used the kit together, collaboratively, and as a “bonding tool.” For instance, in one family the child read a coping card to her parents, they talked together about the subject and then parents referred to the corresponding booklet page to look for additional ideas. Parents also highlighted using a *tool-oriented approach*; viewing the Cellie Kit as a preparational, informational, quick reference and topic introduction tool. One mother noted that the kit “helps give a quick answer,” another called it a "conversation starter” and a father noted that the kit can help “prepare for things to come.”

The theme of *Feasibility Barriers and Suggested Changes* captured the difficulties families ran into engaging with or using the Cellie Kit and their suggestions for improvement. One set of challenges focused on *intervention delivery or content - related barriers* and included areas for growth in supporting families' understanding of content and how to use individual kit components. For instance, one parent noted that "For other kids, they may need more hands-on direction from the therapist [on] how to use it and how to understand how helpful it can be if they haven't been in therapy before." Families also brought up concerns regarding the developmental appropriateness of language within the coping cards as a parent described the cards as "hard to follow word for word" and some language as "more grown up." Families provided suggestions for *changes to the intervention format, delivery and content* that included requests for an online version or virtual component (e.g., application for a tablet) and a version for younger children that included fewer words and more pictures to help explain content. Another barrier theme pertained to *physical or functional barriers* in which there were certain aspects of kit materials that made it difficult to use or were not esthetically pleasing. For example, the physical design of the coping cards was challenging for a child with fine motor difficulties to manipulate and Cellie's appearance (e.g., eyes) was also initially off putting for some. Another child said the face made her nervous and was a "little scary." *Physical and functional suggested changes* included adding page numbers to the coping cards that correspond to the caregiver booklet page, and attaching a keychain or ring to the coping cards to keep them together, among others. Suggested changes to the Cellie doll included offering different colors, making the pocket bigger, and creating a mini keychain sized Cellie to increase transportability. Finally, families ran into barriers associated with the *realities of everyday life* which included difficulties finding time in their busy schedules to engage with the Cellie Kit, especially around the Winter holidays and participant acute illness.

Acceptability

Aim 2 encompassed the exploration of acceptability and relevance of the Cellie Kit coping topics and intervention materials to families' experiences. A summary of satisfaction questionnaire frequencies can be found in Table 5. Content related responses indicated that children and caregivers found the kit information to be trustworthy (100%), felt that the kit gave good tips and advice (100%) and they learned new information and coping tips (100%). Regarding helpfulness, most children (75%) reported that the kit helped them feel better when they were feeling challenging emotions and helped them talk about or ask questions about their condition. Caregivers reported that the kit helped them feel more confident helping their child cope with their condition (100%). Families also reported that the Kit was fun to use (100%), and they would recommend the kit to other kids and families (100%). Design focused responses indicated that the number of words in the caregiver book were appropriate (100%), there were too many words on the coping cards for some parents, the Cellie doll's appearance was likeable (100%), and the pictures on coping cards were esthetically pleasing (100%).

Qualitative themes related to *Acceptability and Factors that Influence Acceptability* were identified. A subtheme of *acceptability comments* included statements of explicit agreement and affirmations that indicated a family found the kit to be helpful, agreed with content or liked tip suggestions. For example, parents reported that the Cellie Kit is a "great tool for coping with the stress and physical/emotional discomfort of having a craniofacial condition." Illustrating specific topic areas, all families indicated that the surgeries and procedures focused sections were or would be helpful in the future, and a mother reported liking the idea of providing handouts to school to share about her child's condition. Relatedly, families reported on *likeable or engaging materials and aspects of the kit* including the broad applicability of coping tips, comprehensiveness, accessibility of information, and engaging visual presentation. Families called out individual kit components as well; one child commented "It's my favorite," referring to the Cellie doll. *Individual and family factors* were also identified as contributors and challenges to engaging with the Cellie Kit within and between families. As one father put it, "Every parent and every child is different." On one hand, comorbid psychiatric conditions like attention deficit/ hyperactivity disorder (ADHD) or autism may have contributed to difficulty attending to kit materials, understanding them, and finding materials interesting, but they also expanded the number of topics that were relevant to children's experiences. The father of an autistic child stated "the quotes and strategies are useful for parents who are in a situation where I am where the child is a hybrid of having craniofacial, along with a separate condition mixed together." At the family level, caregivers are also differentially equipped to relate to their child, discuss topics, and gauge their child's readiness to discuss topics.

Additionally, *Relevance* themes emerged as part of the acceptability of the Cellie Kit. *Topic relevance* refers to which coping card topics and caregiver booklet sections were or were not applicable to the child and family. For example, for a family with a daughter with microtia, the booklet page "I don't know how much information to share with my child about their treatment and diagnosis" felt relevant, but "helping my child understand and accept their gear" was not applicable to their experiences because the parents already made wearing hearing-aid attachments a positive experience by "gathering fashionable headbands." Children who struggled with anxiety, depression, or emotion regulation related to ADHD, found the coping card sections "Dealing with Emotions" and "Everyday Coping Strategies" like Belly Breathing relevant. In addition to topic, timing within a child's development emerged as a relevance theme. Comments around whether past, present, or in the future would be the appropriate time to be exposed to the kit in general or specific topics were captured within the theme *Timing Relevance*. Regarding the past, most parents expressed wishes that the kit had been available earlier or should be available to families with younger children. A mother noted "it would be good to start kids on this as young as possible so families can grow with it." A father noted that the kit could be helpful for children as young as 3, because having "Cellie to carry around could contribute to a calmer experience" for a young child. Regarding use of the kit in the present, when a stressor or situation occurred during study participation, it created opportunities for kit utilization and children's engagement with that material. For example, having a doctor's appointments or going to the hospital during the course of the study, lead to kit use in medical settings. Moreover, every family identified a future use for the kit, noting the value of having the kit in the present to prepare for planned or potential experiences or to have on hand for when an applicable scenarios or experiences occurs. A mother reported that one reason she joined the study was because she "wanted to know how to talk about" an upcoming surgery with her daughter.

Qualitative themes related to *What Families Learned* provided evidence that the Cellie Kit has the potential to increase families' communication skills, understanding of the condition, and ability to cope with challenges. The subtheme *parent-child communication* captured how the Cellie Kit supported new and ongoing conversations between parents and children, both increasing parents' confidence and effectiveness in conversations with their child. For example, one father reported "I learned about ways to approach things that I would not have otherwise known… like this section- helping my child manage emotions and self-esteem…There are things in there that I guess never occurred to me, or I didn't really think about in a way that would allow me to like approach [my daughter] in a way to be like: hey, how are you doing? Does this happen to you?…" Within *communicating with others,* children and parents commented on responding to people's reactions and questions related to the child's craniofacial condition generally, as well as child- peer communication and parent-school communication. One child highlighted that through Cellie she learned "How to talk about my condition more confidently." Parents reported learning how to prepare their child for other's reactions such that "If someone does say something, she will know how to answer it" and noted ways the kit supports communicating with their child's school. The theme of *psychoeducation* captured parent and child statements related to the ways in which the kit increased or had the potential to increase their knowledge about the child's condition, treatment, or coping skills. One child noted "I learned about my microtia. That you get a new ear." Another child reported "I learned some new ways to deal with my condition." Multiple caregivers reported learning more about supporting their child's emotional coping including one father reported learning how to better assess his daughter's emotional well-being and a mother learning ways to support her child's medical anxiety.

Discussion

Preliminary pilot test findings suggest that the Cellie Coping Kit for Craniofacial Conditions (Cellie Kit) is a promising new coping resource that may help address the need for accessible, evidence-based coping intervention tools that manage the challenges associated with having a craniofacial condition. Mixed-methods analyses demonstrated that families found the Cellie Kit and intervention feasible and acceptable. The study sample represented diverse experiences of families of children with craniofacial and psychiatric conditions as well as life experiences and identities external to the child's condition. Building off the strengths of previous Cellie Kits, the current kit sought to expand acceptability through a culturally minded adaptation and fully remote delivery of the intervention.

Overall, the initial study enrollment rates are lower (e.g., n = 6, 21.4%) than other studies of the Cellie Kit intervention (e.g., injury = 76%, cancer = 83%, and sickle cell disease = 75%). While research demonstrates that children with craniofacial conditions are at risk for psychosocial challenges, a lower enrollment rate may be a reflection of the broader clinic population. For instance, the degree to which children and families regularly experience challenges associated with having a craniofacial condition varies greatly depending on the condition, severity, and treatment needs and so, at time of recruitment, families who did not respond to outreach or declined to participate may not have felt that additional support was needed at that time. However, given the enrollment of two children with upcoming surgeries and another family that was approaching a surgical decision point, offering a coping tool at times of known distress such as prior to surgery, may increase enrollment. Alternatively, stigmatized beliefs and negative attitudes about mental health services and help-seeking are more common among Latinx and Black communities which make up a majority of the UI Health Craniofacial Center's population (50% and 15%, respectively) (Fripp & Carlson, 2017). Moreover, despite research participation facilitators for Black and Latinx communities including potential benefits of participation (i.e., compensation, access to new and free resources) and low risks associated with participation, they may have also experienced known barriers such as mistrust, stigma, competing demands (i.e., inconvenience and time conflicts), lack of access to information (i.e., misconceptions of research), health insurance coverage (i.e., fear of discrimination), and legal status in the United States (i.e., fear of deportation among immigrants) (George et al., Norris, 2014). Together, any and all of these beliefs and barriers may have negatively influenced families' decision not to participate in this psychology research study.

Despite low enrollment, our follow up rates are similar or higher than other Cellie Kit populations (injury = ½ to 1/3 follow up, vs craniofacial = 2/3) which may reflect clinic and study design strengths including connection to trusted care team members and interventionist continuity (George et al., 2014). For instance, the Craniofacial Center takes a team approach to care and has followed many patients since infancy with annual or biannual team visits that include psychology. Relatedly, a majority of enrolled and interested patients connected with the research team after an introduction to the Cellie Kit study by the clinic psychology team, compared to cold calls and emails, indicating that families may have felt more comfortable participating after learning about it from a trusted member of their care team. Additionally, families worked with the same interventionists from prior to enrollment through final follow up, providing ample opportunities for rapport building.

Additionally, many clinic families contend with psychosocial stressors which may have prevented them from participating in the study or completing participation. Of note, no families falling in the Clinical level of psychosocial risk participated, possibly reflecting their limited capacity to take on additional responsibilities. For the two families that were lost to follow-up, both were single mother, Latinx families, with lower incomes ($35,000-$49,000), Medicaid insurance coverage and children with two psychiatric diagnoses who may have been contending with multiple barriers to continued participation. One family was at a Targeted level of risk, thus experiencing acute distress without sufficient supportive resources at time of enrollment (Kazak, 2006), and while the other fell in the Universal range, the family had recently experienced a death in the family, which is not factored into scoring, and thus the family's distress may not have been fully captured. Future studies should utilize the PAT-CV as a clinical resource as part of the intervention to learn about family stressors and utilize that information to proactively problem solve potential barriers to continued participation. An additional future consideration is to reduce the burden of study measures, as one family dropped out prior to the child completing pre-intervention measures. Integration of offering the Cellie Kit as part of clinical care would support each of these adjustments.

While our study sample is quite small, the families who participated represent diverse craniofacial experiences including conditions, surgical histories and therapy service history. Moreover, enrolled youth and families represented a diverse range of identities and experiences with regard to racial, ethnic, and socioeconomic backgrounds, co-morbid psychiatric diagnoses and engagement with psychotherapy. Feasibility and acceptability within a complex patient population is a study strength. For example, despite potentially expected challenges to engagement for children with ADHD and Autism, they were able to engage and learn from the Cellie Kit. Additionally, caregivers reported that children's co-morbid psychiatric diagnoses expanded the number of relevant topics for their child and previous experiences in psychotherapy laid the groundwork for knowing how to utilize the coping tool. Therefore, psychiatric complexity made the kit more usable. Moreover, despite difficulties with overall recruitment, we were able to reach families at higher risk for health disparities and those experiencing significant psychosocial stress (i.e., half of intervention completer families in Targeted range). However, our sample underrepresents families of color and publicly insured clinic families which may in part be explained by not being able to recruit Spanish-speaking families because the bilingual Cellie Kit is not yet produced. Additionally, while fathers are often missing in pediatric research (Davison et al., 2017; Parent et al., 2017), half of the families who completed the intervention included father participation. These caregivers appeared to share the responsibilities of participation, likely increasing their capacity to complete the study. Of note, the gender of children who participated in the intervention was limited to female patients, thus our understanding of other genders experiences with the Cellie Kit is unknown. Additionally, while enrollment was open to children between the ages of 6 and 12, only children ages 10 years and under were enrolled, again limiting our understanding of the acceptability of the Cellie Kit to middle school-aged children. It is possible that our younger cohort reflects that the Cellie Kit is more attractive to younger children or their caregivers.

Overall, feasibility was demonstrated for both the implementation of the Cellie Kit intervention and usability of the Cellie Kit itself. Regarding implementation, compared to previous studies, initial visits in the current study were conducted via video call rather than in person. Additionally, we offered phone and video call follow-up options rather than in person or phone call, and most families preferred phone calls. Strengths of this method were reducing technological glitches that more often occurred over video and being able to match family preference to hopefully increase comfort sharing openly. However, a weakness of phone calls was that children were less often present for phone calls and it was more challenging for the interventionist to engage child participants and know when children were and were not attending to the conversation. Overall benefits to the fully remote intervention delivery included safety and comfort for families and interventionists during a global pandemic and potentially greater accessibility due to flexibility in scheduling and increased access to interventionist support. For example, interventionists offered to complete measures with all participants over Zoom and two out of five children, who completed measures, utilized this support. One potential drawback to remote delivery was that certain study components may have been more time-consuming due to the separation of certain study steps (i.e., consent, baseline measures, and initial visit) and time at initial visits related to orienting families to materials using a combination of in-hand materials, video demonstrations, and screen sharing, and technical difficulties. While enrolled families did not voice time concerns in their study feedback, some families who expressed initial interest in participating but did not enroll did. Regardless, there was high family retention once families completed their initial visit, which was regarded as a helpful visit by all caregivers. Interventionists were also able to complete nearly all planned intervention components across study visits, together indicating that virtual delivery is feasible.

Additionally, families affirmed Cellie Kit usability. As hoped, all families used their kits, took individualized approaches to use, and plan to continue use. Moreover, families valued each Cellie Kit component. However, the coping cards were easier to apply and understand for some families than others and children needed variable support and encouragement to use them. Some parents noted the need to be highly involved in their child's kit use, which aligns with Kit use expectations, particularly for younger children and children with learning and neurodevelopmental disabilities. Interventionists could expand problem-solving tactics and emphasize family approaches to use, particularly at follow-up visits, to improve trouble shooting.

As with previous Cellie Kits, one of the most common barriers to family implementation was related to time constraints and managing a busy schedule (Marsac et a., 2018). We feel that previous recommendations to integrate the intervention into standard care would increase timing efficiency for families and also support families' understanding of how and when to use the kit (Marsac et al., 2018). This would work particularly well in integrated clinics such as the study clinic, that utilize a team approach in which families meet with several members of their craniofacial team biannually or annually. As previously proposed, future research should assess feasibility of integration within teams including identifying which member(s) of the team should introduce the kit, frequency of team check-ins and who should be responsible for doing so (Marsac et al., 2018). Additional research should also pursue sustainability including funding team member training and implementation time and taking on the burden of cost for kit purchases, which would individually cost a family between $60 to $100 outside of the study (Marsac et al., 2018).

Overall, families liked the Cellie Kit and intervention as is. They offered suggestions for additional kit features that may improve engagement and usability, but did not request changes to the way the intervention was delivered, the core of the Cellie doll, design choices to have caregiver and child focused components, the content itself, nor did they identify any missing topics. Moreover, the only family content complaint was that the readability and maturity of language in the coping cards may have been advanced for younger children. Families did not comment on cultural sensitivity or appropriateness and this should be explicitly asked about by the interventionists in the future. Families' suggestions will be relayed to the Celie Kit creator and designer for discussion of feasibility of implementation of suggested changes. While families had suggestions for design improvements, some families felt that the kit design already contained many engaging visuals and accessible features.

Aim 2 sought to explore if families would find Cellie Kit coping topics and intervention materials acceptable and relevant to their experiences. Reponses across child and caregiver -reported study measures and interview responses provided support for acceptability. Overall families found the Cellie Kit acceptable, helpful, and relevant as is, and all families would recommend it to other families. They found the information trustworthy, felt that the kit provided good tips and advice for managing current and potential future challenges associated with having a craniofacial condition as well as applications for coping strategies outside of the craniofacial context. Through using the Cellie Kit caregivers and children reported that their confidence in communicating about the child's condition, treatment, emotions and coping grew. Families helped to point out that many factors influence which sections and topics are relevant to them including the nature of child's condition, individual differences between children and caregivers, the way each family has handled challenges to date, and families' experiences in school and in the medical settings. However, there were no sections of the kit that were universally noted as unhelpful or irrelevant, and all families pointed out the value of the surgery and procedures sections as this is a common experience across conditions. While the kit is designed to be used with families of children ages 6 to 12 years, many parents pointed out the value of offering the Cellie Kit to families at the start of their craniofacial journeys as the Cellie Kit would help orient caregivers to what their child may experience over time and help to prepare caregivers for the future. The Cellie doll could also act as a friendly, comforting, and familiar support as the child grows up. Thus, clinic providers should consider introducing families to the Cellie Kit early in care and re-offer or remind families to use the kit as relevant stressors occur over the course of childhood.

Limitations

Although these initial results are promising based on affirmative feasibility and acceptability findings, several limitations should be noted. Thus far, the enrollment rate has been lower than in other Cellie Kit samples, clinic families' identities are not proportionally represented (i.e., all female, majority non-Hispanic White caregivers, no pre-teens). Future recruitment will target families holding underrepresented identities. The small sample size limits generalizability of findings and statistical power, which prevented us from running pre-post analyses of psychosocial measures, and should be targeted in future work. There may have also been a self-selection bias among participants such that families who have already had positive experiences with the clinic psychology team were more likely to enroll. However, the hope for the Cellie Kit would be to expand access and acceptability of psychosocial supports such that even if families do not utilize or have access to regular psychological support, they could use this universal support tool. There was also a small study team which resulted in the primary investigator leading study intervention adaptation, developing feasibility and acceptability measures, recruiting and running a majority of participants, and conducting data analyses. While investigator subjectivity is a resource, it also limits interpretation of results and generalizability of intervention implementation. Moreover, while continuity of the study interventionist supported rapport building with families, it may have increased the likelihood of social desirability reporting by families while providing study feedback.

Additionally, cultural and linguistic acceptability of the Cellie Kit was not specifically assessed, but will be a focus of future research on the bilingual Spanish-Spanish Cellie Kit. In the future, interventionists should ask explicit questions that help to learn more about the salient aspects of a child and their caregivers' identities (i.e., religion, culture, values, domains of importance), and condition-related fears, beliefs and acceptance during the baseline visit to guide orientation to Cellie Kit topics (Beverley et al., 2022). Subsequently, at study follow up interventionists can ask about the alignment of Cellie Kit tips and advice with the families' cultural context and values and ask for explicit feedback on cultural sensitivity and language and request suggestions for improvement within the intervention and materials (i.e., update the Study Feedback and Satisfaction questionnaire to include cultural sensitivity questions).

Conclusion

Building off previous Cellie Kit interventions and integrating culturally responsive adaptations, we created a low-cost, accessible, remotely deliverable, psychosocial intervention tool that was well received by families of children with craniofacial conditions. Although most families were able to independently use the Cellie Kit after a brief virtual introduction, some benefited from additional check-ins and problem solving and would benefit from additional support, particularly prior to major treatment events. The Cellie Kit is a universal level support tool that can be used in conjunction with additional supports. Based on family descriptions of their experiences, using the Cellie Kit may promote resilience through: 1) teaching caregivers and school-aged children condition-specific coping strategies and emotion regulatory skills; and 2) promoting supportive communication between caregivers and children, as well as children with peers, caregivers with schools and families with their medical teams throughout the course of childhood experiences with a craniofacial condition. Although more research is needed to determine intervention efficacy, these results are promising and suggest that families are able to utilize the Cellie Kit. Moreover, its use may help to reduce the development of pediatric medical traumatic stress by increasing family preparedness to navigate and cope with condition-specific and healthcare-related challenges.

References

ACPA-CPF. (2018). Parameters for evaluation and treatment of patients with cleft lip/palate or other craniofacial differences. *The Cleft Palate-Craniofacial Journal*, *55*(1), 137–156. https://doi.org/10.1177/ 1055665617739564

Barrera Jr, M., & Castro, F. G. (2006). A heuristic framework for the cultural adaptation of interventions. *Clinical Psychology: Science and Practice, 13*(4), *311-316.* https://doi.org/10.1111/j.1468-2850.2006.00043.x

Bernal, G., Bonilla, J., & Bellido, C. (1995). Ecological validity and cultural sensitivity for outcome research: Issues for the cultural adaptation and development of psychosocial treatments with Hispanics. *Journal of Abnormal Child Psychology*, *23*(1), 67-82. https://doi.org/10.1007/BF01447045

Beverley, M., Holson, D. A., & Hewlett, D., Jr (2022). Patient engagement and cultural sensitivity as a strategy to improve health inequities: The solutions are as simple as they are complex. *Journal of the National Medical Association, 114*(6), 578–583. https://doi.org/10.1016/j.jnma.2022.09.005

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, *3*(2), 77-101. https://doi.org/10.1191/1478088706qp063oa

Braun, V., & Clarke, V. (2022). Conceptual and design thinking for thematic analysis. *Qualitative Psychology*, *9*(1), 3–26. https://doi.org/10.1037/qup0000196

Broder, H. L., & Wilson-Genderson, M. (2007). Reliability and convergent and discriminant validity of the Child Oral Health Impact Profile (COHIP Child's version). *Community Dentistry and Oral Epidemiology*, *35*(s1), 20-31. http://doi.org/10.1111/j.1600-0528.2007.0002.x

Broder, H. L., Wilson-Genderson, M., & Sischo, L. (2012). Health disparities among children with cleft. *American Journal of Public Health*, *102*(5), 828-830. https://doi.org/10.2105/AJPH.2012.300654

Canfield, M. A., Mai, C. T., Wang, Y., O'Halloran, A., Marengo, L. K., Olney, R. S., … & Copeland, G. (2014). The association between race/ethnicity and major birth defects in the United States, 1999–2007. *American Journal of Public Health*, *104*(9), e14-e23. https://doi.org/10.2105/AJPH.2014.302098

Carmona, R., Jones, T. A., & Rosenberg, J. (2020). Barriers to Accessing Medical Care for Hispanic Individuals With Craniofacial Conditions. *Plastic Surgical Nursing*, *40*(2), 73- 80. http://doi.org/10.1097/PSN.0000000000000302

Cole, D., Weiss, D., Kohser, K. L., Jones, C., Kassam-Adams, N., Brown-Whitehorn, T., ... & Marsac, M. L. (2017). The Cellie Coping Kit for Children with Food Allergy: A Pilot Study. *Pediatric Allergy, Immunology, and Pulmonology*, *30*(1), 39-44. http://doi.org/10.1089/ped.2016.0698

Crerand, C. E., M. Kapa, H., & Litteral, J. (2017). A review of psychosocial risks and management for children with cleft lip and/or palate. *Perspectives of the ASHA Special Interest Groups*, *2*(5), 23-34. http://doi.org/ 10.1111/j.1752-7325.2012.00338.x

Crerand, C. E., Kapa, H. M., Litteral, J., Pearson, G. D., Eastman, K., & Kirschner, R. E. (2018). Identifying Psychosocial Risk Factors Among Families of Children With Craniofacial Conditions: Validation of the Psychosocial Assessment Tool–

Craniofacial Version. *The Cleft Palate- Craniofacial Journal*, *55*(4), 536-545. http://doi.org/10.1177/1055665617748010

Crerand, C. E., Rosenberg, J., Magee, L., Stein, M. B., Wilson-Genderson, M., & Broder, H. L. (2015). Parent-reported family functioning among children with cleft lip/palate. *The Cleft Palate- Craniofacial Journal*, *52*(6), 651-659. http://doi.org/10.1597/14-050

Davison, K. K., Charles, J. N., Khandpur, N., & Nelson, T. J. (2017). Fathers' perceived reasons for their underrepresentation in child health research and strategies to increase their involvement. *Maternal and Child Health Journal*, *21(2)*, 267-274. http://doi.org/10.1007/s10995-016-2157-z

Dawadi, S., Shrestha, S., & Giri, R. A. (2021). Mixed-Methods Research: A Discussion on its Types, Challenges, and Criticisms. *Journal of Practical Studies in Education*, *2*(2), 25-36. https://doi.org/10.46809/jpse.v2i2.20

Feragen, K. B., & Stock, N. M. (2017). Psychological adjustment to craniofacial conditions (excluding oral clefts): a review of the literature. *Psychology & health*, *32*(3), 253-288. http://doi.org/ 0.1080/08870446.2016.1247838

Fripp, J. A., & Carlson, R. G. (2017). Exploring the influence of attitude and stigma on participation of African American and Latino populations in mental health services. *Journal of Multicultural Counseling and Development, 45*(2), 80–94. https://doi.org/10.1002/jmcd.12066

George, S., Duran, N., & Norris, K. (2014). A systematic review of barriers and facilitators to minority research participation among African Americans, Latinos, Asian Americans, and Pacific Islanders. *American Journal of Public Health. 104*(2), e16-31. http://doi.org/10.2105/AJPH.2013.301706.

Jacquez, F., Vaughn, L., Zhen-Duan, J., & Graham, C. (2016). Health care use and barriers to care among Latino immigrants in a new migration area. *Journal of Health Care for the Poor and Underserved*, *27*(4), 1761–1778. http://doi.org/10.1353/hpu.2016.0161

Kazak, A. E. (2006). Pediatric Psychosocial Preventative Health Model (PPPHM): Research, practice, and collaboration in pediatric family systems medicine. *Families, Systems, & Health*, *24*(4), 381. http://doi.org/10.1037/1091-7527.24.4.381

Mai, C. T., Cassell, C. H., Meyer, R. E., Isenburg, J., Canfield, M. A., Rickard, R., ... & Cho, S. J. (2014). Birth defects data from population-based birth defects surveillance programs in the United States, 2007 to 2011: highlighting orofacial clefts. *Birth Defects Research. Part A, Clinical and Molecular Teratology*, *100*(11), 895-904. http://doi.org/10.1002/bdra.23329

Marsac, M. L., Hildenbrand, A. K., Clawson, K., Jackson, L., Kohser, K., Barakat, L., ... & Alderfer, M. A. (2012). Acceptability and feasibility of family use of The Cellie Cancer Coping Kit. *Supportive Care in Cancer*, *20*(12), 3315-3324. http://doi.org/10.1007/s00520-012-1475-y

Marsac, M. L., Klingbeil, O. G., Hildenbrand, A. K., Alderfer, M. A., Kassam-Adams, N.,Smith-Whitley, K., & Barakat, L. P. (2014). The Cellie Coping Kit for sickle cell disease: Initial acceptability and feasibility. *Clinical Practice in Pediatric Psychology*, *2*(4), 389 –399. https://doi.org/10.1037/cpp0000062

Marsac, M. L., Weiss, D., Kohser, K. L., Van Allen, J., Seegan, P., Ostrowski-Delahanty, S., ... & Kassam-Adams, N. (2018). The Cellie Coping Kit for Children with Injury: Initial feasibility, acceptability, and outcomes. *Child Care Health and Development*, *44*(4), 599- 606. http://doi.org/10.1111/cch.12565

Nidey, N., & Wehby, G. (2019). Barriers to health care for children with orofacial clefts: a systematic literature review and recommendations for research priorities. *Oral Health Dent Stud*, *2*(1), 1-12. http://doi.org/10.31532/OralHealthDentStud.2.1.002

Parent, J., Forehand, R., Pomerantz, H., Peisch, V., & Seehuus, M. (2017). Father participation in child psychopathology research. *Journal of Abnormal Child Psychology*, *45*(7), 1259-1270. https://doi.org/10.1007/s10802-016-0254-5

Peck, C. J., Pourtaheri, N., Shultz, B. N., Parsaei, Y., Yang, J., Park, K. E., ... & Steinbacher, D. (2020). Racial Disparities in Complications, Length of Stay, and Costs among Patients Receiving Orthognathic Surgery in the United States. *Journal of Oral and Maxillofacial Surgery*. http://doi.org/10.1016/j.joms.2020.09.023

Sischo, L., Broder, H. L., & Phillips, C. (2015). Coping with cleft: a conceptual framework of caregiver responses to nasoalveolar molding. *The Cleft Palate-Craniofacial* -650. *Journal*, *52*(6), 640-650. http://doi.org/10.1597/14-113

Zulauf, C., Spinelli, T., & Rosenberg, J. (2018). Advocating for the child: The role of pediatric psychology for children with cleft lip and/or palate. *Plastic Surgical Nursing*, *38*(3), 114-120. http://doi.org/10.1097/PSN.0000000000000232

Figure 1.

Cellie Coping Kit for Craniofacial Conditions

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Figure 2.

Recruitment and enrollment flowsheet

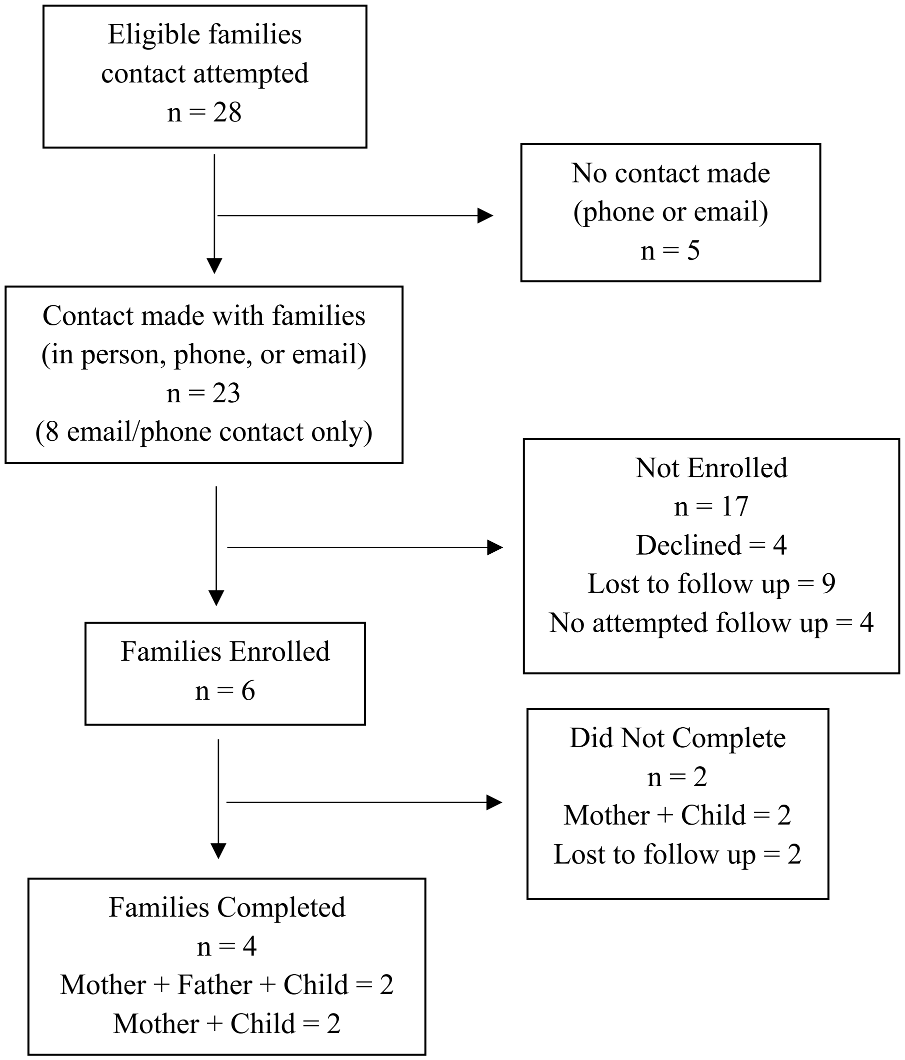


Table 1.

Study flow including timepoints, intervention activities and assessment constructs

Study Timepoint	*Intervention Activities*	*Assessment Constructs and Reporter*
Baseline Enrollment	○ Schedule Initial Visit ○ Coordinate Cellie Kit delivery	Child ○ Pre-intervention measures Caregiver ○ Demographics ○ Family psychosocial risk
Initial/Baseline Visit	○ Overview of Kit ○ Identify craniofacial-related stressors (CRSs)/topics of interest and appropriate kit coping strategies ○ Role play/discuss kit implementation ○ Problem-solve perceived barriers to kit use ○ Schedule 2-week booster	Researcher ○ Fidelity checklist
2-Week Booster	○ Review kit use and previous/new CRSs/topics, problem solve barriers ○ Schedule 4-week booster and post-intervention follow up	Researcher ○ Fidelity checklist
4-Week Booster and Post-Intervention Follow Up	○ Review kit use and previous/new CRSs/topics, problem solve barriers ○ Child and caregiver provide feedback on study and kit	Child/Caregiver ○ Post-intervention measures ○ Satisfaction questionnaire/semi-structured interview Researcher ○ Fidelity checklist

Table 2.

Caregiver and family demographic characteristics

Child Demographic Characteristics	***n* (%)**	**Participating Caregiver and Family Demographic Characteristics**	***n* (%)**
Biological Sex		Participating Caregiver(s)	
Female	5 (83.33%)	Mother	6 (100%)
Male	1 (16.67%)	Father	2 (33.33%)
Race/Ethnicity		Race/Ethnicity	
Non-Hispanic White	2 (33.33%)	Non-Hispanic White	5 (62.5% of parents)
Hispanic and Black	1 (16.67%)	Hispanic	3 (37.5% of parents)
Hispanic	2 (33.33%)		
Asian	1 (16.67%)	Caregiver Relationship Status	
Age		Married/Partnered	3 (50.0%)
6	1 (16.67%)	Single	3 (50.0%)
8	1 (16.67%)		
9	1 (16.67%)	Languages Spoken at Home	
10	3 (50.0%)	English Only	4 (66.67 %)
Medical History		English and Polish	1 (16.67 %)
Medical Diagnosis		English and Spanish	1 (16.67%)
Cleft Lip & Palate	3 (50%)		
Microtia	1 (16.67%)	Family Socioeconomics	
Apert's Syndrome	1 (16.67%)	Child's Health Insurance Coverage	
Osteogenesis Imperfecta	1 (16.67%)	Medicaid	3 (50.0%)
Sleep Apnea	1 (16.67%)	Private Insurance	3 (50.0%)
Asthma	2 (33.33%)		
Timing of Child's Craniofacial Diagnosis		Mother's Highest Level of Education	
Prenatal	1 (16.67%)	Started school but did not finish	1 (16.67%)
At birth	4 (66.67%)		
Early Childhood	1 (16.67%)	Finished high school/got certificate/GED	1 (16.67%)
Children's Surgical History			
Number of past surgeries		Started college/ university/ trade/ business school	3 (50.0%)
0	1 (16.67%)		
1	0 (0%)	Finished college/ university/ trade/ business school	1 (16.67%)
2	1 (16.67%)		
3	3 (50.0%)		
4	1 (16.67%)	Parent Work Status	
Upcoming surgery		One or both parents working	5 (83.33%)
No	4 (66.67%)	One or both parents not working	1 (16.67%)
Yes	2 (33.33%)		

Child Demographic Characteristics	***n* (%)**	**Participating Caregiver and Family Demographic Characteristics**	***n* (%)**
Cranial Vault Reconstruction	1 (16.67%)	Family Income	
Mid-face Reconstruction	1 (16.67%)	<$15,000	1 (16.67%)
Lip Closure	3 (50.0%)	$35 – 49,999	3 (50.0%)
Lip Repair	1 (16.67%)	$100-149,999	1 (16.67%)
Palate Closure	2 (33.33%)	$150-199,999	1 (16.67%)
Bone Graft	2 (33.33%)		
Ear Tubes	2 (33.33%)	Family Psychosocial Assessment	
Dental Surgery	1 (16.67%)	Psychosocial Risk Level	
Adenoids Removal	1 (16.67%)	Universal	3 (50.0%)
Hand Surgery	1 (16.67%)	Targeted	3 (50.0%)
Children's Psychiatric Diagnoses, Therapy History and School Supports		Clinical	0 (0%)
Psychiatric Diagnoses[a]			
Anxiety	2 (33.33%)		
Depression	1 (16.67%)		
Disruptive Mood Dysregulation Disorder	1 (16.67%)		
Attention Deficit/Hyperactivity Disorder	4 (66.67%)		
Autism Spectrum Disorder	1 (16.67%)		
Learning Disability	1 (16.67%)		
Therapy Type (Past/Present)[b]			
Developmental Therapy	1 (16.67%)		
Speech Therapy	5 (83.33%)		
Physical Therapy	3 (50.0%)		
Occupational Therapy	4 (67.67%)		
Individual or Family Psychotherapy	3 (50.0%)		
School Supports			
504 Plan	1 (16.67%)		
Individual Education Program	4 (66.67%)		
None	1 (16.67%)		

[a] All children had ≥ 1 psychiatric diagnosis

[b] All children had ≥ 2 therapies

Table 3.

Intervention Implementation Feasibility

	Baseline ***n* (%)**	**2-Week Booster** ***n* (%)**	**4-Week Booster/ Follow Up** ***n*(%)**	**Total Visits** ***n* (%)**
Intervention Delivery Mode				
Video Call	5/5 (100%)	1/4 (25%)	1/4 (25%)	7/13 (53.8%)
Phone Call	0/5 (0%)	3/4 (75%)	3/4 (75%)	6/13 (46.2%)
Fidelity				
Intervention session attendance	5/5 (100%)	4/5 (80%)	4/5 (80%)	13/15 (86.7%)
Average Session components completed[a]	13.3/14 (95%)	7.75/8 (96.9%)	9.88/10 (98.8%)	30.9/32 (96.7%)

[a] intervention components per session: Baseline [14], 2-Week Booster [8], 4-Week Booster/FU [10]

Table 4.

Participant Reported Feasibility and Intervention Usability

	Children ***n* (%)**	**Caregiver(s)** ***n* (%)**
Introduction received to the Cellie Coping Kit was helpful	-	3/3 (100%)
Child knows how to use the Cellie Coping Kit	4/4 (100%)	-
Used the Cellie Coping Kit	4/4 (100%)	4/4 (100%)[a]
Caregiver book easy to use	-	3/3 (100%)
Coping cards easy to use	3/3 (100%)[b]	2/3 (67%)[c]
• Child did not use coping cards	1/4 (25%)	-
• Child used coping cards by self	1/4 (25%)	-
• Child used coping cards with parent	2/4 (50%)	-
Caregiver book is easy to understand	-	3/3 (100%)
Coping cards are easy for child to understand	-	2/3 (67%)[c]
Directions on coping cards are confusing	0/3 (0%)[b]	-
Kit Components Used		
• Cellie toy	4/4 (100%)	-
• Coping cards	3/4 (75%)	2/3 (75%)
• Caregiver book	2/4 (50%)	3/3 (100%)
Frequency of use		
• Never to 1 time	0/4 (0%)	0/3 (0%)
• 2-5 times	2/4 (50%)	1/3 (33%)
• 5-9 times	1/4 (25%)	2/3 (67%)
• 10 or more times	1/4 (25%)	0/3 (0%)
Locations in which kit was used[a]		
• Home only	2/4 (50%)	2/3 (67%)
• Medical setting only	0/4 (0%)	0/3 (0%)
• Home and in medical setting	2/4 (50%)	1/3 (33%)
• Additional setting (car)	1/4 (25%)	0/3 (0%)
Would use Cellie Coping Kit again	4/4 (100%)	3 (100%)

[a] Includes abstracted response from interview for caregiver who did not complete questionnaire

[b] Does not include a response from child who did not use coping cards

[c] Includes abstracted response from interview for caregiver who did not complete questionnaire and does not include a response from caregiver who did not use coping cards

Table 5.

Intervention Acceptability

	Children ***n* (%)**	**Caregiver** ***n* (%)**
Information is trustworthy	4/4 (100%)	3/3 (100%)
Kit gives good tips/advice	4/4 (100%)	3/3 (100%)
Kit is fun to use	4/4 (100%)	3/3 (100%)
Would recommend to other kids/families	4/4 (100%)	3/3 (100%)
Caregiver book has too many words	-	0/3 (100%)
Coping cards have too many words	0/3 (0%)**	1/2 (50%)[b]
Liked appearance of Cellie some/a lot	4/4 (100%)	3/3 (100%)
Liked the pictures on Coping Cards/ in Caregiver Book some/ a lot	4/4 (100%)	3/3 (100%)
Liked the coping cards/caregiver book tips some/ a lot	4/4 (100%)	3/3 (100%)
Learned new information/coping tips	4/4 (100%)	3/3 (100%)
Kit helped me feel better when I was feeling emotions like sad, angry, and nervous	3/4 (75%)	-
Kit helped me to talk about or ask questions about my condition	3/4 (75%)	-
Kit helped me feel more confident in helping my child cope with his/her condition	-	3/3 (100%)

[b] Does not include a response from caregiver or child who did not use coping cards

Table 6.

Qualitative Themes

Themes and Subthemes	Codebook Definition	Family Quote Examples
Theme: Usability		
Individual participation and kit use	Who used which components and engaged more with the Kit overall (i.e., caregiver vs child)	*"I think when we first got it we sat down and kind of like paged through [the caregiver booklet] on our own."*
Ease of use	What made the kit easy to use (i.e., intervention delivery, aspects of materials or personal experiences)	*"I really like that it has the flash cards as well as the book. That makes it, so it could be portable."*
Frequency and length of use	How often families used the Kit and for how long they used it	*"Have used 1 time per week, so 2 times total; 20 min per time"*
Locations of use	here families reported using the Cellie Kit (i.e., home, medical setting)	*"Took it to the hospital recently"*
Plans for future use	When and how families plan to use the Kit in the future (i.e., topic, approach)	*"Cellie will be good in the future if I have any surgery"*
Theme: Approaches to Using Cellie Kit (How Families Used the Cellie Kit)		
Flexible child-centered approaches	Used kit and its components in a way that met child's learning style, needs, and parent and child engagement styles	*After hitting a wall directly reading coping cards to their 6-year-old, a father shifted tactics to engaging in other play activities like building a tower while asking questions like "When someone asks you about your ear, how does that make you feel?"*
Relationship-centered approaches	Used kit as a family activity; used kit together, collaboratively, and as a "bonding tool"	*Mother asked daughter to "pick a color" to choose their topic and then read aloud and asked questions while cuddled in daughter's bed "in the dark, with a light"*
Tool-oriented approaches	Used kit as preparational, informational, quick reference and topic introduction tool	*The topic of procedures/surgeries "got convo going" about a reconstructive ear*

Themes and Subthemes	Codebook Definition	Family Quote Examples
Theme: Barriers to Use and Suggested Changes		
Intervention delivery or content barriers	Difficulties understanding content (i.e., reading/developmental level) or how to use kit components	*Some of the coping card language is "over her head." We "weren't getting anywhere with the cards."*
Suggested changes to the intervention format, delivery, and content	Suggested changes to format of materials, presentation of content or delivery mode of content to improve accessibility and engagement	*"If they made it into an app with games and could play on tablet"*
Physical or functional barriers	Aspects of kit materials that made it difficult to use or were esthetically displeasing	*"The face at first made me nervous, was a little scary, but Cellie is a friendly monster."*
Physical and functional suggested changes	Suggested changes to the look, function or physical design of the cards, book, or Cellie toy	*"Bigger pocket, different colors of Cellie, and Keychain of Cellie for school [or] Keychain of the note cards for school."*
Realities of everyday life	Difficulties finding time to use Kit including busy family schedule, acute illness, holidays etc.	*"Don't want to use when she is feeling sick"*
Theme: Acceptability and Factors that Influence Acceptability		
Acceptability comments	Statements of explicit agreement or affirmation that indicated a family found the kit to be helpful, agreed with content or liked tip suggestions	*It "was very helpful and had great, useful ideas for how to deal with the various scenarios affecting children with craniofacial conditions."*
Likeable or engaging materials and aspects of the Kit	What families liked or found engaging about the kit and materials (e.g., broad applicability, individual components, and visual presentation)	*"I love that it has like the pictures of Cellie. And it's like colorful. And that makes it look really like interesting, for the kids."*
Individual and family factors	Contributors and challenges specific to each family, child or caregiver that impacts engagement with the Kit (e.g., psychiatric diagnoses, previous experiences, individual characteristics)	*She "has been in therapy in the past so she was open to using it and adapted it easily." vs Regarding a child with autism, she "doesn't like to talk about things," only things "she is interested in"*

Themes and Subthemes	**Codebook Definition**	**Family Quote Examples**
Theme: Relevance		
Topic relevance	Individual coping cards and caregiver booklet pages and sections that were or were not relevant to family or child because of their identity or medical condition(s)	*A mother was interested in pages on responding to people's reactions and teasing to help prepare her daughter for junior high, so that it is "not a surprise when people say something" and "to know how to communicate with her about it, when it comes up."*
Timing relevance	Comments on the appropriate time (past, present, or in the future) to be exposed to the kit generally or specific topics	*"I wish we had this when she first went to school, then you know, we could have prepared her... [for] questions that people might have."*
Theme: What Families Learned		
Parent-child communication	Kit supported new and ongoing conversations between parents and children, increasing parents' confidence and effectiveness in conversing with their child about child's condition, treatment, emotional functioning, and coping	*"Never talked about surgery before Cellie" and through using Cellie, broached the topic and learned "That [the daughter] feels pretty calm and optimistic about the idea of having surgery on her ear."*
Communicating with others	Improved skills to communicate with others (e.g., school, medical team, peers) related to the child's craniofacial condition (e.g., advocacy, approaching conversations, responding to other's comments)	*"The fact that I was able to communicate to her with the doctors ...the other day, that helped her" &* *"I learned ... to figure out how to help others around me also know" about my condition.*
Psychoeducation	Ways in which the kit increased or had the potential to increase knowledge about the child's condition, treatment, emotion regulation, or coping skills.	*"I wish we had had it earlier..." because even if "you see your kid in their day-to-day and they seem to be coping, ... that doesn't mean that we're always giving them all the tools that they could use."*

Appendix: Study Measures and Intervention Manual

Child Intervention Satisfaction and Feedback Questionnaire

Adapted from previous Cellie Interventions by Dr. Meghan Marsac

Part 1. We're asking kids to tell us what they think of the Cellie Kit. We really want to know what you think, even if there are parts you don't like. Thanks for helping up make this kit better.

1. Did you use the Cellie Kit in the last 4 weeks (since we gave it to you?)
    a. Yes _____ No ______
    b. (If yes, continue to #2. If no, skip to end)

2. What parts of the Cellie Kit did you use and did you use it by yourself, with your parents or both? (Circle everyone you used each part with)
    a. The Caregiver Book [by myself/with my parents]
    b. The Coping Cards [by myself/ with my parents]
    c. The Cellie toy [by myself/ with my parents]

3. How often did you use the Cellie Kit?
    a. 1 time
    b. 2-5 times
    c. 5-9 times
    d. 10 or more times

4. Where did you use the Cellie Kit?
    a. At home
    b. In the hospital/clinic [outpatient or inpatient visits]
    c. Other places (i.e., school, friends' house - please specify): __________________

5. Did you learn anything from using the Cellie Kit? If so, what did you learn?

6. Was there anything that got in the way of using [or made it difficult for you to use] the Cellie Kit?

7. Would it be helpful to have someone call you or meet with you more to help you use the Cellie Kit?

8. What should we change about the Cellie Kit?

9. What should we change about the way we teach families to use the Cellie Kits?

10. Thinking about the way we met for study visits. How well do you think each kind of visit went: in-person, video, and phone?

Part 2: Circle/Press “yes” if you agree with the sentence and “no” if you do not agree. There are not right or wrong answers.

1. I know how to use the Cellie Kit.	**YES**	**NO**
2. The Coping Cards have too many words.	**YES**	**NO**
3. The Coping Cards have enough pictures.	**YES**	**NO**
4. The Coping Cards are easy to use.	**YES**	**NO**
5. The Coping Cards give good advice/tips.	**YES**	**NO**
6. The directions on the Coping Cards are confusing.	**YES**	**NO**
7. Cellie and the Coping Cards are fun to use.	**YES**	**NO**
8. I think the information in the Coping Cards is true.	**YES**	**NO**
9. I learned something new from the Cellie Kit	**YES**	**NO**
10. The Cellie Kit helped me to talk about or ask questions about by medical condition.	**YES**	**NO**
11. The Cellie Kit helped me feel better when I was feeling emotions like sad, angry, and nervous.	**YES**	**NO**
12. I will continue to use the Cellie Kit.	**YES**	**NO**
13. I would tell other kids with my condition to use the Cellie Kit.	**YES**	**NO**

Part 3. Circle/Press your answer to tell us how much you like the different parts

	I LIKED IT…		
14. I like the way Cellie looks.	**A LOT**	**SOME**	**NOT AT ALL**
15. I like the tips on the Coping Cards.	**A LOT**	**SOME**	**NOT AT ALL**
16. I like the pictures on the Coping Cards.	**A LOT**	**SOME**	**NOT AT ALL**

Is there anything else you want to tell us about what you thought about the Cellie Kit or how we could make it better?

____

____

____

Caregiver Intervention Satisfaction and Feedback Questionnaire

Adapted from previous Cellie Interventions by Dr. Meghan Marsac

Part 1. We're asking caregivers to tell us what they think of the Cellie Kit. We really want to know what you think, even if there are parts you don't like. Thanks for helping up make this kit better.

1. Did your family use the Cellie Kit in the last 4 weeks (since we gave it to you?)
    a. Yes _____ No ______
    b. (If yes, continue to #2. If no, skip to end)

2. What parts of the Cellie Kit did your family use and who used them?
    a. The Caregiver Book [by myself/with my child]
    b. The Coping Cards [by myself/ with my child]
    c. The Cellie toy [by myself/ with my child]

3. How often did you use the Cellie Kit?
    a. 1 time
    b. 2-5 times
    c. 5-9 times
    d. 10 or more times

4. Where did your family use the Cellie Kit?
    a. At home
    b. In the hospital/clinic [outpatient or inpatient visits]
    c. Other places (i.e., school, store - please specify): __________________

5. Did you learn anything from using the Cellie Kit? If so, what did you learn?

6. Was there anything that got in the way of using [or made it difficult for you to use] the Cellie Kit?

7. Would it be helpful to have someone call you or meet with you more to help you use the Cellie Kit?

8. What should we change about the Cellie Kit?

9. What should we change about the way we teach families to use the Cellie Kits?

10. Thinking about the way we met for study visits. How well do you think each kind of visit went: in-person, video, and phone?

Part 2: Circle/Press "yes" if you agree with the sentence and "no" if you do not agree. There are not right or wrong answers.

1. I felt that the introduction I received to the Cellie Craniofacial Coping Kit was helpful.	**YES**	**NO**
2. The Caregiver Book has too many words.	**YES**	**NO**
3. The Coping Cards have too many words.	**YES**	**NO**
4. The Caregiver Book is easy to understand.	**YES**	**NO**
5. The Coping Cards are easy for my child to understand	**YES**	**NO**
6. The Caregiver Book is easy to use.	**YES**	**NO**
7. The Coping Cards are easy to for my child to use.	**YES**	**NO**
8. The Cellie Kit materials give good advice/tips.	**YES**	**NO**
9. The Cellie Kit is fun to use.	**YES**	**NO**
10. I think the information in the Coping Cards is trustworthy.	**YES**	**NO**
11. I learned something new from the Cellie Kit.	**YES**	**NO**
12. My child learned something new from the Cellie Kit.	**YES**	**NO**
13. The Cellie Kit helped me feel more confident in helping my child cope with his/her condition.	**YES**	**NO**
14. I will continue to use the Cellie Kit with my child.	**YES**	**NO**
15. I would recommend the Cellie Kit to other parents of children with craniofacial conditions.	**YES**	**NO**

Part 3. Circle/Press your answer to tell us how much you like the different parts

	I LIKED IT…		
16. I like the way Cellie looks.	**A LOT**	**SOME**	**NOT AT ALL**
17. I like the tips in the Caregiver Book and Coping Cards.	**A LOT**	**SOME**	**NOT AT ALL**
18. I like the pictures in the Caregiver Book and Coping Cards.	**A LOT**	**SOME**	**NOT AT ALL**

Is there anything else you want to tell us about what you thought about the Cellie Kit or how we could make it better?

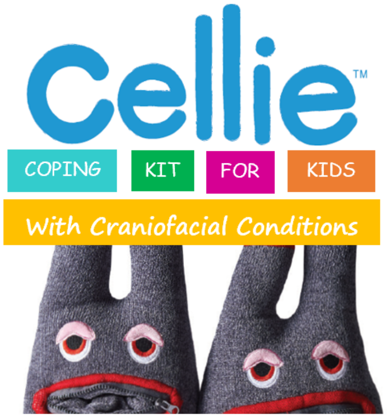
____

____

____

Daily Kit Use Tracking Log

Week 1



Date	Did someone use the kit today? (circle one)	Who used the kit? (circle)
Sunday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Monday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Tuesday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Wednesday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Thursday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Friday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No
Saturday	Yes / No	Child: Cellie / Cards Caregiver: Booklet Did you use the kit together? Yes/No

Craniofacial Cellie Coping Intervention Feasibility Measure

Adapted from Cellie Injury Intervention by Dr. Meghan Marsac

Study ID: ______ Intervention Time Point: T1, boost 1, boost 2, T2 Date: ________

Interventionist(s): ________________________ **Date: __________________**

Mode of Delivery (check which format)

___ in-person ____ phone ____ video call

Implementation of Session Components (check boxes for completed components)

T1: Initial Session:

- ☐ Introduce Intervention Program and Establish Rapport
- ☐ Speak to both the child and the caregiver
- ☐ Provide Overview of the Cellie Coping Kit for Kids with Craniofacial Conditions
- ☐ Stress the importance of caregivers and children utilizing the kit together and caregivers acting as coping teachers for their children
- ☐ Reflect the child's and caregiver's statements
- ☐ Determine 3 Stressors Relevant to the Family's Craniofacial Experiences
- ☐ Thank children and caregivers for sharing their experiences with you
- ☐ Select Relevant Coping Strategies to Address Family Needs
- ☐ Encourage families to ask questions and clarify any misunderstandings
- ☐ Ask family to discuss and role play how they would implement these tips/skills
- ☐ Identify and problem-solve any expected barriers to using the Kit
- ☐ Discuss logistics and confidentiality
- ☐ Thank family for speaking with you
- ☐ Schedule the first follow-up phone session.

___/ 14 = ____ % of components completed

Booster 1 or 2

- ☐ Rejoin with the family
- ☐ Review use of the Kit
- ☐ Troubleshoot any problems in using the Kit
- ☐ Review stressors/Potential new stressors
- ☐ Determine if coping strategies selected are being implemented, if they are helpful, and if new strategies are indicated
- ☐ Discuss logistics
- ☐ Thank family for speaking with you
- ☐ Schedule next phone session (if applicable). Or discuss study follow up.

___/ 8 = ____ % of components completed Booster 1

T2: Post-Intervention Study Follow Up

- ☐ Deliver/coordinate intervention satisfaction feedback forms completion
- ☐ Deliver/coordinate study follow up/post-intervention measures completion

___/ 10 = ____ % of components completed Booster 2 + T2 Follow Up

Psychosocial Assessment Tool- Craniofacial Version

Craniofacial version

PSYCHOSOCIAL ASSESSMENT TOOL©

(adapted all-literacy version – for all ages)

About your child (the patient):

`

Today's date: Month Day Year

Your child's date of birth: Month Day Year

Is your child: ☐ Male ☐ Female

Diagnosis: ☐ Cleft lip only ☐ Cleft palate only ☐ Cleft lip and palate ☐ Other craniofacial diagnosis: ____________

Time of diagnosis: ☐ Prenatal ☐ At birth ☐ Other: ____________

About you (person completing form): ☐ Mother ☐ Father ☐ Other: ____________

Race / Ethnicity

Patient: ☐ Hispanic ☐ Asian ☐ Black ☐ Native Am./First Nation ☐ White ☐ Pacific Islander ☐ Other: ____________

Mother: ☐ Hispanic ☐ Asian ☐ Black ☐ Native Am./First Nation ☐ White ☐ Pacific Islander ☐ Other: ____________

Father: ☐ Hispanic ☐ Asian ☐ Black ☐ Native Am./First Nation ☐ White ☐ Pacific Islander ☐ Other: ____________

How far did you get in school? (✓one box)

☐ Started school but didn't finish
☐ Started college / university / trade / business school
☐ Started university master's or doctoral program
☐ Finished high school / got certificate / GED
☐ Finished college / university / trade / business school
☐ Finished university master's or doctoral program

Work Status of the child's primary caregiver(s): (✓all that apply)

☐ One or both are working full or part-time (including self-employed)
☐ One or both are not working due to child's health
☐ One or both are not working at this time
☐ One or both are currently looking for work

1. Tell us who lives in your home: (include your child, you, and other children or adults living in your home)

First and Last Names	Age	Relationship to your child	First and Last Names	Age	Relationship to your child
		My child			
		Me			

2. Is anyone in your home pregnant or planning to adopt a child? ☐ No ☐ Yes

3. Your marital status: (✓one box) ☐ Single ☐ Married/Partnered ☐ Separated/Divorced ☐ Widowed
☐ Other (list) ____________

4. Who can you ask to help with: (✓all boxes that apply)

	Husband/ Wife/ Partner	Child's Grandparents	Other family members	Friends	People at work	Church/ Spiritual Community	Other (list)	No One
a. Childcare / Parenting								
b. Emotional Support								
c. Money / Financial Support								
d. Information								
e. Everyday tasks (for example: meals, errands, transportation)								

5. How will you get to the hospital or clinic for appointments? (✓all boxes that apply)

☐ Own car ☐ Rides from others ☐ Public (bus, subway, train) ☐ Medicaid transportation ☐ Not sure / Don't know

6. Your child's type of health coverage / insurance: (✓all boxes that apply)

☐None ☐Self-purchased / low cost ☐COBRA ☐CHIP ☐Medicaid ☐Military health care ☐Private (through work)

7. Is your family having money problems? (✓ one box)

☐ No problems ☐ Some problems ☐ Many problems ☐ It's hard to meet our basic needs

8. In what areas are you having money problems? (✓all boxes that apply)

☐ None ☐ Phone/ heat / light bills ☐ Paying rent / mortgage ☐ Buying food ☐ Car costs (upkeep /gas /insurance)

☐ School expenses / child care ☐ Medical bills ☐ Other (list): ______________________________

9. Does your child know about his or her diagnosis?

☐ Yes ☐ No, too young to know ☐ No, decided not to tell him / her

10. What is your child's current schooling: (✓all boxes that apply) ☐ **Too young for school** (go to next question)

Public or private:

☐ Head Start/Early childhood ☐ Preschool / Pre-K ☐ K-12; Grade?____ ☐ High school / collegiate institute

☐ High school certificate/diploma ☐ Dropped out ☐ Home schooled ☐ Not attending by parent's choice

☐ Attending trade / vocational / business / college / university

Special services:

☐ Early intervention (0-3) ☐ Special Education ☐ Homebound ☐ Gifted ☐ Speech Therapy

☐ Has a 504 plan ☐ Response to Intervention (RTI) ☐ Has individualized education plan (IEP)

11. Due to his/her medical problems, does your child (the patient): (✓ one box for each question)

	No	Some-times	Yes
a. Get teased or bullied a lot?			
b. Have motor skill / coordination problems (walking, throwing a ball, riding a bike, etc.)			
c. Have language problems (talking clearly, using sentences, understanding written or spoken words, etc.)			
d. Avoid situations / activities that require talking?			
e. Have problems with hearing?			
f. Have problems with vision?			
g. Get upset, worried, or avoid activities due to how he/she looks?			
h. Get upset about comments / questions from others about how he/she looks?			
i. Have sleep problems?			
j. Have problems eating or drinking?			

12. In general, does your child (the patient): (✓ one box for each question) →				**13. Is this a problem for your other children?** ☐ No other children	
	No	**Some – times**	**Yes / Getting Help**	**No**	**Yes**
a. Seem moody / change moods a lot?					
b. Seem sad or keeps to herself / himself?					
c. Have developmental problems compared to kids the same age?					
d. Been a victim of crime, or abuse, or violence inside or outside of the home?					
e. Have other medical problems besides cleft / craniofacial? (List)					
If your child is <u>under</u> 2 years old, does she or he…				**Is this a problem for your other children under 2?**	
f. Cry a lot?					
g. Have trouble with a sleeping routine?					
h. Have trouble with feeding habits?					
i. Act like she/he doesn't want to be close to you?					
If your child is <u>2 years or older</u>, does she or he…				**Is this a problem for your other children 2 or older?**	
j. Act younger than his or her age?					
k. Get upset about going to the doctor or dentist?					
l. Seem overly active or can't sit still?					
m. Have problems paying attention?					
n. Cry or get upset easily?					
o. Get distracted easily?					
p. Worry a lot?					
q. Have learning problems / problems in school?					
r. Use cigarettes, drugs, alcohol, or other substances?					
s. Act shy or cling to you or other adults?					
t. Have problems making or keeping friends?					
u. Talk about / has tried to kill or harm him/herself or someone else?					
v. Have a mental health problem? (List)					
w. Take medication for any of the issues above?					

14. Regarding your child's medical care, how confident are you that you can: (circle a number for each question below)	**Not at All**	**Some-what**	**Fairly**	**Very Much**
a. Find the time for your child's appointments (getting time off, arranging childcare, arranging travel)?	0	1	2	3
b. Follow through on your child's treatment plan?	0	1	2	3
c. Deal with the ongoing emotional effects of treatment (for example: surgery, other stressors)?	0	1	2	3

15. About the adults in your child's home… (✓one box for each question)	**No**	**Yes**
a. Has anyone had a lot of worry, fear, or anxiety at times?		
b. Have drugs or alcohol caused problems for anyone in the family?		
c. Has anyone been sad or depressed at times?		
d. Does anyone have problems learning problems or problems with paying attention or staying focused?		
e. Have there been relationship problems, fights, or talk about breaking up or divorce?		
f. Has anyone been in jail / is anyone now in jail?		
g. Has anyone been told that he or she drinks too much alcohol?		
h. Have there been legal problems with child custody or with who should raise your child?		
i. Is anyone really sick or have a serious medical problem? (List)		
j. Does anyone have other mental health problems? (List)		
k. Has anyone in the family died during the past year?		
l. Has anyone talked about or attempted to kill or harm him/herself or someone else?		
m. Has anyone been a victim of crime, abuse, or domestic violence?		
n. Are there other family problems or stresses? (List)		

16. Regarding your child's medical problems, how much do you believe. . . . (✓one box for each statement below)				
	Not true for me	**A little true for me**	**Mostly true for me**	**Very true for me**
a. My child's medical team will know what to do				
b. My child will be in a lot of pain				
c. Our family will be closer because of this				
d. I can express my concerns to the medical team				
e. My child's treatment is scary for him/her				
f. We can make good treatment decisions				
g. My child will be treated differently because of his/her medical problems				
h. My child will face a lot of complications or challenges				
i. My child is strong enough to get through this				
j. I'll be a good parent through all of this				
k. I must have done something to cause my child's medical problems				
l. We'll be able to manage this				

Thank you for your time!

Intervention Program Manual

Cellie Coping Kit For Kids With Craniofacial Conditions Intervention Program Manual

Adapted from Cellie Injury Intervention by Dr. Meghan Marsac

Introduction

The Cellie Coping Kit for Kids with Craniofacial Conditions Intervention Program is designed for children who have craniofacial conditions (e.g., cleft lip, cleft palate, microtia, Goldenhar Syndrome, Pierre Robin sequence etc.) and their caregivers. The purpose of the intervention is to provide children with a variety of potentially adaptive coping strategies, appraisals, and psychoeducation about their condition and treatment. Using the Cellie Kit, caregivers can guide children in selecting the strategies that work best for their family to decrease their distress related to living with a craniofacial condition, undergoing ongoing treatment, and the psychosocial impact of having a craniofacial condition. This intervention is intended to be used across settings (e.g., at home, at clinic, in the hospital, during procedures) and providers (e.g., with the child's nurse, doctor, etc.) with the support of the child's caregiver and an intervention researcher. Below are the craniofacial-related stressors addressed by this intervention:

- Craniofacial/treatment information sharing
- Emotional distress (anxiety, sadness, frustration) related to craniofacial condition/treatment/general
- Sleep difficulties
- Physical changes
- Social anxiety/bullying/teasing
- Connections with friends/family
- Working with the school for accommodations
- Care team member roles/ clinic visits/ medical equipment
- Procedural anxiety & pain
- Preparing for surgery and recovery
- Taking medications
- Others reactions to child's diagnosis/treatment
- Supporting siblings/other family members
- Parent self-care

This intervention manual comprises the information and materials needed by intervention researchers to conceptually understand and implement the Cellie Coping Kit for Kids with Craniofacial Conditions Intervention Program.

Content of the Cellie Coping Kit for Kids with Craniofacial Conditions

- A washable, engaging stuffed toy critter ("Cellie")
    - The Cellie toy is used to help engage the child in the Cellie Kit.
    - The Cellie toy is also integrated into some suggested coping tips (e.g., write down questions for your doctor and put them in Cellie's pocket).
- A deck of coping cards
    - Each card lists a specific craniofacial-related stressor with a selection of strategies to use to deal with that stressor.
- A caregiver book

  - Each page in the caregiver book contains a specific craniofacial-related stressor with a selection of strategies that caregivers can help their child use to deal with that stressor; the caregiver book parallels the child’s coping cards, with additional information about caregiver self-care and caring for other family members (e.g., siblings).

Three Core Constructs

These three core constructs are instrumental to the implementation of the Cellie Coping Kit for Kids with Craniofacial Conditions Intervention Program and should be evident in all stages of the program.

1. The child and caregiver are the experts on their experience and the intervention is to be tailored to their experience.
2. Parents are a natural resource for children and serve a key role in helping their child cope with their condition and treatment.
3. Children need support at the time of or in preparation for craniofacial-related stressors.

Training for Intervention Researchers

Intervention researchers require general training in child development, children’s coping, family systems, and pediatric craniofacial conditions. Specific intervention training requires the intervention researchers to practice (using role playing with the supervision of a clinical psychologist or doctoral student in clinical psychology) implementing the Session 1 and telephone booster sessions by role playing scenarios. A 4-hour interactive workshop is recommended for intervention researcher training, but individual training sessions with the PI or Faculty Sponsor are also appropriate. Practice scenarios should include each of the potential stressors and coping tips covered by the Kit. Additionally, intervention researchers should be trained on when to seek more help (e.g., concerns about depression, noncompliance with medical care), and who to contact for additional assistance (PI, Faculty Sponsor, medical team staff) should concerns arise.

For the purpose of the proposed study: Intervention researchers and the supervising clinical psychologist will meet bi-weekly throughout the intervention to ensure the intervention is being delivered consistently and address any questions/ problems that may arise. The supervising psychologist will also be available to intervention researchers to address potential questions/ concerns and should be contacted immediately if concerns arise.

Recommended Readings for Training:

1. Child development
   - Davies, D. (2004). Middle Childhood Development. In *Child Development: A Practitioner's Guide*. New York: Guilford Press.
   - Centers for Disease Control and Prevention (2009). Positive Parenting Tips for Healthy Child Development. Retrieved from http://www.cdc.gov/ncbddd/child/middlechildhood.htm
2. Children’s coping
   - Power, T. (2004). "Stress and coping in childhood: The parents' role." Parenting: Science and Practice **4**(4), 271-317.
   - Skinner, E. and M. Zimmer-Gembeck (2007). "The development of coping." Annual Review of Psychology ***58***: 119-144.

- Harbeck-Weber, C., Fisher, J.L., & Dittner, C.A. (2003). Promoting coping and enhancing adaptation to illness. In M.C. Roberts (Ed.), *Handbook of Pediatric Psychology*. New York: Guilford Press.

3. Family systems
   - Kazak, A. (2006). "Pediatric Psychosocial Preventative Health Model (PPPHM): Research, practice, and collaboration in pediatric family systems medicine." Families, Systems, & Health **24**(4): 381–395.
   - Smith, C. (2006). "Children's coping strategies and coping efficacy: Relations to parent socialization, child adjustment, and familial alcoholism." Development and Psychopathology **18:** 445-469.
   - Kliewer, W., Fearnow, M., & Miller, P. (1996). "Coping socialization in middle childhood: Tests of maternal and paternal influences." Child Development **67**: 2339-2357.
   - Blount, R. (1991). "The influence of environmental factors and coping style on children's coping and distress." Clinical Psychology Review **11**: 93-116.
   - Kapa, H. M., Litteral, J. L., Pearson, G. D., Eastman, K., Kirschner, R. E., & Crerand, C. E. (2019). Assessment of psychosocial risk in families of children with craniofacial conditions using the psychosocial assessment tool—craniofacial version.
4. Pediatric Craniofacial Conditions
   - Crerand, C. E., M. Kapa, H., & Litteral, J. (2017). A review of psychosocial risks and management for children with cleft lip and/or palate. Perspectives of the ASHA Special Interest Groups, 2(5), 23-34.
   - Johns, A. L., Luquetti, D. V., Brajcich, M. R., Heike, C. L., & Stock, N. M. (2018). In their own words: caregiver and patient perspectives on stressors, resources, and recommendations in craniofacial microsomia care. The Journal of craniofacial surgery, 29(8), 2198.
   - Feragen, K. B., Stock, N. M., Myhre, A., & Due-Tønnessen, B. J. (2020). Medical stress reactions and personal growth in parents of children with a rare craniofacial condition. The Cleft Palate-Craniofacial Journal, 57(2), 228-237.
   - Luquetti, D. V., Brajcich, M. R., Stock, N. M., Heike, C. L., & Johns, A. L. (2018). Healthcare and psychosocial experiences of individuals with craniofacial microsomia: Patient and caregivers perspectives. International journal of pediatric otorhinolaryngology, 107, 164-175.
   - Stock, N. M., & Feragen, K. B. (2019). Comparing psychological adjustment across cleft and other craniofacial conditions: implications for outcome measurement and intervention. The Cleft Palate-Craniofacial Journal, 56(6), 766-772.
5. Cellie Coping Kit intervention
   - Marsac, M.L., Hildenbrand, A., Clawson, K., Jackson, L, Kohser, K., Barakat, L, Kassam-Adams, N., Aplenc, R., Vinsel, A, Alderfer, M.A. (2012). Preliminary data on acceptability and feasibility of the Cellie Cancer Coping Kit. Supportive Care in Cancer, **20**(12), 3315-3324. PMID: 22572922
   - Marsac, M.L., Klingbeil, O.G., Hildenbrand, A.K., Alderfer, M.A., Kassam-Adams, N., Smith-Whitley, K., Barakat, L.P. The Cellie Coping Kit for Sickle Cell Disease: Initial acceptability and feasibility. Clinical Practice in Pediatric Psychology, **4**(2): 389-399, 2014. doi: 10.1037/cpp0000062
   - Cole, D., Weiss, D., Kohser, K.L., Jones, C., Kassam-Adams, N., Brown-Whitehorn, T., Lewis, M., Devine, P., Marsac, M.L. The Cellie Coping Kit for Children with Food Allergy: A Pilot Study. Pediatric Allergy, Immunology, and Pulmonology, in press.

- Marsac, M. L., Weiss, D., Kohser, K.L., Van Allen, J., Seegan, P., Winston, F.K., Kassam-Adams, N. The Cellie Coping Kit for Children with Injury: Initial Feasibility, Acceptability, and Learning Outcomes, under review.

Goals and Overview of Intervention Sessions

Session 1: Introduction to the Cellie Coping Kit for Kids with Craniofacial Conditions

Goal 1: To establish a collaborative relationship with the family
Goal 2: To provide an overview of the intervention tool
Goal 3: To collaborate with the child and caregiver to identify the top 3 stressors the child is facing
Goal 4: To work with the family to select and determine how to use coping strategies for each stressor
Goal 5: To identify and problem-solve any expected barriers to using the Kit

Phone Booster Sessions:

Goal 1: To continue rapport-building with the family
Goal 2: To determine how the family is using the Kit
Goal 4: To re-visit stressors and identify potential new stressors
Goal 5: To determine if coping strategies selected are being implemented, if they are helpful, and if new strategies are indicated
Goal 3: To troubleshoot any difficulties in using the Kit

Intervention Protocol

Session 1: Introduction to the Cellie Coping Kit for Kids with Craniofacial Conditions: Identifying stressors and relevant coping strategies

The first session is for the intervention researcher and family to establish rapport, identify craniofacial-related stressors, and determine relevant coping strategies from the book and cards to be implemented by the family. The intervention researcher will meet with caregiver and child together. Initially, the Cellie intervention researchers will join with the children and caregivers by establishing a collaborative relationship. Then the intervention researcher will orient the child and caregiver to the Kit by giving a brief overview of the program (e.g. presenting the Kit, explaining the use of the Kit, and discussing follow-up booster sessions). Parents and children will be presented with the reasoning for implementing the Cellie Intervention and explore how the Kit is relevant to the family's craniofacial experience. Collaboratively, the intervention researcher and family will identify the three most pressing craniofacial-related stressors for the family. The intervention researcher will then determine which information in the book and in the coping cards is most relevant to those stressors and have the family discuss and role play how they would implement these tips/skills. Parents will be encouraged to act as key coping teachers for their children throughout this intervention program, utilize the kit across settings (i.e. at home, during hospitalizations, during procedures, at follow-up appointments) and with different providers (i.e. doctors, nurses, etc.).

Throughout Session 1, remember to:

- Stress the importance of caregivers and children utilizing the kit together and caregivers acting as coping teachers for their children
- Reflect the child's and caregiver's statements
- Speak to both the child and the caregiver
- Thank children and caregivers for sharing their experiences with you
- Encourage families to ask questions and clarify any misunderstandings

Introduction and Establishing Rapport

- **"Hi. I'm (name) from the Cellie project. I'm a (job title/role in hospital). And you are (names of participants)? What do you prefer to be called?"**
- **"I understand that (name of who has approached the family previously) spoke to you about meeting with me today. I want to thank you for taking the time to meet with me."**
- Ask how the child and family are doing today.
- Focus on getting to know the family. **"I'm interested in learning a little bit about your family." "What do you think is important for me to know about your family?"**
- Ask questions to help you better understand the family. For instance: **"How old are you (child)? Who else lives at home with the family? How old are (child's) siblings?"**

Provide Overview of the Cellie Coping Kit for Kids with Craniofacial Conditions

- **"Having a craniofacial condition and its treatment can be tough to handle. We have developed this Kit** [show Kit] **to help children with craniofacial conditions and their families come up with ideas of how deal with some parts of the condition experience."**
- Invite the caregiver or child to ask questions about the intervention program at any time during your session.
- Check in to see what they already know about the Kit and what you will be doing today: **"Do you know why we are meeting together today?" "Have you heard of this Kit?"**
- Clarify your role and the purpose of meeting together.
- Introduce the kit and how to use the Cellie Kit by introducing Cellie and cards to children. Present the book of tips for caregivers and explain how they can be used.
- Provide examples of how families may use the kit, i.e. child squeezing Cellie during procedures or caregivers and children playing a game with the Coping Cards to promote discussions about the child's craniofacial condition and its treatment.

Determining Stressors Relevant to the Family's Craniofacial Experiences

- **"Now that we've talked about the Cellie kit, let's talk about how your family might use it. Can you give an example of some of the things that are hardest for you about having a craniofacial condition and treatment?"**
- If family has difficulty naming stressors, reflect on stressors they may have already mentioned or discuss stressors commonly experienced by families of children with craniofacial conditions.
- Stress to the caregiver and child the importance of information from their points of view.
- After reviewing the list of stressors, promote a discussion with caregivers to narrow stressors down to the three most pressing: **"Of the things you mentioned, can you help pick the three that you would like to try to use the Cellie kit to help with?"**

Selecting Relevant Coping Strategies

- Thank family members for sharing with you and acknowledge any painful experiences, thoughts, or feelings they may have disclosed.

- Reflect back what they have told you and introduce concepts from the book/cards that are directly related to what they stated (i.e. if family chose the stressor of child experiencing bullying or teasing, present the child with the bullying and teasing card and the caregiver with section of book that discusses strategies to help the child during bullying or teasing).
- Repeat this for all three of the most pressing stressors they disclosed.
- Remind caregivers to act as teachers for their children utilizing the coping techniques together.
- Ask family to discuss and role play how they would implement these tips/skills
- Ask families for feedback regarding feasibility of coping strategies and encourage families to ask any questions.

Discuss logistics and confidentiality

- Review confidentiality. **"All of the information you share with me during these sessions is confidential."** (Refer back to prior discussions with study members and remind them of the consent forms they completed).
- **"I will be calling you two times over the next 4 weeks. I would like to speak with both (child's name) and (caregiver's name). We can set up a phone session at convenient times for you so we can discuss how things are going. The first phone session will take around 10-15 minutes. The first part of the second phone session will take 10-15 minutes. The second part will be the study follow up session and will take between 10 - 60 minutes. If you would like to complete the follow up study measures together, it will take closer to 60 minutes. When is a good time for me to call in two weeks?"**
- **"Do you have any other questions?"**
- Thank family for speaking with you and schedule the first follow-up phone session.

Phone Booster Sessions: 2 week and 4 week:

Phone sessions will be conducted with each participating caregiver and child. The same intervention researcher will meet and conduct phone follow-ups with each family for all sessions. The two follow-up phone sessions will be initiated 2 week and 4 weeks following the initial session. The intervention researcher will continue to build a collaborative relationship with the family and emphasize the kit's use by caregivers and children together during their medical care. The intervention researcher will discuss with the family how they utilized the Cellie Intervention throughout the past two weeks. If caregiver or child experienced any difficulties implementing intervention, the intervention researcher will collaborate with the caregiver and problem solve the difficulty. The intervention researcher will review the three most pressing stressors experienced during prior two weeks. If stressors changed, the intervention researcher will assist families in determining how to use the kit with new emerging stressors.

Rejoin with the family

- **"Hi. This is (name) from the Cellie Project. Do you remember talking with me? Is this a good time to talk?"**
- **"How are you doing today? How is (child's name)? Thank you for speaking with me today."** Ask about treatment and discuss any changes.

- **"As you know this is our (refer to number of meetings, e.g. our second of three phone sessions). It will take about 10-15 minutes to complete. At the end of today's conversation we will schedule our next session."**
- **"Did you have any questions or concerns from the last time we spoke?"**

Review use of the Kit and troubleshoot any problems in using the Kit

- **"Let's start by discussing how you and your family have used the Kit during the past two weeks."**
- Ask if the family used the Kit during the past two weeks. Clarify who used the Cellie Kit (i.e. child only, caregiver and child, or caregiver only).
- Determine how each family member used the kit:
  - Did child use the cards? How did the child use the cards?
  - How did the child engage with the cards/Cellie?
  - Did the child use the cards/Cellie by themselves or with others? If with others, with whom?
  - Who directed child's play with cards/Cellie? Did caregivers encourage child to play with cards/Cellie or did child play with cards/Cellie on their own?
  - Did the cards/Cellie seem to be helpful to the child?
  - Did the caregivers review the book? If so, where did they read the book?
  - Did caregivers use any of the strategies included in the book?
  - Was the book helpful?
  - Did the child or caregiver have any difficulties in using any part of the kit? If so, brainstorm with caregiver new ways to use the Kit and offer helpful suggestions on ways that the kit can be more helpful.

Review stressors

- **"Two weeks ago we spoke about some things that were particularly hard during that time (list stressors), are these things still challenging for your family? Are there any new challenges since last time we met?"**
- If the family notes the same stressors are still present: reinforce coping strategies and offer any new coping strategies that are relevant to the particular stressors.
- If the family adds new stressors: discuss new stressors and determine relevant coping strategies. Discuss new coping strategies with caregiver and how they can implement strategies throughout the next two weeks.

Discuss logistics

- Ask if caregiver or child has any questions or concerns.
- Close phone session by thanking caregiver and child for their time. Thank them for sharing with you and reinforce how helpful their participation in the study is for the development of the Cellie Kit intervention program.
- Schedule next phone session (if applicable). Or discuss study follow up.

Study Follow Up

Study follow up sessions will be conducted along with the week 4 booster session and involve coordinating study follow up measure completion. Families will have the option to complete follow up questionnaires with the intervention researcher over the phone/Zoom or complete on their own after the session. Families will also be asked to complete the intervention satisfaction feedback form, which they will have the choice of having the intervention researcher deliver verbally or completing it on their own after the session.