BMJ Open

Telehealth-delivered naturalistic developmental behavioural intervention with and without caregiver acceptance and commitment therapy for autistic children and their caregivers: protocol for a multi-arm parallel group randomised clinical trial

Laurie McLay,[1,2] Lisa Marie Emerson,[1] Hannah Waddington,[3] Jenna van Deurs ,[1] Jolene Hunter,[1] Neville Blampied,[4] Aaron Hapuku,[1] Sonja Macfarlane,[5] Nicholas Bowden,[6] Lauren van Noorden,[1] Mandy Rispoli[7]

To cite: McLay L, Emerson LM, Waddington H, *et al*. Telehealth-delivered naturalistic developmental behavioural intervention with and without caregiver acceptance and commitment therapy for autistic children and their caregivers: protocol for a multi-arm parallel group randomised clinical trial. *BMJ Open* 2023;**13**:e071235. doi:10.1136/bmjopen-2022-071235

► Prepublication history and additional supplemental material for this paper are available online. To view these files, please visit the journal online (http://dx.doi.org/10.1136/bmjopen-2022-071235).

Received 21 December 2022
Accepted 11 May 2023

For numbered affiliations see end of article.

Correspondence to
Professor Laurie McLay;
laurie.mclay@canterbury.ac.nz

ABSTRACT

Introduction Timely access to early support that optimises autistic children's development and their caregiver's mental health is critical. Naturalistic developmental behavioural interventions (NDBIs) and acceptance and commitment therapy (ACT) are evidence-based supports that can enhance child learning and behaviour, and adult well-being, respectively. The traditional face-to-face delivery of these approaches is resource intensive. Further, little is known about the benefit of parallel child-focused and caregiver-focused supports. The aims of this trial are to evaluate the effectiveness and social validity of telehealth-delivered, caregiver-implemented, child-focused NDBI and caregiver-focused ACT when delivered alone and in parallel, on autistic children's social communication and caregiver well-being.

Methods and analysis The study will use a randomised, single-blind clinical trial with three parallel arms: NDBI; ACT and ACT+NDBI. We will recruit a minimum of 78, 2–5-year-old autistic children and their families throughout Aotearoa New Zealand. Support will be delivered over 13 weeks using a combination of culturally enhanced web-based modules and online group coaching. Primary outcome variables include children's social communication/engagement with their caregiver as well as caregiver stress and will be evaluated using a repeated measures multivariate analysis of variance. Outcome variables are assessed at baseline (before randomisation), immediately postparticipation and at 3-month follow-up.

Ethics and dissemination The trial is approved by the Health and Disability Ethics Committee (2022 FULL 12058). The findings of this trial will be disseminated through peer-reviewed journals and national and international conference proceedings regardless of the magnitude/direction of effect. Additionally, data will be shared with stakeholder groups, service providers and health professionals.

Trial registration number Australian New Zealand Clinical Trials Registry (ACTRN12622001134718).

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This trial will consist of three support arms, thus enhancing our understanding of the relative and combined effectiveness, and social and cultural acceptability of acceptance and commitment therapy (ACT) and naturalistic developmental behavioural intervention (NDBI).
⇒ Telehealth-delivery offers an equitable and accessible method of delivering caregiver-focused and child-focused support, that has scarcely been examined.
⇒ The ACT and NDBI programmes developed for this study were created in consultation with the autistic community and the tangata whenua (indigenous people) of Aotearoa New Zealand.
⇒ Our multi-arm randomised clinical trial compares three active interventions, so all participants will receive support.
⇒ The ACT and NDBI programmes are currently only available in English which prevents non-English speaking caregivers benefitting from these supports.

INTRODUCTION

Autism/takiwātanga (a te reo Māori term for autism meaning 'my/their own time and space')[1] is a form of neurodiversity characterised by differences in cognitive, sensory and social processing. Reported rates of autism among children in Aotearoa New Zealand (NZ) have increased from 1.2% in 2014 to 2.5% in 2021.[2] This is reflective of an overall global trend suggesting that 1 in 44 children are autistic.[3] (Identity-first language has been used in this protocol

following consultation with the autistic community in Aotearoa New Zealand).

The increased recognition and rates of autism accompany a contemporaneous rise in demand for services. This has resulted in an unmet need for support, posing a major challenge for health systems. For example, in NZ, there is a 2 to 3-year delay between primary caregivers or parents (henceforth, caregivers) seeking initial support and receiving a diagnosis.[4 5] Consequently, many children do not receive support until school age.[5] Timely, evidence-informed early supports can maximise learning and development for autistic children.[6 7] Naturalistic developmental behavioural interventions (NDBIs) are a category of supports informed by behavioural and developmental theories that are implemented within natural contexts. They teach skills in a developmentally sequential manner using behavioural techniques, naturalistic teaching strategies, and shared control between the adult and child.[8] NDBIs can be comprehensive, focusing on supporting skills in a range of developmental domains (eg, adaptive behaviour, language, play), or have a more specific focus (eg, targeting social communication alone).[8] Common elements of NDBIs include an emphasis on relationship building, face-to-face interaction, following the child's lead, use of positive affect, language modelling, responding to communicative attempts, natural rewards and direct teaching. Many NDBI programmes teach caregivers to be the primary support agents, thus providing opportunities for NDBI techniques to be implemented throughout the day and across environments. This may facilitate generalisation and maintenance of skills over time, as well as providing the most meaningful social context for learning.[9] Recent meta-analyses found evidence that NDBIs support development of social communication, language, play and cognition in young autistic children.[10 11]

While many support options for autistic children focus on improving child outcomes (child-focused supports), directly supporting the well-being of the family may also facilitate meaningful change.[12] Parenting an autistic child brings much joy,[13] however, research also suggests that caregivers of autistic children can experience high rates of stress and poor psychological well-being.[14 15] Although child-focused supports can lead to secondary improvements in caregiver mental health, they do not directly provide caregivers with tools to manage their own well-being.[16]

Acceptance and commitment therapy (ACT) is a third-wave cognitive behavioural therapy that focuses on the way people respond to internal experiences (eg, thoughts and feelings) through six core processes: acceptance, defusion, contact with the present moment, self-as-context, values and committed action.[17] Recent systematic reviews suggest that ACT for caregivers of autistic children can contribute to reductions in stress, depressive symptoms and anxiety; improvements in caregiver competence and quality of life;[18 19] and can enhance child well-being and behaviour.[20]

Māori, the tangata whenua (indigenous people) of NZ, continue to face significant disparities in access to healthcare compared with non-Māori due to systemic racism and economic hardship, stemming from the ongoing impacts of colonisation.[21] This is true of other indigenous populations globally, such as in Australia,[22] the USA and Canada.[23] To address these inequities, it is essential for ethnic minorities to have access to culturally appropriate supports. However, both ACT and NDBI are culturally bound therapies grounded in assumptions from Western society. As culture strongly influences understanding autism and supporting autistic people,[24] evidence-based Western support options may not be appropriate for people from non-Western cultures and may act as a barrier to indigenous populations accessing and/or benefiting from these supports.[25] Cultural considerations are necessary to ensure the available supports also meet the needs of indigenous populations.

Technological innovations have resulted in a surge in telehealth-delivered support, with the recent COVID-19 pandemic, and long waitlists for services, further highlighting the need for remote access.[26] Telehealth provides consumers with additional ways to access supports as programmes can be accessed anywhere, any time and at minimal cost.[26] This may help reduce inequities in healthcare access faced by Māori and other ethnic minorities. While telehealth-delivered models of NDBI and ACT have been developed, no studies have examined the relative or combined effect of both caregiver-focused ACT and child-focused NDBI delivered via telehealth, nor those which have been culturally enhanced for use with indigenous and non-indigenous populations alike.

The proposed study will evaluate: (1) the effectiveness of culturally enhanced telehealth-delivered, caregiver-implemented, child-focused NDBI and caregiver-focused ACT when delivered alone and in parallel, on children's social communication/engagement with their caregiver and internalising and externalising behaviour, as well as caregiver mental health and well-being; and (2) the social validity and cultural acceptability of telehealth-delivered NDBI and ACT. We hypothesise that: (1) telehealth-delivered NDBI will result in improvements in children's social communication/engagement as well as internalising and externalising behaviour; (2) telehealth-delivered ACT will result in improved mental health outcomes for caregivers; (3) parallel delivery of NDBI and ACT will deliver the greatest benefits in children's social communication/engagement, and internalising and externalising behaviour, as well as caregiver mental health, and be rated most favourably; (4) each programme will result in improved outcomes for both Māori and non-Māori participants; and (5) caregivers will rate each programme as acceptable, feasible and culturally appropriate.

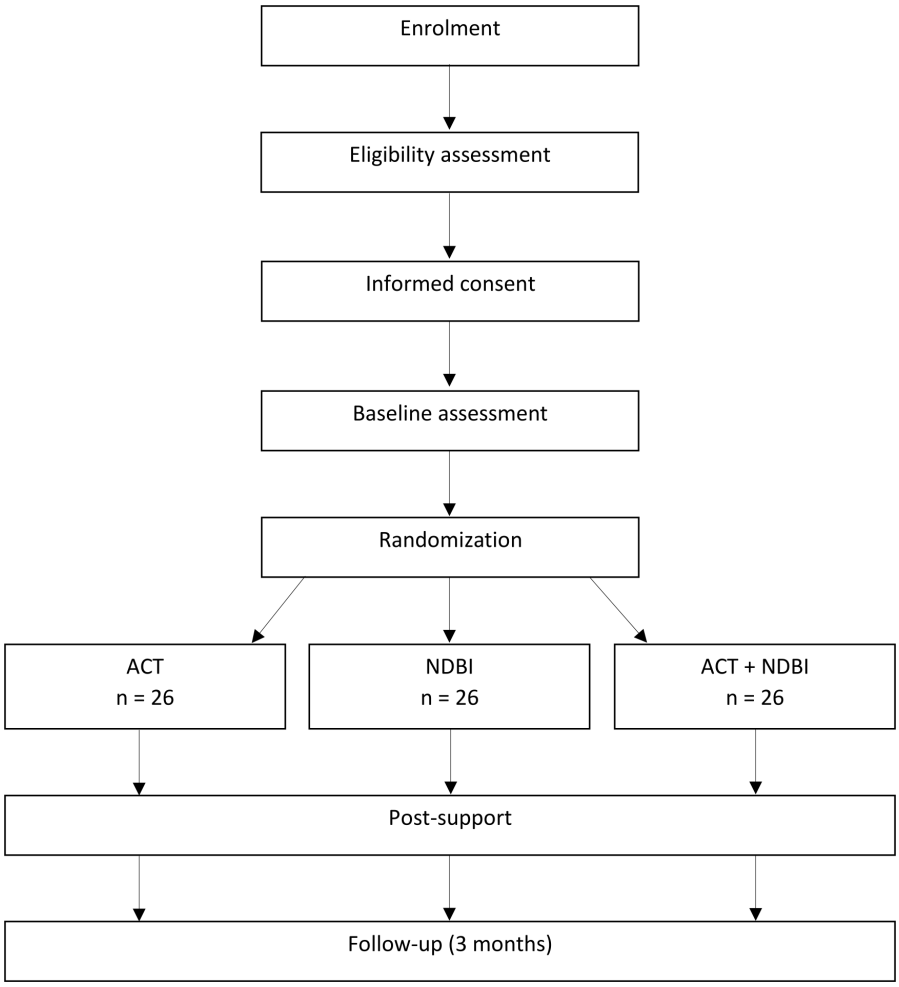

Figure 1 Consort participant flow chart identity-first language has been used in this protocol following consultation with the autistic community in Aotearoa New Zealand. ACT, acceptance and commitment therapy; NDBI, naturalistic developmental behavioural intervention.

METHODS

Design

This will be a multi-arm parallel group, single blind (rater), randomised clinical trial. Caregiver–child dyads will be randomly allocated to one of three support arms: ACT alone, NDBI alone or NDBI+ACT. Randomisation will be performed as block randomisation with a 1:1:1 allocation using minimisation and stratification by ethnicity (participants who self-identify as Māori or non-Māori). Participant flow throughout the trial is depicted in figure 1. This study and protocol were developed in accordance with the Standard Protocol Items: Recommendations for Interventional Trials,[27] the Consolidated Standards of Reporting Trials (CONSORT)[28] statement and the CONSORT extension for multi-arm parallel-group randomised trials.[29]

Eligibility criteria

Participants will include a minimum of 78 caregiver–child dyads who meet the following inclusion criteria: (1) the child has a formal diagnosis of autism or has been identified as having a high likelihood of being autistic by an appropriately qualified clinician and confirmed by a 'high likelihood' score on the Social Attention and Communication Surveillance-Revised (SACS-R) or SACS-Preschool (SACS-PR)[30] instrument; (2) the child is between 2 years 0 months and 5 years 11 months of age at the first point of contact (ie, screening) and is not attending primary school; (3) the primary caregiver speaks sufficient English to understand study requirements and written content, and to participate in group sessions; (4) the primary caregiver consents to randomisation; (5) the child is not receiving >15 hours a week of professionally delivered support; (6) the primary caregiver is not currently engaged in another parenting programme and (7) the child does not have a sibling or twin already participating in the study.

Recruitment

Participants will be recruited throughout NZ using a flyer shared on social media and distributed to community and cultural organisations, support groups and service providers. Referrals from service providers and self-referrals will be accepted via the programme website or direct contact with the research team.

Setting

Screening will be conducted via phone. The administration of the SACS-R and SACS-PR will be conducted in person at prospective participants' homes, at university clinics or via Zoom (those residing outside of university sites). Information and consent forms (see online supplemental file 2) will be emailed or posted to eligible caregivers, depending on preferences. Caregivers will access web-based educational content via the programme website (www.waioratamariki.org.nz) and group coaching will be delivered via Zoom. Post-participation interviews will be conducted via Zoom or phone, and paper copies of assessment tools and outcome measures will be posted to caregivers or administered over the phone. Video recordings of caregiver–child interactions will be recorded by caregivers within their homes.

Randomisation and blinding

Computer-generated fixed block size randomisation with an allocation ratio of 1:1:1, will generate a random allocation order, stratified by ethnicity. An independent statistician will conduct the randomisation process and give the randomisation schedule to an independent researcher who will enclose assignments in sequentially numbered sealed envelopes. When each caregiver has completed the enrolment phase the project coordinator will open the next envelope and inform the caregiver which condition they have been allocated to. This trial is single-blinded. Caregivers and therapists will be aware of the allocated condition, whereas outcome assessors will be blind to the allocation. An independent statistician who is blind to participant allocation will conduct key analyses of the specified primary and secondary outcome measures. Investigators will conduct subsidiary analyses after the blind is broken.

Programs of support

Caregivers will engage in the ACT and NDBI programmes over 13 weeks, including six (self-directed) web-based modules (released two weekly) and seven caregiver group sessions conducted via Zoom. Caregivers will be randomly divided into cohorts of up to six people who will participate in group sessions together. Group sessions will last up to 2 hours. Session one will orient caregivers to the programme and introduce caregivers and facilitators. Subsequent sessions will include a module summary, discussion of homework and caregiver reflections, trouble-shooting, and tailoring strategies to the individual and their family. All group sessions will follow elements of Māori tikanga, such as opening and closing the session with a karakia (prayer) and draw on hui and pōwhiri processes (rituals of encounter).[31] Te reo Māori terminology will be incorporated within programme content and resources.

Group sessions will be cofacilitated by a caregiver of an autistic child who is not part of the study and a trained practitioner (ie, psychologists, intern psychologists or PhD students). Where feasible, groups that include Māori participants will be cofacilitated by a Māori caregiver of an autistic child. Caregiver cofacilitators will not need to be trained in ACT nor NDBI as their role is to relate to the experience of raising an autistic child and facilitate related conversations, as opposed to providing therapeutic advice. However, they will be provided with access to modules prior to coaching sessions. There will only be a small number of clinical and caregiver facilitators in order to maintain integrity in programme delivery (ie, to ensure consistent levels of training and performance).

Online modules will take up to 1 hour to complete. Web-based content is presented in a range of formats, including written text, video, pictures, comics and audio files. Caregivers will receive guidance on strategies to practice in the home to assist in the application of newly learnt skills. Caregivers will also be given the opportunity to engage in a therapist-monitored, online discussion forum with others in their cohort.

An outline of both programmes is presented in table 1.

ACTion in Caregiving

ACTion in Caregiving is a parent-focused programme based on ACT, which was developed for this project to teach strategies to reduce caregiver stress. Each module and its associated group session will focus on one of the six core components of ACT.

The ACTion in Caregiving website includes te ao Māori (Māori worldview) imagery (eg, koru designs) to symbolise parent growth, nurturing of their well-being, caregiver–child connections and mindfulness. Established ACT techniques and metaphors have been adapted to suit an NZ audience. Participants will receive written instructions during the modules to engage in worksheets, experiential exercises (eg, thought suppression strategies) and therapeutic activities (eg, mindfulness meditations) to facilitate understanding of ACT concepts and consolidate learning.

Play to Learn

The Play to Learn programme was created for this research, and is based on the principles of NDBI. Play to Learn is designed to teach caregivers strategies to support the development of a broad range of social communication skills in their children.

Te ao Māori imagery for the Play to Learn website uses ngutukākā (plant shaped like parrot beak) and mangōpare (hammer head shark known for its strength) border designs representing communication and strengthening social connections. The online modules include video exemplars of NDBI techniques which represent families from a diverse range of cultures (eg, Māori, Pacific peoples). While completing the modules caregivers will be provided with relevant resources and directions regarding how to create their own resources (eg, Social Stories). In addition, caregivers will be given several opportunities to share video recordings of themselves practicing the skills from Play to Learn, to receive feedback from facilitators.

Table 1 ACTion in Caregiving, Play to Learn, and ACTion in Caregiving+Play to Learn programme content

Structure	ACTion in Caregiving content	Play to Learn content
Introductory group	Programme overview, introductions to group members and facilitators	Programme overview, introductions to group members and facilitators
Module 1/ Group 1	Creative hopelessness (eg, futility of controlling thoughts and feelings); acceptance/willingness in the context of unpleasant internal experiences; introduction to values	Engaging your child (eg, face-to-face interactions) and following their lead (eg, joining child-chosen activity)
Module 2/ Group 2	Mindfulness (eg, awareness of present moment with attitude of non-judgement) and application within daily life (eg, mindfulness of the breath)	Engaging through play (with people and objects) and incorporating in everyday routines
Module 3/ Group 3	Identifying caregiver cognitive fusion and techniques to facilitate defusion	Principles of learning (eg, role of antecedents and consequences) as applied to teaching new skills using behavioural techniques (eg, chaining)
Module 4/ Group 4	Introduction to self-as-context and applying awareness, acceptance and defusion to this	Principles of learning applied to replacing behaviours that are harmful to the child, others or property
Module 5/ Group 5	Living by values (eg, ongoing clarification of values and extent to which caregivers are currently living by these; using values as a guide for action)	Teaching non-verbal communication using behavioural techniques (eg, modelling, creating opportunities) and visual supports
Module 6/ Group 6	Taking action to live by values (eg, goal-setting), while managing vulnerabilities and processes of psychological inflexibility (eg, avoidance)	Teaching verbal communication using behavioural techniques (eg, prompting, responding to speech/ attempts) and visual supports

ACT+NDBI

Families in the combined support group will participate in ACTion in Caregiving and Play to Learn programmes concurrently. Group sessions will occur during alternate weeks.

Adherence strategies

If caregivers have clinical questions between group sessions that are not confidential, they will be directed to ask these at the next group session or on the discussion forum so the answer is available to all caregivers in their cohort. If caregivers have not logged into the web-based programme for 7 days they will receive a text or email from a researcher to ask if they require support. Follow-up phone calls will be made by group facilitators if caregivers miss a group session, to clarify reasons for non-attendance and trouble-shoot barriers. Families will be able to continue to access the web-based content independently postparticipation.

Outcome measures

Outcome variables will be assessed at baseline (before randomisation), immediately postparticipation and at 3-month follow-up. Online supplemental table shows the time points for data collection for each outcome measure.

Eligibility assessment measures

Screening interview

Screening interviews will be completed to ascertain the safety and suitability of the programme, collect demographic details, and to gather information about the child's presenting strengths and challenges and caregiver mental health.

SACS-R and SACS-PR

The SACS-R (24–30 months) and SACS-PR (31–60 months) observational developmental surveillance tools will be administered by a trained member of the research team if the child participant does not yet have a formal diagnosis of autism.[32 33] Assessors rate whether the child's behaviour is consistent with autism or not across the SACS-R (≤30 months of age) or SACS-PR (>31 to 60 months of age) items. A child is considered to have a 'high likelihood' of autism if at least three of the key items are consistent with autism.[34] The SACS-R has demonstrated high diagnostic accuracy (positive predictive value (PPV)=83%; negative predictive value (NPV)=99%), high specificity (99.6%) and modest sensitivity (62%).[34] The SACS-R+SACS-PR has also demonstrated high diagnostic accuracy (PPV=78%; NPV=99.9%), sensitivity (96%) and specificity (98%).

Primary outcome measure: child engagement

The primary child outcome is the difference in the child's engagement with their caregiver across time points. Naturalistic, in-home, child–caregiver play episodes (10 min) will be video recorded by caregivers at each time point. An adapted version of Bakeman and Adamson's[35] method will be used to code three forms of child engagement (unengaged, passive engagement and active engagement) during video observations.

Bakeman and Adamson's[35] procedure has excellent inter-rater reliability (intra-class correlation coefficients (ICC)=0.94).[36]

Primary outcome measure: caregiver

Parenting Stress Index-Fourth Edition, Short Form

The primary caregiver outcome is the difference in caregiver stress, as measured by the Parenting Stress Index-Fourth Edition, Short Form (PSI-4-SF) Parental Distress (PD) subscale, across time points.[37] The PSI-4-SF is a 36-item abbreviated version of the caregiver-report measure. It evaluates stress in the child–caregiver system by summing scores across PD, Parent-Child Dysfunctional Interaction (P-CDI) and Difficult Child (DC) subscales to provide a Total Stress scale score. Higher scores indicate higher parenting stress. Within the current study, only the PD subscale will be administered as this is directly relevant to trial aims and has high validity and reliability (α=0.88), whereas P-CDI and DC subscales have been found to have poor validity for caregivers of young autistic children.[38]

Secondary outcome measures: child

Child Behavior Checklist for Ages 1.5-5

The Child Behavior Checklist for Ages 1.5-5 (CBCL) is a 99-item caregiver-report measure of child internalising and externalising behaviour and will evaluate change in child emotional and behavioural functioning across time points.[39] Scores across seven subscales (Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Attention Problems, Aggressive Behavior and Sleep Problems) are summed to provide composite scores for Internalizing, Externalizing and Total Problem Behavior scales. Higher scores are indicative of increased difficulties. The CBCL has strong construct validity, high internal consistency (Internalizing α=0.89, Externalizing α=0.92, Total Problems α=0.95) and high test–retest reliability (r=0.87–0.90).[40]

Vineland Adaptive Behavior Scale, Third Edition

The Vineland Adaptive Behavior Scale, Third Edition (Vineland-3) parent/caregiver rating form is a standardised measure of adaptive behaviour. Four domain scores (Communication; Daily Living Skills; Socialization and Motor Skills) and the global score (Adaptive Behavior Composite) will be measured across time points. Higher scores are indicative of more advanced adaptive behaviours. The Vineland-3 has adequate internal consistency (Communication α=0.95, Daily Living Skills α=0.94, Socialization α=0.96, Motor Skills α=0.90, Adaptive Behavior Composite α=0.98) and high test–retest reliability (r=0.73–0.92).[41]

Systematic Analysis of Language Transcripts, New Zealand/Australia

The 10-minute video observations used for the child engagement measure will be transcribed and analysed using the Systematic Analysis of Language Transcripts, New Zealand/Australia (SALT-NZAU) V.20 software to detect change in speech and language from baseline.[42] Analyses will include total number of utterances, total number of words, total number of different words, mean lengths of utterances in words and morphemes, percentage of grammatically accurate utterances, percentage of maze words and percentage of intelligible utterances. Video coders will receive training in the SALT-NZAU V.20 software and 20% of the videos will be coded separately by two coders to assess inter-rater reliability.

Secondary outcome measures: caregiver

Depression, Anxiety and Stress Scale-21

The Depression, Anxiety and Stress Scale-21 (DASS-21) will be used to evaluate change in caregiver psychological distress from baseline. The DASS-21 yields subscale scores on three dimensions (Depression, Anxiety and Stress) as well as a Total Scale score, with higher scores indicating higher distress. The DASS-21 has been demonstrated to have high internal consistency (Depression scale α=0.88, Anxiety scale α=0.82, Stress scale α=0.90, Total scale α=0.93) and convergent and discriminant validity.[43]

Parental Acceptance and Action Questionnaire

The Parental Acceptance and Action Questionnaire (PAAQ) is a 15-item self-report measure of caregiver experiential avoidance and will be used to measure change from baseline. The PAAQ consists of two subscales, Unwillingness and Inaction, as well as a Total Scale score.[44] The Unwillingness subscale measures caregiver's ability to tolerate their child's experience of an unpleasant emotion and the latter evaluates the caregiver's ability to effectively manage their reactions to their child's affect. Higher scores indicate higher experiential avoidance. Items from the original version of the PAAQ that refer to child anxiety will be adapted to include a broader range of unpleasant child emotional states (eg, anger, sadness).[45] The PAAQ shows satisfactory internal consistency (α=0.67–0.82).[45]

Parent Sense of Competence Scale

The Parent Sense of Competence Scale (PSOC) is a 16-item caregiver-report questionnaire which will be used to assess change in caregiver efficacy and satisfaction in their caregiving role from baseline. The PSOC yields a Total score as well as Efficacy and Satisfaction scores, designed to capture caregivers' perceived competence in child rearing and enjoyment derived from caregiving, respectively. Higher scores are indicative of higher caregiver efficacy and satisfaction. The PSOC has adequate internal consistency (total score α=0.79, efficacy score α=0.76, satisfaction score α=0.75).[46]

Process measures

Social validity

The Treatment Acceptability Rating Form-Revised (TARF-R)[47] is a 20-item self-report measure that will be completed postparticipation to assess programme acceptability. Ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) are summed to provide a total acceptability score, with higher scores indicating higher acceptability. The TARF-R has adequate internal consistency (Effectiveness α=0.95; Reasonableness α=0.90; Willingness α=0.83; Cost α=0.71; Negative side-effects α=0.77; Disruption/time

α=0.69, Total α=0.92).[47 48] In addition to the TARF-R, caregivers will be interviewed using a semi-structured interview schedule to further assess the social validity and cultural acceptability of the programme.

Programme adherence and implementation

While receiving support, caregivers in each support arm will complete weekly logs of their engagement in ACT and/or NDBI practices at home, time spent engaging in these practices (min), and amount of additional professional support received (ie, type and duration). The 10-minute video observations used for the child engagement measure and language analyses will also be analysed to evaluate caregiver use of NDBI strategies. The same analysis will not be conducted for ACT strategies as they are not always observable; ACT implementation will be measured by weekly logs. Website analytics (eg, frequency and duration of engagement with website modules) and group session attendance will provide a measure of support dosage. Finally, therapist procedural integrity in delivery of ACTion in Caregiving and Play to Learn group sessions will be assessed by an independent observer for at least 20% of sessions using a checklist.

Interobserver agreement

To assess interobserver agreement (IOA), 15%–20% of all 10-minute caregiver–child video observations will be randomly selected and recoded by a second observer for child engagement, child language and caregiver use of NDBI strategies. An autistic research assistant blinded to participant allocation will code 15%–20% of original IOA coded videos. This is to obtain an autistic perspective on the data collected and strengthen interpretation. IOA will be calculated using ICC.

Patient and public involvement

Consultation with the autistic community (including autistic adults and caregivers of autistic children) has informed the development of the research questions, design and measures. We also received input from the autistic community, and Māori and Pacific people regarding support implementation, website layout, functions, usability, content and cultural appropriateness. We will provide participants with a summary of trial results and will seek patient and public involvement in the development of dissemination methods, such as how to communicate findings to the community.

Sample size

Sample size estimates were calculated a priori (power=0.80; α=0.05) and indicated that a minimum of 78 participants (caregiver/child dyads; N=26/group) is sufficient to detect a large effect (d=0.80) for the primary analysis. Effect size estimates were based on Kasari *et al*[9] which demonstrated a strong effect size (Cohen's d=0.87) following caregiver implementation of the JASPER programme with autistic toddlers. This sample size allows for 15% attrition between preparticipation and postparticipation.

Data analysis methods

First, an exploratory analysis of the data will be conducted. Then the three support groups will be compared for equivalency across variables at baseline, starting with descriptive examination. Where descriptive examination suggests that one or more groups may differ substantively from the others, the statistical significance of such differences will be tested by independent sample t-tests and χ^2 with alpha set at $p<0.05$. If substantive differences are detected, these variables may be used as covariates in subsequent analyses.

A repeated measures 3 (groups; NDBI vs ACT vs NDBI+ACT) by 3 (time; baseline vs post-support vs follow-up) multivariate analysis of variance (MANOVA) design will compare baseline scores for primary and secondary outcomes with scores postsupport, and at follow-up. Caregiver and child outcomes will be analysed separately. Depending on the statistical significance of the MANOVA findings, post hoc procedures such as discriminant descriptive analysis and discriminant ratio coefficients, will be used to describe differences among groups and identify which variables contribute to these differences.[49] Point-biserial correlation will evaluate whether categorical variables (eg, self-identified ethnicity) moderate any significant outcomes. Descriptive statistics (eg, linear regression) will be used to examine data gathered from process variables, including the relationship between process and outcome variables and differences in process variables across groups and time.

Within and between group differences may also be analysed by Modified Brinley Plots, Cohen's d (standardised mean difference) effect size with 95% CIs, and the per cent superiority effect size.[50 51] The degree of clinically significant therapeutic change will be assessed by the Reliable Change Index.[52] A one-way ANOVA will be performed to compare caregivers' overall mean satisfaction with support received across groups. Point-biserial correlation will evaluate whether self-identified ethnicity classification and caregiver autistic/non-autistic identification, moderate effects. Postparticipation interviews will be analysed using inductive thematic analysis.

We will conduct an intention to treat analysis, whereby multiple imputation is used to handle missing data. We will also conduct a per protocol analysis whereby participants who withdraw will not be included in the final comparisons. Depending on the sample size and number of partial completers we may conduct a separate MANOVA to determine the effect of supports for this group specifically. We plan to describe the reasons for missing data, clarify whether there are meaningful differences between individuals with complete and incomplete data, and any baseline characteristics which may predict non-completion.

Data collection, storage and protection

The research team will make every reasonable effort to collect data from enrolled caregivers across time points, including sending email reminders and phone calls. Access to the full participant dataset will be restricted to the research team. Each caregiver–child dyad will be allocated a unique ID code following study enrolment and information will be recorded under this code. Research data will be stored on secure password-protected servers at the University of Canterbury which are only accessible by approved members of the research team. Identifiable data and source documents will be retained for at least 10 years after the youngest child has turned 16 years old, then destroyed by secure deletion.

Data monitoring and harms

There are no anticipated serious adverse events for this trial, therefore a formal data monitoring committee is not required and no interim analyses or a priori stopping rules will be implemented. If any adverse events arise (eg, worsening of outcomes for caregivers/children), these will be recorded by the research team and reported to the relevant ethics committees as well as being reported in relevant publications as it pertains to feasibility outcomes. Participants will be in regular contact with members of the research team. If there are any perceived risks (eg, caregiver response to questionnaire indicates severe distress) we will advise participants to seek care from their healthcare professional and/or contact relevant statutory authorities, and pause involvement in the study if necessary. The study may end early for participants if the level of problem behaviours (eg, aggression or self-harm) reported by the caregiver is considered to be unsafe towards them or other family members. Involvement will also end immediately if the caregiver requests withdrawal. Caregiver reasoning for discontinuation will be recorded if they are contactable and provide an explanation.

Ethics and dissemination

This study has been approved by the NZ Health and Disability Ethics Committee 2022 FULL 12058). Any substantive protocol modifications and amendments will be submitted to the NZ Health and Disability Ethics Committee for approval prior to implementation.

Potential caregiver participant will be given detailed information sheets describing the study and are requested to contact the research team to arrange further discussion and initial screening. If they agree to participate in the study, they will be provided with a consent form, but informed that their participation is voluntary and they can withdraw at any time without consequences. It will not be possible to obtain informed consent from child participants, given their age, therefore their assent to participate and be videoed will be obtained.

Trial findings will be disseminated through peer-reviewed journals and national and international conference proceedings regardless of the magnitude/direction of effect. Data will be shared with stakeholder groups and service providers. If the supports are shown to be effective and socially valid the web-based modules have the potential to be disseminated widely.

Author affiliations
[1]Faculty of Health, University of Canterbury, Christchurch, New Zealand
[2]Child Well-being Research Institute, University of Canterbury, Christchurch, New Zealand
[3]Faculty of Education, Victoria University of Wellington, Wellington, New Zealand
[4]School of Psychology, Speech, and Hearing, University of Canterbury, Christchurch, New Zealand
[5]Institute of Education, Massey University, Palmerston North, New Zealand
[6]Department of Women's and Children's Health, University of Otago, Dunedin, New Zealand
[7]School of Education and Human Development, University of Virginia, Charlottesville, Virginia, USA

Twitter Nicholas Bowden @NickBowdenNZ

Acknowledgements The authors are grateful for and would like to acknowledge the contribution of the people within our focus groups as well as our peer reviewers.

Contributors LM, LME, HW and MR designed the study. JvD, JH, LME, HW, LM and LvN worked together to create the ACT and NDBI modules. NBI, NBo and JvD drafted the statistical analysis plan. SM and AH provided cultural guidance and support. JvD drafted the initial protocol and manuscript and all authors contributed to refinement of the study protocol and approved the final manuscript. The sponsor and funders had no role in the design of this study and will not have any role during its execution, analyses, interpretation of the data or decision to submit results.

Funding The study is cofunded by Cure Kids and A Better Start - National Science Challenge (Reference Number 7017) for 2 years. This covers investigator salaries and caregiver cofacilitators, website development, equipment, travel and training. The design, management, analysis and reporting of the study are entirely independent of study funders.

Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Not applicable.

Provenance and peer review Not commissioned; externally peer reviewed.

ORCID iD
Jenna van Deurs http://orcid.org/0000-0002-6862-2223

REFERENCES

1 Opai K. A time and space for Takiwātanga. *Altogether Autism Journal* 2017;2:13. Available: https://issuu.com/lifeunlimited1/docs/altogether_autism_journal__issue_2
2 Ministry of Health. Annual data explorer 2020/21: New Zealand health survey. 2021. Available: https://minhealthnz.shinyapps.io/nz-health-survey-2020-21-annual-data-explorer/_w_322ca116/#!/explore-indicators

3 Maenner MJ, Shaw KA, Bakian AV, *et al*. Prevalence and characteristics of autism spectrum disorder among children aged 8 years—autism and developmental disabilities monitoring network, 11 sites, United States, 2018. *MMWR Surveill Summ* 2021;70:1–16.
4 Eggleston MJF, Thabrew H, Frampton CMA, *et al*. Obtaining an autism spectrum disorder diagnosis and supports: New Zealand parents' experiences. *Research in Autism Spectrum Disorders* 2019;62:18–25.
5 Meer L, Evans K. *The autism diagnostic process in New Zealand: Final Report*. Autism CRC, 2021. Available: https://www.autismcrc.com.au/sites/default/files/reports/1-058_NZ-Diagnostic-Practices_Final-Report_2021.pdf
6 Whitehouse AJO, Varcin KJ, Alvares GA, *et al*. Pre-Emptive intervention versus treatment as usual for infants showing early behavioural risk signs of autism spectrum disorder: a single-blind, randomised controlled trial. *The Lancet Child & Adolescent Health* 2019;3:605–15.
7 Zwaigenbaum L, Bauman ML, Stone WL, *et al*. Early identification of autism spectrum disorder: recommendations for practice and research. *Pediatrics* 2015;136 Suppl 1(Suppl 1):S10–40.
8 Schreibman L, Dawson G, Stahmer AC, *et al*. Naturalistic developmental behavioral interventions: empirically validated treatments for autism spectrum disorder. *J Autism Dev Disord* 2015;45:2411–28.
9 Kasari C, Gulsrud AC, Wong C, *et al*. Randomized controlled Caregiver mediated joint engagement intervention for toddlers with autism. *J Autism Dev Disord* 2010;40:1045–56.
10 Crank JE, Sandbank M, Dunham K, *et al*. Understanding the effects of naturalistic developmental behavioral interventions: A project AIM meta-analysis. *Autism Res* 2021;14:817–34.
11 Tiede G, Walton KM. Meta-analysis of naturalistic developmental behavioral interventions for young children with autism spectrum disorder. *Autism* 2019;23:2080–95.
12 Rosenbrock GJ, Jellinek ER, Truong DM, *et al*. Treatment acceptability in parent-mediated interventions: considerations for maximizing outcomes for children with autism. *Practice Innovations* 2021;6:236–50.
13 Ooi KL, Ong YS, Jacob SA, *et al*. A meta-synthesis on parenting a child with autism. *Neuropsychiatr Dis Treat* 2016;12:745–62.
14 Barroso NE, Mendez L, Graziano PA, *et al*. Parenting stress through the lens of different clinical groups: A systematic review & meta-analysis. *J Abnorm Child Psychol* 2018;46:449–61.
15 Pinquart M. Featured article: depressive symptoms in parents of children with chronic health conditions: A meta-analysis. *J Pediatr Psychol* 2019;44:139–49.
16 Dykens EM, Fisher MH, Taylor JL, *et al*. Reducing distress in mothers of children with autism and other disabilities: A randomized trial. *Pediatrics* 2014;134:e454–63.
17 Hayes SC, Luoma JB, Bond FW, *et al*. Acceptance and commitment therapy: model, processes and outcomes. *Behav Res Ther* 2006;44:1–25.
18 Byrne G, Ghráda ÁN, O'Mahony T, *et al*. A systematic review of the use of acceptance and commitment therapy in supporting parents. *Psychol Psychother* 2021;94 Suppl 2:378–407.
19 Juvin J, Sadeg S, Julien-Sweerts S, *et al*. A systematic review: acceptance and commitment therapy for the parents of children and adolescents with autism spectrum disorder. *J Autism Dev Disord* 2022;52:124–41.
20 Bodden DHM, Matthijssen D. A pilot study examining the effect of acceptance and commitment therapy as parent counseling. *J Child Fam Stud* 2021;30:978–88.
21 Kingi TK, Durie M, Elder H, *et al*. *Maea te toi ora: Māori health transformations*. Huia Publishers, 2017.
22 Bailey B, Arciuli J. Indigenous Australians with autism: A Scoping review. *Autism* 2020;24:1031–46.
23 Gone JP, Hartmann WE, Pomerville A, *et al*. The impact of historical trauma on health outcomes for indigenous populations in the USA and Canada: A systematic review. *American Psychologist* 2019;74:20–35.
24 Freeth M, Milne E, Sheppard E, *et al*. Autism across cultures: perspectives from Nonwestern cultures and implications for research. In: Volkmar FR, Paul R, Rogers SJ, et al., eds. *Handbook of autism and pervasive developmental disorders*. 4th edn. John Wiley, 2014: 997–1013.
25 Tupou J, Curtis S, Taare-Smith D, *et al*. Māori and autism: A Scoping review. *Autism* 2021;25:1844–58.
26 Ellison KS, Guidry J, Picou P, *et al*. Telehealth and autism prior to and in the age of COVID-19: A systematic and critical review of the last decade. *Clin Child Fam Psychol Rev* 2021;24:599–630.
27 Chan A-W, Tetzlaff JM, Altman DG, *et al*. SPIRIT 2013 statement: defining standard protocol items for clinical trials. *Ann Intern Med* 2013;158:200–7.
28 Moher D, Hopewell S, Schulz KF, *et al*. CONSORT 2010 explanation and elaboration: updated guidelines for reporting parallel group randomised trials. *BMJ* 2010;340:c869.
29 Juszczak E, Altman DG, Hopewell S, *et al*. Reporting of multi-arm parallel-group randomized trials: extension of the CONSORT 2010 statement. *JAMA* 2019;321:1610–20.
30 Barbaro J, Dissanayake C. Prospective identification of autism spectrum disorders in infancy and Toddlerhood using developmental surveillance: the social attention and communication study. *J Dev Behav Pediatr* 2010;31:376–85.
31 Lacey C, Huria T, Beckert L, *et al*. The Hui process: A framework to enhance the doctor-patient relationship with Māori. *N Z Med J* 2011;124:72–8.
32 Barbaro J, Dissanayake C. Early markers of autism spectrum disorders in infants and toddlers prospectively identified in the social attention and communication study. *Autism* 2013;17:64–86.
33 Barbaro J, Ulusoy E, Dissanayake C. Determining the efficacy of the social attention and communication study in identifying Preschoolers with Asds: development of the SACS-preschool [paper]. 12th Annual Meeting of the International Society for Autism Research; Donostia/San Sebastian: Spain, 2013
34 Barbaro J, Sadka N, Gilbert M, *et al*. Diagnostic accuracy of the social attention and communication surveillance-revised with preschool tool for early autism detection in very young children. *JAMA Netw Open* 2022;5:e2146415.
35 Bakeman R, Adamson LB. Coordinating attention to people and objects in mother-infant and peer-infant interaction. *Child Dev* 1984;55:1278–89.
36 Nordahl-Hansen A, Fletcher-Watson S, McConachie H, *et al*. Relations between specific and global outcome measures in a social-communication intervention for children with autism spectrum disorder. *Research in Autism Spectrum Disorders* 2016;29–30:19–29.
37 Abidin RR. *Parenting stress index*4th edn. PAR, 2012.
38 Zaidman-Zait A, Mirenda P, Zumbo BD, *et al*. An item response theory analysis of the parenting stress index-short form with parents of children with autism spectrum disorders. *J Child Psychol Psychiatry* 2010;51:1269–77.
39 Achenbach TM. *Child Behavior Checklist for Ages 1.5-5*. ASEBA, 2001.
40 Achenbach TM, Rescorla LA. *Manual for the ASEBA preschool forms & profiles*. University of Vermont, Research Center for Children, Youth, and Families, 2000.
41 Sparrow SS, Cicchetti DV, Saulnier CA. *Vineland Adaptive Behavior Scales*Third Edition (Vineland-3). Pearson, 2016.
42 Miller JF, Gillon GT, Westerveld MF. *Systematic Analysis of Language Transcripts, New Zealand/Australia (SALT-NZAU 16) [Computer software]*. Madison, WI: SALT Software LLC, 2015.
43 Henry JD, Crawford JR. The Short-Form version of the depression anxiety stress scales (DASS-21): construct validity and normative data in a large Non-Clinical sample. *Br J Clin Psychol* 2005;44:227–39.
44 Cheron DM, Ehrenreich JT, Pincus DB. Assessment of parental experiential avoidance in a clinical sample of children with anxiety disorders. *Child Psychiatry Hum Dev* 2009;40:383–403.
45 Emerson LM, Aktar E, de Bruin E, *et al*. Mindful parenting in secondary child mental health: key parenting predictors of treatment effects. *Mindfulness* 2021;12:532–42.
46 Johnston C, Mash EJ. A measure of parenting satisfaction and efficacy. *Journal of Clinical Child Psychology* 1989;18:167–75.
47 Reimers TM, Wacker DP, Cooper LJ, *et al*. Acceptability of behavioral treatments for children: analog and naturalistic evaluations by parents. *School Psych Rev* 1992;21:628–43.
48 Reimers TM, Wacker DP, Cooper LJ, *et al*. Clinical evaluation of the variables associated with treatment acceptability and their relation to compliance. *Behavioral Disorders* 1992;18:67–76.
49 Sajobi TT, Fick GH, Lix LM. On statistical significance of discriminant function coefficients. *J Mod Appl Stat Methods* 2020;18:1–25.
50 Blampied NM. Analyzing therapeutic change using modified Brinley plots: history, construction, and interpretation. *Behav Ther* 2017;48:115–27.
51 Lakens D. Calculating and reporting effect sizes to facilitate cumulative science: a practical Primer for T-tests and Anovas. *Front Psychol* 2013;4:863:863.:.
52 Jacobson NS, Truax P. Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. *J Consult Clin Psychol* 1991;59:12–9.