

Mapping and Evaluating Services for Children with Learning Disabilities and Behaviours that Challenge (MELD): Stage 2

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SIGNATURE PAGE

The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the study in compliance with the approved protocol and will adhere to the principles outlined in the relevant study regulations, GCP guidelines, and CTU SOPs. I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the intervention without the prior written consent of the Sponsor.

I also confirm that I will make the findings of the study publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the study will be given; and that any discrepancies from the study as planned in this protocol will be explained.

Chief Investigator:		
Richard Hastings		26 May 2022
Name	Signature	Date

General Information This protocol describes Stage 2 of the MELD study, and provides information about the procedures for the study. Every care has been taken in drafting this protocol. However, corrections or amendments may be necessary. These will be circulated to the known Investigators in the study. Problems relating to the study should be referred, in the first instance, to the CI.

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This protocol has been developed by the MELD Study Management Group (SMG).

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Glossary of abbreviations

AE	Adverse Event
ASD	Autism Spectrum Disorder
BtC	Behaviours that Challenge
CAMHS	Child and Adolescent Mental Health Service
CI	Chief Investigator
DAG	Directed Acyclic Graph
FCAG	Family Carer Advisory Group
GCP	Good Clinical Practice
GDPR	General Data Protection Regulation
HS&DR	(NIHR) Health Services and Delivery Research
iDMEC	independent Data Monitoring and Ethics Committee
ISRCTN	International Standard Randomised Controlled Study Number
LD	Learning Disability
NHS	National Health Service
NIHR	National Institute for Health Research
PAG	Professionals Advisory Group
PPI	Patient and Public Involvement
R&D	Research and Development
RA	Research Assistant
REC	Research Ethics Committee
SAE	Serious Adverse Event
SAP	Statistical Analysis Plan
SMF	Study Master File
SMG	Study Management Group
SSC	Study Steering Committee
VABS3	Vineland Adaptive Behaviour Scales 3 rd Edition
WP	Work Package

1 Amendment History

The following amendments and/or administrative changes have been made to this protocol since the implementation of the first approved version.

Amendment No. <i>(specify substantial/non- substantial)</i>	Protocol version no.	Date issued	Summary of changes made since previous version
1. Non- substantial	v1.1	14.03.2022	The Sponsor reference number was added in. Changes were made to the number of service models found in Stage 1, services and participants to be sampled, as well as the sampling method. Dr Liz Schroeder was added as the study Health Economics Researcher.
2. Non- substantial	V2.2	25.05.2022	The IRAS reference number was added in. Correction made to the number of participants needed (from n=234 to n=244) throughout the protocol.

2 Synopsis

Title	Mapping and Evaluating Services for Children with Learning Disabilities and Behaviours that Challenge
Acronym	MELD
Funder and ref.	NIHR 129577
Study design	<p>a) An observational study to evaluate outcomes across service models (as identified in an earlier Stage 1 of the research) for children with learning disabilities and behaviours that challenge at service referral (or re-referral) and 12-months post-referral.</p> <p>b) A multiple case study design of children with learning disabilities and behaviours that challenge, their family carers, service staff, and a commissioner i) in one service within each service model type, and ii) in services using co-production.</p>
Study participants	<p>Family carers of for children with a learning disability and behaviours that challenge (observational study and case studies).</p> <p>Staff in community services for children with a learning disability and behaviours that challenge (observational study and case studies).</p> <p>Children with learning disabilities and behaviours that challenge (case studies).</p> <p>Health or social care/education staff who receive training or advice, or are involved in the individual cases but who are not a part of the service (observational study and case studies).</p> <p>Commissioners of community services for children with a learning disability and behaviours that challenge (case study 2.2a only).</p> <p>Staff (internal or external) who are involved in co-production in services (case study 2.2b only).</p>
Planned sample size	<p>Observational study: 244 children with learning disabilities and behaviours that challenge, and a family carer for each child. Up to 244 health or social care/education staff who are involved in the individual cases but who are not a part of the service. Each service will also complete</p>

	<p>a service-level data collection proforma about key elements of their service (including: reach, mean time to treatment, mean time to discharge, take-up of services, costs).</p> <p>Case studies (a): 6-7 children with learning disabilities and behaviours that challenge (including at least one child with more severe learning disabilities), 4-5 family carers, 2-3 service staff, 2-3 staff external to the service but involved in the cases, and 1 key commissioner for one service within each service model type.</p> <p>Case Studies (b): With input from our family carer and professionals advisory groups, as well as our SMG, we will also select up to 4 case studies to explore the co-production of services with children and/or family members. This would include interviewing 4-5 children with learning disabilities and behaviours that challenge, 4-5 family carers, 4-5 staff (internal or external) working with co-production, and 2-3 staff who are external to the service but are involved in the cases.</p>
Inclusion criteria	<p>The inclusion criteria for services are:</p> <ol style="list-style-type: none"> 1. They were a service included in the analysis for Stage 1, and 2. Nothing significant has changed in their service model since the Stage 1 analysis <p>The inclusion criteria for children referred/referred to services included in Stage 2 are:</p> <ol style="list-style-type: none"> 1. Child (0-15 years of age) has learning disabilities as defined administratively or otherwise by the service 2. Child has been referred at least in part for support in relation to behaviours that challenge (including those referred to the relevant learning disability/behaviours that challenge pathway in any broader service) 3. Child's parental caregiver consents to take part in the research 4. Child's parental caregiver is able to complete, by questionnaire or interview with a researcher, study outcome measures in English

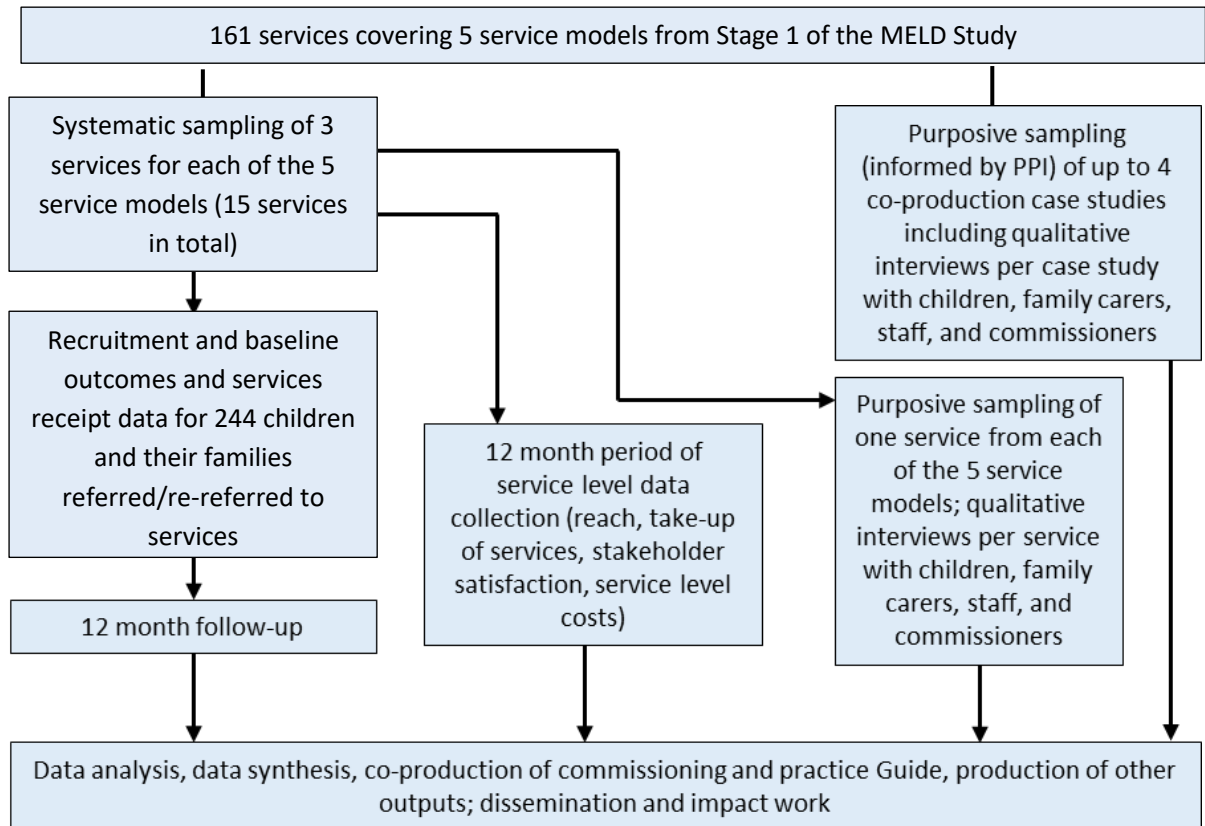
	<p>For the case studies:</p> <ol style="list-style-type: none"> 1. WP2.2a only: Researchers will only contact parental caregivers about interviews with them and/or their children where initial consent to be contacted about interviews was obtained at baseline 2. WP2.2b only: Researchers will only contact parental caregivers about interviews with them and/or their children if they agreed for their contact details to be shared by the service with the research team 3. Children and young people who will be interviewed will be between 6 and 15 years of age. Consent from their parental caregiver will be obtained before they take part, and their assent noted 4. Professionals (service staff, external staff, and commissioners) will be identified by the services, and researchers will only contact them if they agreed for their contact details to be shared by the service with the research team
Exclusion criteria	<p>The exclusion criteria for services are:</p> <ol style="list-style-type: none"> 1. Services that have been established for less than one year, and so have had little chance to become reasonably stable within the current study timeframe 2. Services that have a typical referral/re-referral rate over 6 months of fewer than 10 children 3. Services that indicate at Stage 1 they would not wish to be contacted about involvement in Stage 2 4. Co-applicants Lovell and Liew's services – to address potential conflicts of interest
Planned study period	22 months between 01 March 2022 and 31 December 2023
Primary objective	To evaluate outcomes for children and their families, and costs for service models of community services for children with learning disabilities and behaviours that challenge in England.
Secondary objectives	To understand the experiences of children and families, and the processes by which child LD and BtC services impact on the lives of children with LD and BtC, and their families and carers.

	<p>To describe the ways that services for children with LD and BtC co-produce services with families and carers and children and young people.</p> <p>To inform and support decision-making on commissioning and developing services for children with LD and BtC and their families and carers.</p>
Methodology summary	<p>WP2.1. Five service models were identified in Stage 1, we will recruit 15 services representing these models and 244 children and carers to take part in an observational study. Outcome measures assessing child behaviours that challenge, child mental health, child health, child quality of life, and adaptive skills, family carer wellbeing, family carer quality of life, services received, and (follow-up only) family carer experience and satisfaction will be completed by family carers at baseline (referral or re-referral to the service) and 12-month follow-up. Each service will also complete a service-level data collection proforma about key elements of their service (including: reach, mean time to treatment, mean time to discharge, take-up of services, costs). We will ask external stakeholders (e.g., social worker, education professional, referrer) to complete a satisfaction with the service measure about one child who is being supported by the service who they are involved with in a professional capacity. Statistical analyses will examine 12-month outcomes across service models, accounting for baseline scores and potential confounders or other key factors (informed by stakeholder consultation). Health economic analyses will examine the costs and cost effectiveness of each service model.</p> <p>WP2.2a. Using a multiple case study design approach, we will select one service of each service model type within which to gather data through a review of any relevant documentation and interviews (per case study) with 6-7 children with learning disabilities, 4-5 family carers, 2-3 service staff, 2-3 external staff who are involved in the cases, and a key commissioner.</p> <p>WP2.2b. With input from the family carer advisory group, professionals advisory group, and SMG members, we will also select up to four case studies</p>

to examine coproduction of services with children and/or family carers; again reviewing any relevant documentation and interviewing 4-5 children, 4-5 family carers, 4-5 staff (internal or external) working with co-production, and 2-3 external staff who are involved in the cases. Cross-case comparison and synthesis with documentary evidence about each service/coproduction model (and synthesis with quantitative data about each service from Stage 1) will be used to examine experiences, perceptions, and processes.

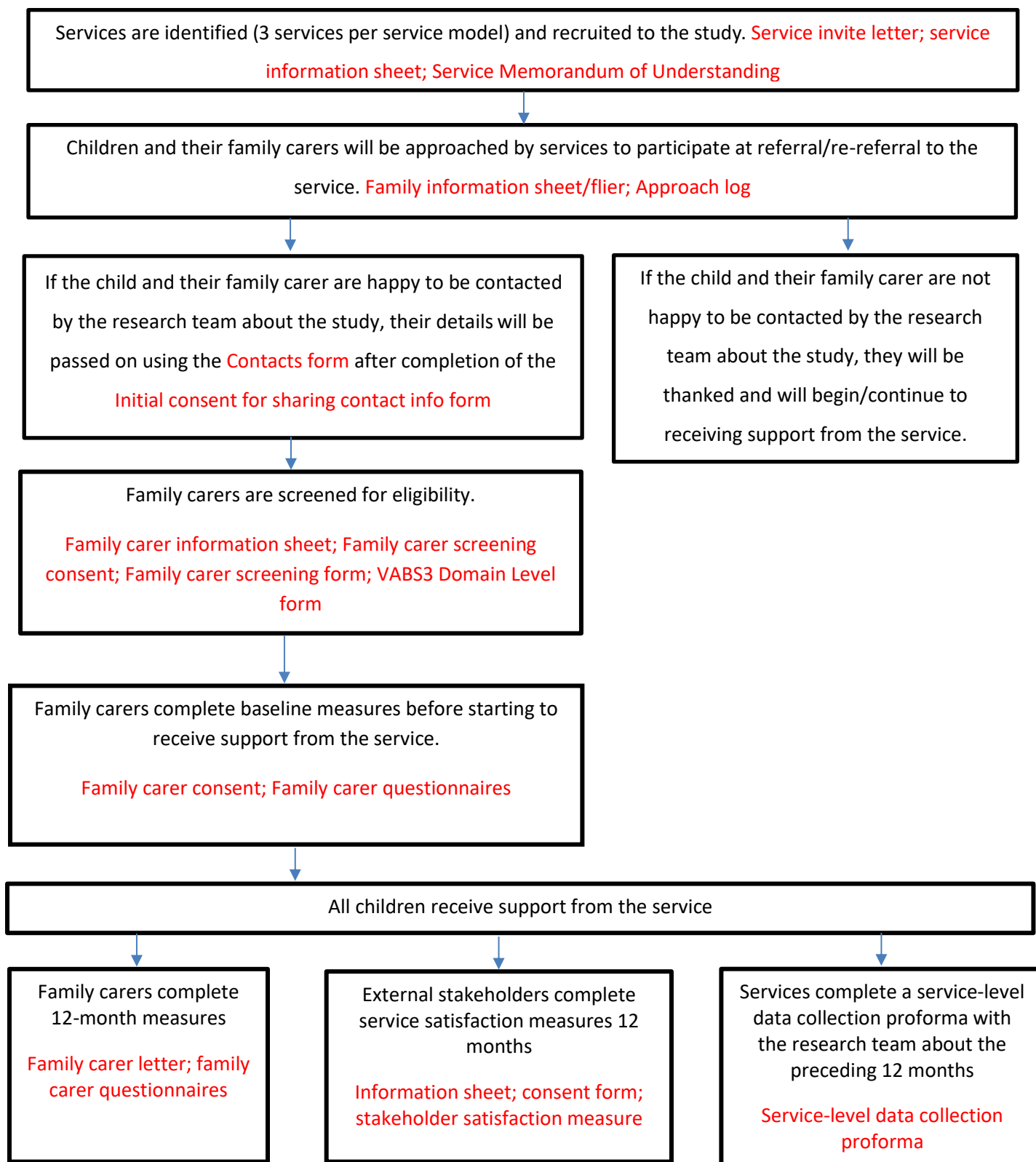
3 Study summary & schema

3.1 Study flow diagram



3.2 Participant flow

Work Package 2.1



Work Package 2.2a

Purposive sampling of one service case study including qualitative interviews per service model with children, family carers, and staff

Service case study invite letter; service case study information sheet; Service case study consent form; Service approach log



A case study, using qualitative methods will be completed about one service per service model:

Participant approach logs

Indicative topic guides for all interviews

6-7 children Child invitation letter for family carer; child information sheet; Parental consent form

4-5 family carers Family carer invitation letter; Family carer information sheet; Family carer consent form

2-3 Health/social care service staff who are not part of the service, but are involved in the case and 2-3 service staff Staff invitation letter; Staff information sheet; Staff consent form

1 Commissioner Commissioner invitation letter; Commissioner information sheet; Commissioner consent form



A review of any relevant documents (e.g., policies) related to how the service operates and delivers support to children with LD and BtC

Work Package 2.2b

Purposive sampling (informed by FCAG, PAG, and SMG) of up to 4 co-production case studies including qualitative interviews per case study with children, family carers, and staff

Service case study invite letter; service case study information sheet; Service case study consent form; Service approach log



A case study, using qualitative methods will be completed about 4-5 children in one service per co-production case study:

Participant approach logs

Indicative topic guides for all interviews

4-5 children Child invitation letter for family carer; child information sheet; Parental consent form

4-5 family carers Family carer invitation letter; Family carer information sheet; Family carer consent form

2-3 Health/social care service staff who are not part of the service, but are involved in the case and 2-3 staff working with co-production (staff may be internal or external to the service) Staff invitation letter; Staff information sheet; Staff consent form



A review of any relevant documents (e.g., policies) related to how the service operates and delivers support to children with LD and BtC



Services complete a service-level data collection proforma about co-production activities within the service with the research team

Service-level data collection co-production proforma

3.3 Study lay summary

Aims

This research is about community-based services across England that support children with a learning disabilities and behaviours that challenge, and their families. We want to find out if the way that services are structured and organised (known as “service models”) has any effect on the outcomes of children with learning disabilities and behaviours that challenge and their families. We also want to understand the experiences of children and families, and the ways that services for children with learning disabilities and behaviours that challenge work with families, carers, children and young people to develop services with them.

Background

1 in every 5 children with a learning disability in the UK display behaviours that challenge. These are not a medical diagnosis, but are behaviours (like aggression or self-injury) that may cause harm to the child or other people or prevent the child being included in the community. Children with learning disabilities and behaviours that challenge are at risk of negative outcomes (like abusive care), their families are more likely to experience stress, and these children’s care is costly for services. When the National Institute of Health and Care Excellence (NICE) reviewed the evidence, they found little research about how best to design and deliver health and care services to children with learning disabilities and behaviours that challenge. This study aims to fill this gap by evaluating the services currently offered to these children.

Design/methods

We will select 15 services (from 5 different service models that we identified in Stage 1 of the MELD project) to study in detail. We will ask families of 244 children newly referred to these services to complete questionnaires when they first come to the service and again after 12 months. We will gather detailed information about each service (like how many children they see in the year, and estimate costs), and we will ask people who are involved in the children’s care (but not employed by the service) how satisfied they are with the service. We will also interview children (using special communication techniques), families, and staff in each service model we identify about their experiences receiving and delivering care, as well as reviewing any relevant documents. Family carers and professionals in our advisory groups, as well as members of the study team, will help us to select from all Stage 1 services or their own networks four examples of co-production; where services work with children with a learning disability and/or families to design or improve services together. We will carry out additional interviews with children, family carers, and staff to find out how co-production is working, how much it costs, and we will review any relevant documents (e.g., policies, if any).

Patient/public involvement

Family carers have been working with us for 5+ years to tell us how services can be better for children with learning disabilities and behaviours that challenge. Their ideas have sparked this research. We will work with an advisory group of families throughout the research to make key decisions, drawing on their expertise, and working together to think about what the findings mean.

4 Background and rationale for the current study

Learning disability (LD), used as the official term in the UK health system, is known as Intellectual Disability internationally. Intellectual Disability/LD is a condition described in ICD-11 as a Disorder of Intellectual Development (Salvador-Carulla et al., 2011). Consistent with contemporary definitions of this condition, LD emerges during the “developmental period” (usually taken to mean before age 18 years), and is characterised by low cognitive ability (using standardised tools an IQ <70) and low levels of adaptive behaviour (such as communication, social skills, independence skills - also assessed using standardised tools). Prevalence studies internationally suggest that approximately 2% of children and adolescents have a LD (Maulik et al., 2011). UK Learning Disability Observatory data also show just over 2% of children in England have been identified by local authorities/schools as having LD (Hatton et al., 2014). Prevalence varies slightly with socio-economic factors but is broadly similar across the UK. In practice, and this is also reflected in the ICD-11 “sub-types” of intellectual disability, it is helpful to distinguish between levels of LD severity: mild (2-3 SDs below the mean on standardised IQ/adaptive behaviour assessments), moderate (3-4 SDs below the mean), and severe/profound (4 or more SDs below the mean). In addition, LD is associated with significantly higher prevalence of other neurodevelopmental conditions; in particular Autism Spectrum Disorder (ASD). The prevalence of LD among children and adolescents with ASD in UK population-based data has been shown to be as high as 52% (95% CI: 42%, 62%) (Totsika et al., 2011).

Children with LD are also likely to display challenging behaviour (or Behaviours that Challenge; BtC). Approximately 1 in 5 children with LD in the UK in contact with services display BtC (Emerson et al., 2001). Recent analysis of UK population data suggest 10-17% of children with LD show aggression towards others (Emerson et al., 2014). In some settings, prevalence rates are higher (e.g., 53% of children in a special school context; Nicholls et al., 2020). BtC are associated with poor care outcomes for children (e.g., increased exposure to restrictive care), for family carers (e.g., increased stress and mental health problems; Hastings, 2016), and increased costs of care to families (Einfeld et al., 2010) and to health and social care services (Iemmi et al., 2016). Children with more severe LD, and those with LD who also have autism, are more likely to display BtC (Nicholls et al., 2020).

BtC are understood theoretically from a contextual perspective in terms of definition, vulnerability factors, and maintaining processes (Bowring et al., 2019; Hastings et al., 2013). First, BtC are a socially defined health and social care issue, rather than a medical disorder or diagnosis; defined as behaviours that are not typical for the culture the person lives in and that occur at a frequency, severity, or duration that places an individual at risk of harm, places carers or others at risk of harm, or that hinder inclusion in typical community settings (Emerson & Einfeld, 2011). BtC are defined in terms of their effects rather than topography. Nevertheless, individuals with LD often engage in a number of behaviours that are typically considered challenging, regardless of context: injuring themselves (e.g., banging their heads against hard surfaces, eye-poking, skin scratching leading to bleeding), physical aggression towards others (e.g., kicking, biting, pulling hair), physically destructive behaviours (e.g., throwing furniture, pulling down curtains), and other actions (e.g., absconding, high rate unusual repetitive behaviours such as body rocking, inappropriate touching, screaming). The second contextual dimension is that the vulnerability factors for BtC are primarily (though not exclusively) psycho-social, relating to the inequalities and life experiences of people with LD (e.g., impoverished social networks, lack of communication skills, exposure to negative life events including abusive care, barriers to accessing health and care services). The third contextual dimension is that BtC are functional for the person engaging in them – they allow a certain amount of control over the (social) environment: the behaviour/response of others is then the main mechanism through which BtC are maintained and may worsen over time.

Given the prevalence of BtC, and continued high profile care scandals (e.g., BBC Panorama exposés of Winterbourne View in 2011, and Whorlton Hall in 2019), effective community-based services and supports are a national priority (NHS England, 2015). However, NICE guidelines for BtC (2015, 2018) found no high quality evidence relating to the design and organisation of services for children. This study focuses on that evidence gap.

There are currently no data on an England-wide basis about the outcomes and costs associated with different models or characteristics of services for children with LD and BtC. Given the lack of evidence overall, the findings from the current research will be directly relevant to the ongoing planning and delivery of health and social care services across the UK.

5 Study objectives and outcome measures

5.1 Study objectives

In the proposed research, we will measure outcomes and costs for identified service models (see MELD Stage 1 protocol) by following children and families referred to selected services.

The *research objectives* are to:

1. To assess the outcomes for children with LD and BtC and their carers for different service models.
2. To estimate the economic costs of different service models for children with LD and BtC, and to assess their economic outcomes within a cost-consequences framework.
3. To understand the experiences of children and families, and the processes by which child LD and BtC services impact on the lives of children with LD and BtC, and their families and carers.
4. To describe the ways that services for children with LD and BtC co-produce services with families and carers and children and young people.
5. To inform and support decision-making on commissioning and developing services for children with LD and BtC and their families and carers.

5.2 Study outcome measures

The following outcomes will be measured at baseline and at 12-months post-referral as a part of the observational study, unless otherwise stated:

- Child-related outcomes (all completed by family carers)
 - Child BtC measure
 - Behaviour Problems Inventory Short Form (BPI-S) (Rojahn et al., 2012)
 - Child physical health measures
 - Children's sleep habit questionnaire (CSHQ) (Owens et al., 2000)
 - Non-communicating children's pain checklist – revised (NCCPC-R) (Breau et al., 2004)
 - Child mental health measure
 - Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997)
 - Child quality of life measures
 - EQ-5D-Y Health Questionnaire (Wille et al., 2011)

- **EITHER** Paediatric Quality of Life Inventory (PedsQL) infant scales (*only for children aged 0-24 months*) (Varni et al., 2011) **OR**
 - Paediatric Quality of Life Inventory (PedsQL) Generic Core Scales (*only for children aged 2 years+*) (Varni et al., 2001)
- Child skills measure
 - GO4KIDDS (Perry et al., 2015)
- Child services received measure
 - Client service receipt inventory (CSRI) (Beecham & Knapp, 1992)
- Family-carer-related outcomes (all completed by family carers)
 - Family carer quality of life measure
 - EQ-5D-5L Health Questionnaire (van Hout et al., 2012)
 - Family carer wellbeing measure
 - Warwick Edinburgh Mental Wellbeing Scale (WEMWBS) (Tennant et al., 2006)
 - Family carer services received measure
 - Client service receipt inventory (CSRI) (Beecham & Knapp, 1992)
 - Family carer experience/satisfaction with service (at 12-months only)
 - Experience of service questionnaire (ESQ) satisfaction with care items (Brown et al., 2014)
- Service-level outcomes (at 12-months only) (completed by services)
 - 'Reach' to population
 - Timings of service delivery (e.g., mean time to starting support from referral, and length of time to discharge)
 - Service take-up
 - Costs at service level
- Other-stakeholder-related outcomes (at 12-months only) (completed by external stakeholders)
 - Satisfaction of external stakeholders (e.g., referrer, health, social-care, or education professionals) brief measure (*Developed for the purposes of this study*)

6 Study design and data collection methods

The study involves two main work packages: a quantitative observational study (Work Package 2.1) and qualitative case studies (Work Package 2.2). In Work Package 2.1, we will use a systematic sampling of service models identified in Stage 1 of the MELD project and monitoring of outcomes for children and their families referred to the selected services and followed up for 12 months. Alongside this quantitative research, we will use qualitative interviews with stakeholder groups (Work Package 2.2). These will be described in more detail below.

Work Package 2.1 – Individual and services quantitative data, including economic data

Individual Case Level Inclusion and Outcome Data

A family carer will provide outcomes data in relation to each child/family recruited into the Stage 2 evaluation. All children referred/re-referred to services during the eight-month window for recruitment will be potentially eligible. The inclusion and exclusion criteria are described in sections 8.1 and 8.2.

The child's primary parental caregiver will complete the outcomes at baseline, and at a 12-month follow-up. The child's primary parental caregiver will also complete a version of the client services receipt inventory (CSRI) (Beecham & Knapp, 1992) at the same data points for themselves and their child with LD and BtC, adapted on the basis of lessons learnt from an ongoing economic analysis of the Early Positive Approaches to Support (E-PaTS) programme (NIHR PHR 15/126/11) for family caregivers of children with developmental delays.

We have chosen a 12-month follow-up to match with minimal follow-up expectations from RCTs, but also informed by data from service evaluations of specialist BtC services. In our three-year evaluation of one all-age service (39 children, 46 adults), the mean time between baseline and follow-up (at discharge) for the 85 cases included was 45 weeks (SD=29.19), median=37 weeks; range 15-160 weeks (Bowring et al., 2020). Eleven cases seen by a more intensive service for children with severe and complex BtC were in contact with the service for an average of 15 months (Dilks-Hopper et al., 2019).

To provide an opportunity for a longer-term follow-up, especially for health economic analysis, consent will be sought to follow-up families again, including collection of routine service use data. This longer-term follow-up is not included in the current protocol. Associated with socioeconomic

position, parents of children with LD may have limited literacy skills. Therefore, ability to read English is not part of our eligibility criteria. Measures will be completed with family carers by telephone or MS Teams interview if they choose. Following good practice from our recent NIHR and other funded research, during initial recruitment/consent literacy and other support needs that could relate to family carers' potential to participate in the study will be checked with them. Specifically, participants will be given an opportunity to indicate a preference to complete measures by interview in person with a member of the research team (at their own home or a mutually agreed location), via telephone, via MS Teams, or to be sent measures directly for self-completion (online or post). In recent research, 5-15% of family carers chose telephone completion and 8-9% chose face-to-face data collection. We have not asked why parents chose these options but providing the options as a standard practice means that parents do not have to reveal needs to researchers that they do not wish to (e.g., difficulty reading English).

At baseline, the child's primary parental caregiver will also be asked to complete a questionnaire about key family demographic information and child characteristics including "co-morbidities" such as autism diagnosis, epilepsy, and physical/motor impairments.

Assessment of Severity of Learning Disability

The Vineland Adaptive Behavior Scales (VABS) 3rd Edition Interview Form Domain Level assessment (Sparrow et al., 2016), will be carried out as a telephone/MS Teams interview with the primary parental caregiver as part of the eligibility check to determine severity of LD, although this would not form part of the exclusion criteria. If family carers have opted to complete outcomes in a face-to-face/telephone/virtual meeting with a researcher, the VABS will also be carried out at the same time rather than as a separate interview. The VABS is a standardised adaptive behaviour assessment, and the Domain Level version can be completed by a trained researcher in 10-15 minutes with a parental caregiver. Although formal diagnosis of LD in clinical settings should also include an IQ assessment, based on extensive research experience in the field such data are very unlikely to be available in children's clinical files and direct testing of the children by research staff is expensive and can be challenging for very young children and those with severe disabilities. Therefore, an adaptive behaviour assessment is the best option within the research to estimate LD severity. Children with a VABS composite score of <50 will be considered in the present study to have moderate to severe/profound LD. The VABS assessment is not a valid method to clearly identify further severity levels of LD as defined in ICD-11 (i.e., mild, moderate, severe/profound). However, the VABS composite score (as a continuous measure) or <50 vs. 50+ categorisation can be used in statistical

analyses, and to describe the obtained sample of children. Given the observational nature of the study design, the VABS data will not be used to determine eligibility for inclusion in the research, since services will have their own eligibility criteria for inclusion in their child LD BtC service or pathway.

Service-Level Outcomes.

Data about each of the services will be gathered for a 12-month period during the time they are active in the observational study. Data will be gathered for the service as a whole (not limited to those who consent to take part in individual case data collection) on:

- ‘Reach’ to population (proportion of the local population of children with LD and BtC who receive services – based on a national population estimate applied to local population statistics).
- Mean time to starting support or assessment from the point of referral/re-referral, and length of time to discharge.
- Take-up: proportion of referrals that go on to have some assessment and/or support.
- Costs at a service level (based primarily on service level information about staffing, other resources, accommodation etc.) over the 12-month period.

These data will be gathered in a visit to each service by a researcher who will work with the service manager/administrator, local finance leads, and clinicians to gather the relevant data using a standardised procedure. A top-down costing approach using primary accounting methods will be used to cost each service and will be based on an assessment of staff costs, employer on-costs, training costs, and revenue and capital overheads, delineated on the basis of service type by workload activity.

These data will be used in part to describe the services taking part in Stage 2, in part as potential variables in the main statistical analysis, and in part to enable health economic analysis.

WP2.2 Qualitative Data Collection – Design Summary

Utilising a multiple case study design, informed by Yin (2018) and Stake (2006), there will be two aspects to qualitative data collection.

A. Service Model case studies (WP2.2a). One service reflecting each of the 5 service models will be identified purposively (reflecting rural/urban mix, strength of other community services and any

other key factors identified in consultation with the family carer advisory group and professionals advisory group after Stage 1) to be the focus for qualitative data collection. If the first service selected declines to participate, another will be selected until an agreement is obtained. In each service, the following data will be gathered to explore views and experiences of the work the service carried out:

- Interviews with 6-7 children (6-15 years of age) with LD and BtC using a communication tool Talking Mats piloted already by our team (Bradshaw et al., 2018). We aim to collect data from at least one child per case study with more severe LD who is not able to use Talking Mats (details below)
- Interviews with 4-5 family carers
- Interviews with a 2-3 staff working within the service
- Interviews with 2-3 staff who are external to the service, but are involved in the cases (e.g., education, health, and social care professionals)
- Interview with the key commissioner of the service
- Review of any relevant written documentation (e.g., policy, if any).

In terms of interviews with 6-7 children with LD and BtC (6–15 years of age) for each case study, parents/guardians of children with LD and BtC will provide consent for them to take part in Talking Mats facilitated interviews or informal observations to assess the preferences of children with severe learning disability. Parents will also be asked about their child's communication preferences and any specific requirements in terms of dealing with the child's BtC - to aid the researchers to prepare for each child's interview or observation. Parents will also provide information about how their child may communicate discomfort or indicate lack of willingness to take part in an activity. Children with LD will be asked for their assent and research staff trained to understand non-verbal clues suggesting that the children may not be comfortable with the data collection procedure.

A Talking Mat is a framework for supporting communication. It involves a topic, a scale (positive, unsure, negative) and a series of options to place on the mat, all by using symbols. A series of Talking Mats will be prepared on a range of topics, including:

- Mats about 'my support' will enable us to ask children about what they like to do and how they feel about where they spend time and the people they are supported by/cared for. This will help us to gather data relevant to characteristics perceived as important and what people's experiences are.

- A mat about 'good for me' will enable children to tell us a little more about the positive outcomes (though probably with a focus on more concrete outcomes) they experience and from what we know about where they are living/services they receive, we can learn something about their experiences of different service models.

Placement of symbols by children on their Talking Mats will be recorded by taking a photograph of each mat. Each session will also be video recorded and a transcription of the session will be generated, capturing both verbal and non-verbal communication to understand children's reasoning behind the placement of symbols. These data will then be used in the analysis of the Talking Mats facilitated interviews with children.

We wish to be as inclusive as possible and aim to interview at least one child with severe LD per case study who is not able to use the Talking Mats method. This interview engagement will be exploratory, since established methods are not available. Co-investigators Bradshaw and Gore contributed to a Challenging Behaviour Foundation (our PPI partner)/Mencap guide about good practice in seeking the views of children with severe to profound LD (CBF/Mencap, 2017). Seeking the views of children with more severe LD needs careful tailoring but normally involves a series of steps to be taken with the parents/carers and the child.

Before the visit: A telephone interview is carried out with a family carer to ask questions about communication skills, support, activities, behaviour support and how the young person shows what they do and don't like. Based on this information the visit is tailored to the young person's communication needs (for example, sending a tailored information sheet to explain the visit, refreshing relevant Makaton signs, preparing symbols etc).

The aims of this session are to:

- identify communication preferences,
- identify how the young person shows what they do and don't like,
- agree proactive and positive strategies for reducing the likelihood of challenging behaviour within sessions, and
- agree reactive strategies to diffuse challenging behaviours and keep the child and others safe if these were to occur.

During the visit: Children are visited at their home. The person engaging with the child spends time being in their environment and ensuring the child feels comfortable in their presence and interacting with them. The following methods are used to gather the views and experiences of children.

1. **Direct engagement in activities:** Specific activities will vary depending on what the child enjoys and what is practical within the visit, examples include: playing with various sensory toys, puzzles, art and craft, reading a book, trampoline, and eating a meal. Activities are video recorded for analysis after the visit.
2. **Observation:** Informal observations of the child interacting with a family member or carer are video recorded. “Observable indicators” are used to identify what the child enjoys and engages with and what they don’t enjoy.

We will also interview 4-5 family carers, 2-3 staff working within the service, 2-3 health or social care/education staff who receive training or advice or are involved in the individual cases but who are not a part of the service, and a key commissioner of the service, as well as reviewing any relevant documentation (e.g., policies, if any). Purposive sampling will be utilised to ensure a diverse range of participants, including gender, length of time in contact with services/as part of the service, and for children/carers, the severity of LD as this was a dimension identified in PPI preparation that should be considered, the presence of an additional diagnosis of ASD, age of young person (in particular, to include some young people if possible in teen years closer to transition to adult services), and ethnicity.

Interviews (facilitated using Talking Mats) with children will be face-to-face or could be online should the need arise (see Section 20.5). Data collection with children with severe LD will be face-to-face, and separate protocols would need to be developed for virtual observations should the need arise (see Section 20.5). All other interviews will be face-to-face or on the telephone/videoconference (i.e., MS Teams), with the participant choosing the format they prefer and taking into account any government recommendations in force due to COVID-19.

For each case study there will be up to 19 interviews (7 of which with children with LD and BtC using Talking Mats, or observations, and 12 other semi-structured interviews). Thus, across 5 cases (services) there will be up to 35 Talking Mats interviews or observations and 60 semi-structured interviews. We will ensure fully informed consent/assent is obtained from each participant. Semi-structured interviews will be digitally audio recorded using encrypted recorders or within MS Teams,

whereby the recording will only be available to the interviewer, person transcribing the interview, and participant until it is deleted, and transcribed verbatim. The transcripts will then be checked carefully against the recording for accuracy and to ensure they are anonymised.

We plan to use our first case as a pilot study, and review whether amendments are needed for the subsequent case studies.

B. Co-production case studies (WP2.2b). Our family carer advisory group, professionals advisory group, and SMG members will review information about co-production in the 161 services in Stage 1 and will identify co-production examples from their own networks. These groups will work with co-investigator Seers to select up to four co-production case studies (for working with family carers, and also with children with LD if there are examples) for in-depth exploration (Yin, 2018). Data per case study will be collected by reviewing any relevant documentation (e.g., policies, if any) and interviewing:

- i. 4-5 children on their views of being involved in co-production,
- ii. 4-5 family carers,
- iii. 2-3 external health or social care/educational staff,
- iv. 4-5 staff working with co-production (staff may be internal or external to the service).

Views of facilitators and barriers to co-production will be explored. Semi-structured interviews will be used, and questions and prompts will be co-produced with relevant stakeholders. In each case study, detailed information allowing costing of the co-production activities will also be gathered using a structured proforma. This will include accurately measuring all time inputs for professionals and lay members involved in the co-production activities. For professionals, a record of the type and grade of staff involved in the co-production activities is required. For lay members, some description of their usual activities is required (e.g., employment role, and the type of activity that has been displaced by their involvement in the co-production activities). In addition, if there are face-to-face meetings, then the cost of venue hire and travel costs will need to be recorded, as well as any other types of resource inputs that are involved in the co-production process.

There will be up to four co-production case studies where parents/carers have co-produced the service model and/or its delivery.

We will purposively sample to include those services who have undertaken strategic coproduction involving children with LD and BtC, as well as those who have worked with carers of children with LD and BtC. For each case study, in addition to reviewing the service's documentation on coproduction, we will use purposive sampling (outlined above) to interview: i) 4-5 children on their views of being involved in the co-production (facilitated with Talking Mats), ii) 4-5 family carers, iii) 2-3 health or social care/educational staff, iv) 4-5 staff working with co-production (staff may be internal or external to the service). Thus, for the co-production case studies we will have up to 20 Talking Mat interviews with children, and up to 52 semi-structured interviews.

A series of Talking Mats will be prepared on a range of topics, including:

- Mats about 'being involved' will enable us to ask children about what they have been involved in within the service. This will help us to gather data relevant to whether children with learning disabilities have been involved in making choices about their own care.
- A mat about 'help to be involved' will enable children to tell us a little more about how happy they have been with the methods that have been used to help them to be involved in making choices about their own care.

As outlined above, Talking Mat interviews will be video recorded using either a video camera or MS Teams. Each mat will be photographed, and interviews will be transcribed. Semi-structured interviews will be digitally audio recorded using encrypted recorders or MS Teams and transcribed verbatim. The transcripts will then be checked carefully against the recording for accuracy and to ensure they are anonymised. In each case study, detailed information allowing costing of the co-production activities will also be gathered, as described above. The first case will be used as a pilot study and carefully reviewed to determine if any amendments are required.

6.1 Participant voucher payments

Family carer participants will receive a £10 high-street voucher for completion of the baseline questionnaire and £20 for completion of the 12-month follow-up questionnaire. The higher amount at follow-up is to support participant retention at follow-up.

All non-NHS staff participants (i.e., parent carers, children with LD, staff external to the service, commissioners) who take part in an interview will be given a £15 high street voucher for completion of an interview with a researcher.

6.2 Risk assessment

This is a low-risk study. The research is purely observational, and no interventions will be implemented outside of the treatment as usual within services. Involvement in the study is entirely voluntary, and informed consent will be obtained prior to any data collection with participants. Participants will be asked questions about their/their child's wellbeing, mental health, health, quality of life, and experience of the service using measures that are routinely used in research with children and young people with learning disabilities.

We will complete a formal risk assessment before the study commences.

7 Site and Investigator selection

Each service, and each co-production case study will be a separate site, and a PI will be identified within each site. Identification of services is described in detail in Section 8 below. Identification of Co-Production Case Studies will be undertaken with the involvement of the family carer advisory group, professionals advisory group, and the SMG.

The study will be carried out at University of Warwick, under the supervision of the Chief Investigator (Hastings). Fully trained Research Fellows and Research Assistants at the University of Warwick will be responsible for recruitment and all data collection. A Site Delegation Log and Roles and Responsibilities document will be completed and full contact details will be recorded for all Sites. A Site File, containing all relevant study documents will be prepared at the University of Warwick.

Once all study documentation is in place and a Site Initiation Visit has been completed, recruitment of services and participants into the study will begin.

Occasionally during the course of the study, amendments may be made to the study documentation, required approvals obtained, and the latest approved versions will be added to the Site File.

8 Selection of Services and Participants

All eligible services from Stage 1 of the MELD study will be considered (N=161), and sampling of services will be according to a systematic sampling process to select 15 Sites from the approximately 161 services included in Stage 1 (3 services per Class).

Family carers of children with LD and BtC will be approached by services upon referral/re-referral to the service and provided with an information sheet. They will be asked whether they agree for their contact details to be shared with the research team, who will contact them with more information about the study, and to undergo an eligibility check with the research team. Family carers of children with LD and BtC are eligible for the study if their child meets all of the following inclusion criteria and none of the exclusion criteria apply. All queries about participant eligibility should be directed to the Study Manager before recruitment.

Service staff who are supporting children with LD and BtC in the service will be approached by the Site to participate in an interview for one of the case studies (either WP2.2a or possibly WP2.2b). There are no formal inclusion/exclusion criteria for these participants, provided they are working within the service to generally support children with LD and BtC.

Services will be asked to identify someone within the service who can work with a member of the research team to complete the service-level data proforma (described in section 5.2). There are no formal inclusion/exclusion criteria for these participants.

Services will also be asked to identify external stakeholders who are involved in the care of at least one child who is being supported by the service (e.g., education staff, social worker, referrer to the service) who could complete a brief service satisfaction measure at 12-months for one of these children. There are no formal inclusion/exclusion criteria for these participants, provided they are supporting children who are being supported by the service.

External staff who are involved in cases and commissioners (WP2.2a only) will be identified by the Site to be approached to participate in an interview for one of the case studies (WP2.2a, WP2.2b). There are no formal inclusion/exclusion criteria for these participants.

In WP2.2b only, and only if the Site selected was not involved in WP2.1 at all, Sites will be asked to identify family carers and children with LD and BtC to be approached about participating in interviews about their experiences of co-production. If the Site was involved in WP2.1 then only family carer participants who provided initial consent to be contact about an interview for them and/or their child will be contacted by the research team.

8.1 Inclusion criteria

The inclusion criteria for services are:

1. They were a service included in the analysis for Stage 1, and
2. Nothing significant has changed in their service model since the Stage 1 analysis

The inclusion criteria for children referred/referred to services included in Stage 2 are:

1. Child (0-15 years of age) has learning disabilities as defined administratively or otherwise by the service
2. Child has been referred at least in part for support in relation to behaviours that challenge (including those referred to the relevant learning disability/behaviours that challenge pathway in any broader service)
3. Child's parental caregiver consents to take part in the research
4. Child's parental caregiver is able to complete, by questionnaire or interview with a researcher, study outcome measures in English

8.2 Exclusion criteria

The exclusion criteria for services are:

1. Services that have been established for less than one year, and so have had little chance to become reasonably stable within the current study timeframe
2. Services that have a typical referral/re-referral rate over 6 months of fewer than 10 children
3. Services that indicate at Stage 1 they would not wish to be contacted about involvement in Stage 2
4. Lovell and Liew's services – to address potential conflicts of interest

The only exclusion criteria for children referred/re-referred to services included in Stage 2 are not meeting the inclusion criteria.

There are no formal inclusion/exclusion criteria for the other groups of participants, apart from their involvement in the services, and the types of participants who will be approached are outlined in section 8 above.

9 Recruitment, Screening and registration

9.1 Identification of Services

Our main strategy for identification and recruitment of services for WP2.1 is described in Section 8 above, and will be based on the findings of Stage 1 of the MELD project.

For WP2.2a, we will approach a purposive sample of services who were involved in WP2.1, providing them with an information sheet, and will ask if they consent to be involved in the qualitative data collection element of the study (one service from each service model). More details are provided in Section 6.

For WP2.2b, we will approach a purposive sample of services who were involved in Stage 1 of the MELD Study, providing them with an information sheet, and will ask if they consent to be involved in the qualitative data collection element of the study (one service from each service model). More details are provided in Section 6. Other services may be approached that did not take part in Stage 1 of the MELD project if identified as relevant by our family carer advisors.

9.2 Identification of Participants

A. Quantitative data collection (WP2.1)

Our main strategy for identification and recruitment of participants is described in Section 8 above. Potential participants will agree to have their contact details shared with the research team, who will then make contact with potential participants to share further information about the study with them and determine their preferences for completing the questionnaires, and potentially taking part in interviews.

B. Qualitative Case Studies (WP2.2a)

We will identify 6-7 children, 4-5 family carers, 2-3 service staff, 2-3 external staff who are involved in the cases, and one commissioner to interview per service; these interviews will become a case study for that service. Methods for identifying service staff, external staff, and commissioners are outlined in section 8 above.

Family carers will be asked upon recruitment to the study if they would be happy to be contacted about them and their child being invited to interview at a later point in the study. Only family carers who consent to being contacted about interviews will receive an invitation for themselves and/or their child to take part in interviews following the observation study 12-month data collection.

C. Co-Production Case Studies (WP2.2b)

Additionally, the four services selected to be involved in WP2.2b will be asked to identify 4-5 children, 4-5 family carers, 2-3 external staff who are involved in the cases, and 4-5 staff (internal or external) who are involved in co-production. We will approach potential participants to be involved in an interview to explore their experience of co-production within the service. If the Site was involved in WP2.1 then only family carer participants who provided initial consent to be contact about an interview for them and/or their child will be contacted by the research team.

9.3 Approach logs

A. Quantitative data collection

The staff within services will keep a log of all participants considered/approached and whether they agreed to be contacted by the research team. When at Site, logs may contain identifiable information but this **must** be redacted prior to being sent to the research team at the University of Warwick. The approach log should be sent to the MELD Study team every month.

B. Qualitative Case Studies and Co-Production Case Studies

For the qualitative interviews, the research team will maintain an approach log of family carer and child participants. Services will keep an approach log for all other potential participants (i.e., internal staff, external staff, commissioners). For WP2.2b, it may be necessary for services to keep an approach log for family carers and children as well, if the service was not involved in WP2.1. When at Site, logs may contain identifiable information but this **must** be redacted prior to being sent to the research team at the University of Warwick. The approach log should be sent to the MELD Study team every month.

9.4 Recruitment rates

A. Quantitative data collection

15 services will be recruited at the beginning of this project.

A total of approximately 244 children will be recruited at an expected rate of approximately 29 per month during the 8-month recruitment period.

Up to 244 health or social care/education staff who are involved in the individual cases but who are not a part of the service will be recruited at the 12-month follow-up timepoint to complete a brief satisfaction measure.

B. Qualitative Case Studies

We will interview 6-7 children (6-15 years of age) with LD and BtC using Talking Mats (as described above). We aim to interview at least one child per case study with more severe LD who is not able to use Talking Mats. We will also interview 4-5 family carers, 2-3 staff working within the service 2-3 health or social care/education staff who receive training or advice, or are involved in the individual cases but who are not a part of the service, and a key commissioner of the service.

C. Co-Production Case Studies

We will interview 4-5 children (6-15 years of age) with LD and BtC using Talking Mats (as described above). We will also interview 4-5 family carers, 2-3 health or social care/education staff, and 4-5 staff working with co-production (staff may be internal or external to the service).

9.5 Recruitment/Informed consent

A. Quantitative data collection

Family carers:

- All participants will have been sent the Participant Information Sheet and consent form prior to the recruitment telephone call taking place and given sufficient time to read the information. The study will be explained in detail within the telephone call, with participants being encouraged to contact the study team if they have questions about the research. If face-to-face data collection, written consent will be obtained ahead of data collection commencing. If data will be collected over the telephone or MS Teams, verbal consent will be obtained. If data are collected online, an online consent form will be presented before any questionnaires. Verbal consent will also be obtained for postal completions of questionnaires. The Research Assistant will read aloud each statement of the consent form and ask the

participant to agree to each statement individually. The Research Assistant will then sign the consent form on behalf of the participant. A copy of the consent form will then be sent by post to the participant.

- A contacts form will be completed for family carer participants including multiple methods of contact (address, telephone, email address) to minimise loss to follow-up.
- Preferences for follow-up data collection for family carers (face-to-face interview completion, telephone/MS Teams completion, online, or postal questionnaires).
- Baseline data collection for family carers completed (either at time of recruitment by telephone/MS Teams or at a suitable time for the participant by telephone/MS Teams, face-to-face, online, or postal) including:
 - Baseline demographic questionnaire completed
 - Baseline outcome measures completed.

Service staff:

Service-level data collection

- The service-level data collection proforma will be completed.

Service costs data collection for co-production case studies

- The service-level data collection proforma will be completed.

External stakeholders:

- All external stakeholder participants will have been sent the Participant Information Sheet and consent form and given sufficient time to read the information. Postal consent will be obtained for postal completions of the measure, and online consent will be obtained for online completion of the measure.
- The satisfaction measure will be completed.

B. Qualitative Case Studies and Co-Production Case Studies

- All participants will have been sent a Participant Information Sheet and consent form prior to recruitment taking place and given sufficient time to read the information. If required, the

study will be explained in detail within a telephone call, with participants being encouraged to contact the study team if they have questions about the research.

If face-to-face data collection, written consent will be obtained ahead of data collection commencing. If data will be collected over the telephone or MS Teams, verbal consent will be obtained. The Research Assistant will read aloud each statement of the consent form and ask the participant to agree to each statement individually. The Research Assistant will then sign the consent form on behalf of the participant. A copy of the consent form will then be sent by post to the participant.

Parents/guardians of children with LD and BtC will provide consent for them to take part in Talking Mat facilitated interviews or informal observations (for children with severe LD). Parents will also be asked about their child's communication preferences and any specific requirements in terms of dealing with the child's BtC - to aid the researchers to prepare for each child's interview or informal observation (for children with severe LD). Parents will also provide information about how their child may communicate discomfort or indicate lack of willingness to take part in an activity. Children with LD will be asked for their assent and research staff trained to understand non-verbal clues suggesting that the children may not be comfortable with the data collection procedure.

- Interviews will be completed.

9.6 Registration

The MELD study will be registered with the ISRCTN database.

10 Withdrawal

10.1 Withdrawal

Participants have the right to withdraw consent for participation in any aspect of the study at any time until data analysis commences. The participants' care will not be affected at any time by declining to participate or withdrawing from the study, or from an individual aspect (e.g., a qualitative interview).

If a participant initially consents but subsequently withdraws from the study, or an individual aspect of it, clear distinction must be made as to what aspect of the study the participant is withdrawing from. These aspects could be:

1. Withdrawal from all future follow-up assessments

2. Withdrawal from qualitative interview
3. Withdrawal from previously collected data
 - a. Baseline assessments
 - b. Follow-up assessments
 - c. Qualitative interview
 - d. All of the above
4. Withdrawal of consent to all of the above

Participants who consent and subsequently withdraw should complete the study withdrawal form or the withdrawal form should be completed on the participant's behalf by the Research Assistant/ study team member based on information provided by the participant. This withdrawal form should be sent to the MELD Study email address. Any queries relating to potential withdrawal of a participant should be forwarded to the Study Manager.

10.2 Lost to follow up

Family carer participants who do not complete the 12-month follow-up data collection will be considered lost to follow-up. The research team will send reminders before they are considered lost to follow-up. The schedule of reminders will be as follows:

One week before sending the questionnaire: A courtesy text/email to let them know to expect it

One week after sending the questionnaire: A reminder email/text

Two weeks after sending the questionnaire: A reminder telephone call, speaking to them if possible or leaving a voicemail

Three weeks after sending the questionnaire: A postal survey (with a minimum dataset version of the questionnaire for family carer participants)

11 Study procedures

11.1 Baseline and follow-up assessments for WP2.1

Family carers:

Family carer participants will be approached about the study, and provided an information sheet by sites upon approach. If willing to be contacted by a member of the research team, their contact information will be shared with the research team and an eligibility call will be arranged, if eligible, a recruitment/baseline interview (either telephone interview, MS Teams, or face-to-face) will be

arranged and participants' preference for data collection methods (face-to-face, telephone, MS Teams, online, or post) will be determined, and consent taken. Baseline data will then be collected, using participants' preferred methods. Baseline data collection will include:

- Baseline demographic questionnaire completed including family living circumstances (including postal code, allowing coding of neighbourhood deprivation); gender, marital status, ethnic group, level of education, health/disability; parent health-related quality of life using the EQ-5D-5L; minimal information on recent resource use; number of adults and children in household; household income and financial hardship; where the child with LD lives during a normal week.
- Baseline outcome measures completed.
- These family carer participants will be followed up 12 months post-referral/re-referral for all outcome measures, as well as a measure of service satisfaction.

Service staff:

Service-level data collection

Service staff participants will be approached by sites to work with a member of the research team to complete a service-level data proforma, as described above.

External stakeholders:

External stakeholder participants will be approached by sites to complete a brief measure about their satisfaction with the service in terms of the support provided for one child who they support and who is being supported by the service. If willing to be contacted by a member of the research team, their contact information will be shared with the research team, who will arrange for to complete the consent form and data collection proforma with them, ideally in person, but potentially via MS Teams.

11.2 Qualitative case studies WP2.2

A sample of participants will be invited to an interview with a researcher (see Section 8 for more details about how participants will be selected).

If willing to take part, interviews will be conducted face-to-face for children with LD or either face-to-face, via the telephone, or MS Teams for all other participants depending on the participant's preference. Parents/guardians of children with LD and BtC will provide consent for them to take part

in Talking Mat facilitated interviews, or the informal observation with a child unable to use Talking Mats. For all other participants, informed consent will be taken before their interview.

Service-level data collection (WP2.2b)

Service staff participants will be approached by sites to work with a member of the research team to complete a service-level data proforma, as described above, specifically about costs related to co-production activities.

Figure 1. Schedule of enrolment, interventions and assessments¹

Procedures	Study timepoints			
	Screening	Baseline	Service delivery	12-month follow-up
<i>Family carer/child Eligibility</i>	X			
<i>Child Vineland Adaptive Behaviour Scales (VABS)</i>	X			
<i>Family carer Informed consent</i>		X		
<i>Family carer Contacts form</i>		X		
<i>Child and family Demographics</i>		X		
<i>Child Behaviour Problem Inventory Short Form (BPI-S)- completed by parental carer</i>		X		X
<i>Child Children's sleep habit questionnaire (CSHQ)</i>		X		X
<i>Child Non-communicating children's pain checklist (NCCPC-R)</i>		X		X
<i>Child Strengths and Difficulties Questionnaire (SDQ)</i>		X		X
<i>Child EQ-5D-Y Health Questionnaire</i>		X		X
<i>Child Either Paediatric Quality of Life Inventory (PedsQL) Generic Core Scales or Paediatric Quality of Life Inventory (PedsQL) infant scales</i>		X		X
<i>Child GO4KIDDS</i>		X		X
<i>Family carer EQ-5D-5L Health Questionnaire</i>		X		X
<i>Family carer Warwick Edinburgh Mental Wellbeing Scale</i>		X		X
<i>Family carer Experience of service questionnaire (ESQ)</i>				X
<i>Child Client service receipt inventory (CSRI)</i>		X		X
<i>Family carer Client service receipt inventory (CSRI)</i>		X		X
<i>Service-level 'Reach' to population</i>				X
<i>Service-level Timings of service delivery (e.g., mean time to starting support from referral, and length of time to discharge)</i>				X

¹ Taken from the HRA CTIMP protocol template (2016).

<i>Service-level</i> Service take-up				X
<i>Service-level</i> Costs at service level				X
<i>External stakeholders</i> Informed consent for service satisfaction				X
<i>External stakeholders</i> Service satisfaction				X
<i>Child</i> qualitative interviews (and consent)				X
<i>Parent carer</i> qualitative interviews (and consent)				X
<i>Service staff</i> qualitative interviews (and consent)			X	
<i>External staff</i> qualitative interviews (and consent)			X	
<i>Commissioner</i> qualitative interviews (and consent)			X	
Co-production interviews with <i>children</i> (and consent)			X	
Co-production interviews with <i>parent carers</i> (and consent)			X	
Co-production interviews with <i>staff (internal or external) involved in co-production</i> (and consent)			X	
Co-production interviews with <i>external staff</i> (and consent)			X	
<i>Service level</i> co-production costs				X

12 Safety reporting

There are no expected adverse events related to the intervention or research procedures. The ethics committee will be asked to approve that adverse events should not be reported for this study.

However, should any member of the research team become concerned at any point about the well-being or safety of a participant or a child involved in the study, study staff will follow a study-specific Standard Operating Procedure for dealing with harm which will be explained to participants during the consent process and highlighted explicitly in participant information sheets.

13 Statistical considerations

13.1 Sample size

Following Stage 1, we identified 5 different service models. On that basis, 15 examples of services in total will be included at Stage 2 – based on sample size calculations. To detect a standardised effect

size of 0.4 in a one-way ANOVA, with 90% power, a two-sided alpha of 0.05, and assuming five service model types identified in Stage 1, a total sample size of 120 children is required. Inflating this sample size for clustering of children within services, assuming an average number of 13 children recruited from each setting and an intra-cluster correlation coefficient (ICC) of 0.05, the required sample size increases to 195 children from 15 settings. This sample size does not currently account for loss to follow up, so we will allow up to 20% by extending our sample size to N=244 (recruiting approximately 15-16 individuals per service for 15 services). This ICC is likely conservative given the subjective nature of the outcome measures (Adams et al., 2004). In our statistical analysis, we will enhance power and precision by adjusting for individual and cluster level characteristics, including service type (which is by definition a cluster level characteristic) and corresponding baseline measures of the outcome variable.

13.2 Missing data

Detail of missing data will be described in the Statistical Analysis Plan (SAP).

13.3 Procedures for reporting deviation(s) from the original Statistical Analysis Plan

Any deviations from the original SAP will be submitted as substantial amendments where applicable and recorded in subsequent versions of the protocol and SAP.

13.4 Inclusion in analysis

All eligible services from Stage 1 of the MELD study will be considered (N=161), and sampling of services will be according to a systematic sampling process. Initially, a stratified sampling approach to selecting services that were included in MELD Stage 1 analyses was proposed. However, a potential complication of this procedure is that services may not continue involvement in Stage 2. Hence, any achieved apparently random sample would be likely be biased.

We propose the following balancing procedure within service models.

1. Rank services based on criteria indicating how likely they conform to a representative example of the service model
2. Secondly rank according to the quality of existing communication between the services and the research team (e.g., it may not be prudent to approach services whose data were obtained in the end with a FOI request).

3. A screening log describing our approach for each service and the outcome of the approach will be recorded. In addition, we will use service-level data as a proxy (that we could obtain for all services) to explore whether we are at risk of selection bias.

14 Analysis

Statistical Analysis

It is inevitable that the referral of children to different service types will be a non-random process. Therefore, a crude comparison of outcomes of children in different service types will be prone to selection bias. To minimise this, the research team and stakeholders will construct a Directed Acyclic Graph (DAG) prior to data collection in Stage 2, which will outline our core assumptions around the causal pathways between service model (exposure) and child outcomes (Williams et al., 2018). This DAG will be used to identify key quantitative data to collect (e.g., confounders, mediators, and moderators) and will be used as an aid for modelling the relationship between service model and child outcomes. The final statistical models will depend on the DAG, but will broadly be a two-level linear regression model (children nested within services), with the child outcome measure at 12-months as the outcome, service model included as a fixed effect, and the corresponding child outcome collected at baseline also included as a covariate. Results will be reported as adjusted mean differences, 95% confidence intervals, and p-values. Mediation analyses will use the bootstrapped procedure as this method has no distributional assumptions on the indirect effect and is generally more robust (Bollen & Stine, 1990).

A detailed statistical analysis plan will be written and agreed by the study management team prior to any analysis taking place. A missing data strategy will be a part of the SAP. This is most likely to include an exploration of any setting/child determinants of missing outcome data (to infer likely missing mechanisms), and an analysis that is valid under a Missing At Random assumption (e.g. using maximum likelihood methods or multiple imputation).

When fitting models to describe the relationship between our service types and our outcome/s, we will adjust for confounding using multinomial propensity score weighting, as this approach tends to preserve the majority of a study sample and therefore maximises precision of estimates. When specifying our propensity score model, we will include outcome risk factors (informed by the DAG which defines the minimally required adjustment set) and exclude strong predictors of service model inclusion that are not associated with outcome. Following this, we will inspect the distribution of

propensity scores for distributional overlap. We will consider trimming of extreme weights (i.e., probabilities close to zero or one), but will investigate the extent to which trimming limits the generalisability of our findings. We will then estimate the “average treatment effect in the whole population” (i.e., the effect of service model on outcomes in all referred children) by fitting models with inverse probability of treatment weights and robust sandwich-type estimators for variance estimation.

Economic Analysis

An economic evaluation of the selected service models will be incorporated into Work Package 2.1; data gathered about co-production activities will be incorporated into Work Package 2.2. This will take the form of a cost-consequence analysis, which will involve the identification, measurement, and valuation of all costs and consequences of the selected service models and co-production activities as well as the presentation of data in a disaggregated tabular format for comparison. The cost consequence analysis will be carried out from a public sector perspective. Data related to the direct costs of each service model will be informed by primary research (using a standard proforma) that will account for the cost of service delivery, monitoring, follow-up, and associated administrative activities. Individual-level data on broader resource consequences and associated economic costs will be informed by CSRs completed by each child’s primary parental caregiver at baseline and at a 12-month follow-up. In addition, data on each measure included in the outcomes package will be collected and reported separately. Mean costs and outcomes for each service model will be estimated together with appropriate measures of uncertainty such as standard errors and confidence intervals. To handle potential selection biases within the economic evaluation, we will apply propensity score matching methods to balance the characteristics of individuals in each comparator group and to ensure consistency with the statistical analysis, before conducting a cost-consequences analysis of the alternative service model types. Our analytical strategy will be informed by recent guidance on the conduct of health economic analyses using individual patient level observational data (Kreif et al., 2013).

A detailed Health Economics Analysis Plan (HEAP) will be written and approved prior to the completion of Stage 2 data collection.

Qualitative Analysis

A Qualitative Analysis Plan will be written for the project and approved prior to data analysis. Data will be organised using NVivo v12. Parkinson et al. (2016) argue that NVivo improves transparency of

the analysis as decisions and interpretations can be traced back to raw data. Data collection, transcription, and analysis will proceed concurrently. This gives the flexibility to explore unanticipated issues with subsequent cases. We will code the data using a Framework approach (Gale et al., 2013; Ritchie & Spencer, 2000; Parkinson et al., 2016). Ritchie and Spencer (2000) argue that framework analysis is useful to describe context, to examine reasons for what exists, to evaluate what is helpful and what can be improved and for identifying new theories, plans or actions.

Framework analysis involves familiarisation; identifying a framework; indexing; charting; and mapping and interpretation. Familiarisation involves really getting to know the data or immersion in the data. This will be achieved by listening to the interviews, reading transcripts, and assigning codes. Discussing issues that arise is an important part of familiarisation. We will discuss with the Study Management Group, including PPI partners. Identifying a framework is a careful balance between issues from the literature, as well as new and emerging issues that come from the data identified in the familiarisation stage. We will be mindful of Stake's (2006) advice to ensure we recognise new issues as they arise. We will do this by keeping careful field notes, by analysing concurrently with data collection, and discussing of expected and unexpected emerging themes within the study team; so both existing and new emerging issues can be accommodated. Indexing applies the framework to each transcript. Charting is really about making the data manageable, summarising, and organising the summaries into charts. Mapping and interpretation involves looking at the key issues in the data, making sense of the data in light of our research questions. We will also carefully consider any data that do not fit in the framework and which suggest rival explanations.

Drawing on Yin (2018) we will also examine, test and refine the following propositions to guide data collection and analysis. These draft propositions have been based on key themes in our qualitative synthesis of existing literature (Griffith & Hastings, 2014) and individual studies, and will be reviewed by the study team and PPI partners before data analysis commences:

1. Family carers of children with LD and BtC struggle to manage these behaviours, face barriers taking their children into social situations, and can become socially isolated.
2. BtC are functional – serving a purpose/having meaning for the child – and the behaviour/responses of other people is the main mechanism through which BtC are maintained.
3. The quality of the caregiver-professional relationship is central.
4. Some services, or components of services, are effective at providing support for BtC.

5. Organisational barriers can be successfully negotiated in some service models.
6. Co-production of services with family carers and children with LD may enable more appropriate services to be developed.

An important aspect of data analysis will be the cross-case synthesis. In addition to concisely describing each case including its context, we will look across cases to understand the similarities and differences between cases. Our analysis will be informed by Yin's (2018) suggestions of pattern matching, explanation building, and cross case synthesis. This involves looking at the findings in each case, comparing findings across cases, making initial statements, revising the statements in light of the second, third and more cases. We will look for rival explanations in the data (an important part of ensuring rigour) and determine whether these explanations hold up given the actual case study findings. We will refine our propositions. Yin suggests displaying and tabulating data is important, which fits well with our chosen framework analysis approach.

We will ensure there is a clear audit trail that provides a justification of our decisions. Supporting and challenging data will be presented, and in sufficient detail to demonstrate a real understanding of the area.

Integrating qualitative and available quantitative data will be important to enable a more nuanced account of the case study data (Mason et al., 2019). We plan to do this using the techniques proposed by O'Cathain et al. (2010). This will involve both "following a thread" and a "mixed methods matrix" both at the analysis stage. Following a thread was suggested by Moran-Ellis et al. (2006) who proposed an analytical theme or theme in one data set is followed across data sets. They report this as "preserving the value of the open exploratory qualitative inquiry but incorporating the focus and specificity of the quantitative data". The mixed methods matrix is where data are summarised and displayed in a matrix, to look for patterns. This will also fit well with the framework analysis proposed. We plan to use the pillar integration process (Johnson et al., 2019) to display the data. We also note that metaphors for integrated analysis in mixed methods have been proposed by Bazeley and Kemp (2012), and we will draw on these metaphors when discussing integration and interpretation with our stakeholders.

In addition to exploring how services achieve outcomes for children and their families/carers, we will use cross-case comparisons to explore how different service models help or hinder supporting these

processes, how organisational and other barriers are addressed, and how service delivery and acceptability vary according to context.

Talking Mats data

A Talking Mat will be considered completed if children are able to make valid placements of stimuli relevant to the given topic area. A discrete-response coding system will be devised for the current study to verify the validity of children's responses. Researchers will view videos of participants one Talking Mat at a time. Placements will be recorded for each stimulus presented and a confidence rating of high or low will then be made based on an estimation of the validity of the child's response. The observed position of stimuli on a mat following a participant's response will be used to code placements (i.e., the area/column depicting stimuli that is favoured/experienced frequently, or not liked/seldom experienced, or partially liked/experienced). Low confidence ratings will be made if placements appear motivated by acquiescence; are contrary to other communications (e.g., the child says "don't like" and places the item in the highly preferred column); where the child is highly distracted (e.g., placement appears non-intentional), or where placements appear motivated by a sensory stimulation function (e.g., lining items up to create a visually reinforcing display). High confidence will be assumed and rated in the absence of these low confidence indicators.

A second observer will view videos from a randomly selected 50% of participants, in each case coding at least 50% of Talking Mats from the interview and covering all categories of Talking Mat from the study overall. Inter-rater reliability (based on both placements and confidence ratings) will be calculated and reported as a percentage (total agreements divided by total agreements plus total disagreements $\times 100$). In addition, the second observer (a Talking Mats trainer), will complete the Effectiveness Framework of Functional Communication (EFFC) (Murphy and Cameron, 2008) for each mat in their sample. This tool is commonly used in Talking Mats research and provides seven 0–4-point ratings concerning quality of communicative interactions based on the behaviour of both the speaker (child) and listener (researcher). A score of 21 out of 28 represents effective Talking Mats communication.

Researchers will review transcripts in detail and analyse these using a thematic approach (Braun and Clarke, 2022) in relation to each mat, across all children. Transcriptions will be analysed alongside stimuli placement records to support an integration of both data sources and help further explore the manner in which children use the mat and perceive the topic area. Analysis involves comparing what is understood about the child's views, what (and how) views are expressed by the child during

the visit, and how the child is supported to influence interactions and events using whatever skills they have.

Observation data (WP2.2a only)

Observable indicators of the child's engagement, enjoyment, or non-enjoyment will be used to help understand the child's perspectives and experiences.

Observable indicators are observable signs (body language, facial expressions, interactions, speech or vocalisations, behaviours etc.) a person displays that express their feelings. Although there is some similarity between ways people express enjoyment, this can vary and is unique to each individual, for example some people smile when they are happy whereas other people smile when they are nervous. Asking family carers to identify observable signs of when their child is: enjoying something; not enjoying something; engaged; disengaged; and what is important to or liked by them will reduce subjectivity in the analysis. For each child, family carers will be asked to select up to five key indicators of enjoyment and engagement, and up to five key indicators of non-enjoyment and disengagement.

Observation sheets will be completed to look at what the child appeared to engage with, enjoy, and not enjoy based on these 'observable indicators' in the video recordings. This will be completed by looking at the things that happened before (the "antecedents" to) the child displaying one of their 'observable indicators'.

15 Data Management

Source data will be paper versions of the questionnaires. If physical copies of questionnaires are completed by the Research Assistant face-to face or over the telephone/MS Teams, the Research Assistant will return physical copies of questionnaires to the office as soon as possible, and will store them securely (e.g., in a locked room/cabinet in their home) until this is done. If questionnaires are posted to the participants, they will be returned in free-post envelopes to the MELD Study team. questionnaires will only contain a unique identifier (PID) per participant. No other identifiable information will be recorded on the questionnaires.

Once in the office, physical versions of questionnaires will be stored in locked filing cabinets in lockable offices, and only members of the research team will have access to these data. The Research Assistants

will enter physical versions of questionnaire data on to a secure bespoke database. Once data has been entered and checked for completeness and accuracy by an independent member of the research team (i.e., not the person who entered the data) and upon completion of the study, physical versions of the questionnaires will be destroyed. Online versions of questionnaire data will be stored on Qualtrics and downloaded weekly. The downloads will be securely stored in a study-specific folder on the University of Warwick server, only accessible to research team members. Once data collection has completed for any given part of the study, the data collected online will be transferred to the secure bespoke database with the data entered from physical versions of the questionnaires.

Access to the database will be via username and password and restricted to appropriately trained personnel only. The database will be housed on local servers managed by University of Warwick staff in accordance with all appropriate legislation.

Identifiable data will be password protected and stored separately from non-identifiable data.

Wherever possible data will be validated at point of entry, thereby reducing the opportunity for missing or unexpected data. All changes made to the data will be recorded and visible via an audit log within the database.

Qualitative interviews will be recorded on encrypted audio-recorders or on MS Teams and stored on password protected computers at site. Any transcripts will be fully pseudonymised.

A data management plan will be developed to outline the details of how data will be collected, transferred, stored, and accessed by the team.

The following source data will be collected:

Study data	Source Data					
	Contacts form	Baseline demographic questionnaire	Questionnaires	Written documentation within services	Qualitative interview recordings	Observation recordings
Contacts information	X					
Demographics		X				
Study outcomes			X			
Qualitative interview data (including Talking Mats)					X	
Observations and engagement with children with severe LD						X
Service-level data (including costs)			X			
Review of written documentation for WP2.2a				X		
Review of written documentation for WP2.2b				X		

15.1 Completion of questionnaires

The original versions of the questionnaires are to be retained at the University of Warwick. Research Assistants/Research Fellows will be responsible for data entry from questionnaires onto the online study database. A copy of the questionnaires will be checked/queried within approximately four weeks of completion. In accordance with the principles of GCP, the PI is responsible for ensuring accuracy, completeness, legibility and timeliness of the data reported in the questionnaires from services. Questionnaires from family carers will be checked by the research team.

Questionnaire pages and data will be checked for missing, illegible or unusual values (range checks) and consistency over time.

If missing or questionable data are identified, a data query will be raised on a data clarification form. The data clarification form will be sent to the relevant Research Assistant, who shall be requested to respond to the data query on the data clarification form. The original questionnaire pages should not be altered.

All answered data queries and corrections should be signed off and dated by a delegated member of staff. The completed data clarification form should be returned to the Study Manager and a copy retained alongside the participants' questionnaires.

16 Protocol/GCP non-compliance

The Chief Investigator should report any non-compliance to the study protocol or the conditions and principles of Good Clinical Practice in writing to the ethics committee and sponsor as soon as they become aware of it.

17 End of Study definition

The end of the study is defined as the date of final data collection from participants included in the research.

The sponsor must notify the HRA of the end of the study within 90 days of its completion or within 15 days if the study is terminated early.

18 Archiving

The Study Master File (SMF) containing essential documents, as specified in the University of Warwick Clinical Trials Unit Standard Operating Procedures (<https://warwick.ac.uk/fac/sci/med/research/ctu/ctuintranet/conducting/planning/sop>) will be archived following University of Warwick protocols for 10 years.

Archiving will also be undertaken at Sites, with costs included in the SoECAT to cover this.

19 Regulatory Considerations

19.1 Ethical and governance approval

This protocol will receive approval from a NHS Research Ethics Committee, and from the Health Research Authority (due to the involvement of NHS services).

19.2 Data Protection

The research team will act to preserve participant confidentiality and will not disclose or reproduce any information by which participants could be identified, except where specific consent is obtained, or if abusive practice is disclosed that researchers would have a duty to report. Data will be stored in a secure manner and will be registered in accordance with the Data Protection legislation (in accordance with GDPR). Participants will always be identified using a unique PIN. All other identifiable information will not be stored with collected data.

19.3 Indemnity

The University of Warwick has in force a Public and Products liability policy, and a professional Indemnity policy which provides cover for "negligent harm" and the activities here are included with in that coverage subject to the terms, conditions and exceptions of the policy. The University of Warwick does not provide compensation for non-negligent harm.

19.4 Study sponsorship

The University of Warwick will act as Sponsor for study.

19.5 Funding

The study is funded by National Institute for Health Research (NIHR) Health Services and Delivery Research (HS&DR) programme (Ref NIHR 129577).

20 Study management

20.1 SMG (Study Management Group)

The SMG, chaired by the Chief Investigator, will meet monthly and will include all Investigators, and all employed project staff to discuss study progression and key management issues. SMG members have signed up to the remit and conditions as set out in a SMG Charter.

20.2 SSC (Study Steering Committee)

A SSC has been established and will meet at least twice during this stage project. There is an independent chair with expertise in child learning disability applied research. Other independent members include two senior child learning disability NHS clinicians (from Wales and Northern Ireland – independent of this England-focused study), a statistician, a health economist, and two family carer representatives (parent/carers of a child with LD and BtC, one of whom is the carer of a child with more severe LD and BtC); along with non-independent members: CI, Study Statistician (Paul Thompson), the Study Manager (Samantha Flynn), and study Research Assistants (Nicholas Manktelow and Emma Taylor) as observers. The SSC will provide overall supervision for the study and provide advice through its independent chair. The ultimate decision for the continuation of the study lies with the SSC. SSC members will be required to sign up to the remit and conditions as set out in a Stage 2 SSC Charter which will be filed in the SMF. The SSC will determine whether an independent Data Monitoring and Ethics Committee (iDMEC) is required for the study or whether the SSC will take on data monitoring function. As this is a low-risk study with no blinding or delivery of intervention, it is expected that an iDMEC will not be required.

20.3 iDMEC (independent Data Monitoring and Ethics Committee)

See 20.2.

20.4 Project Advisory Groups (PAGs)

To support PPI input and ongoing input from professionals, two Project Advisory Groups (PAGs) have been established: (i) family carers of children with LD and BtC, supported by co-investigators Grant and Cooper and our PPI partner organisation the Challenging Behaviour Foundation, and (ii) professionals working in child LD BtC services (clinicians, commissioners, service managers). These groups will not have a formal governance role, but will contribute to key decisions throughout the research, advise on engaging children with LD and BtC, their family carers, professionals and services, and interpretation of the study findings. The PAGs will also advise on information sheets and other ethics matters, and on co-production of dissemination outputs, act as ambassadors for the research project, and creating communication pathways with family carers and professionals. The Family Carer Advisory Group (FCAG) will meet three times during the project, and the Professionals PAG will meet less frequently but will be consulted regularly by email and in individual or small meetings, to ensure that PPI involvement and consultation with professionals is regular and closely informs the whole project.

20.5 Planning for the effects of COVID-19

Consent and data collection will primarily be completed via telephone, MS Teams, online, and by post and it is anticipated that this can continue during COVID-19 restrictions. Some participants may request that data collection is completed face-to-face so, providing this is possible at the time, we will accommodate these requests. Should face-to-face data collection become impossible because of COVID-19 restrictions, we will collect all data via telephone/online. Provided that services continue to deliver support, the study will continue to collect data in some form.

It may also become impossible to complete Talking Mats data collection with children with LD face-to-face in which case they would be undertaken online. Co-applicant Bradshaw has expertise in completing Talking Mats online and will provide training to other members of the research team in how to complete these face-to-face and online, in case of COVID-19-related restrictions. If COVID-19-related-restrictions are enforced during the course of the study and in person data collection with children with severe LD is not possible, we will look to adapt these procedures to take place virtually. Co-applicant Bradshaw is also experienced in virtual methods of observation.

21 Quality Control and Assurance

21.1 Monitoring

Investigators will facilitate study related monitoring, including audits and regulatory inspections, by providing direct access to source data/documents as required. Participant consent for this will be obtained. Findings generated from any monitoring will be shared with the Sponsor.

21.2 Audits and inspections

This study may be subject to inspection and audit by the University of Warwick under their remit as Sponsor.

22 Publication policy

Outputs from the MELD study will include open access peer reviewed journal articles in international academic journals, presentations at national and international academic conferences and at public engagement/dissemination events. All publications and presentations relating to the study will be authorised by the SMG. A project publications policy and plan will be produced and approved by the

SMG, and will include the key principles from the NIHR Open Access Policy
(<https://www.nihr.ac.uk/documents/nihr-open-access-policy/28999>)

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