Pay More Attention: A national mixed methods study to identify the barriers and facilitators to ensuring equal access to high quality hospital care and services for children and young people with learning disabilities and their families – Phases 2-4

Statement of the problem

Despite comprehensive evidence of health inequalities for adults with learning disability (LD), including poor practice, discrimination and abuse in hospitals, there has yet to be a comprehensive review of how well hospital services are meeting the needs of children and young people (CYP) with LD and their families. Qualitative evidence of parental dissatisfaction with the quality, safety and accessibility of hospital care for CYP with LD exists. However, the extent to which their experience differs from that of parents of CYP without LD is not known. Moreover, the views and experiences of CYP with LD are missing. A major uncertainty is when the inequalities known to exist for adults with LD start to emerge - is it simply when CYP with LD make the transition to adult services or are they apparent as soon as a they are first recognised as having LD? We aim to address this gap in our understanding by comparing how services are delivered to, and experienced by CYP with and without LD and their families to see what inequalities exist, for whom, why and under what circumstances. This information will:

- **provide evidence for the development of new interventions and improved use of existing ones;**
- **promote equality to be better target interventions to those who need them, when they need them;**
- **resulting in a more efficient and effective service that patients and parents are satisfied with.**

Summary of project plan

A transformative, mixed methods case study design[1], prioritising the voices of CYP and their parents. Each case will be represented by National Health Service (NHS) hospitals in England. Data will be gathered concurrently, over 4 phases as shown below:

Phase	Aims	Setting	Methods
1 Completed	-To describe the organisational context for healthcare delivery to CYP with LD and their families -To compare how equipped hospital staff feel they are to identify the needs of CYP with and without LD and their families and provide high quality care to effectively meet these needs.	24 hospitals in England	STAFF: Interviews with senior managers STAFF: Questionnaire to clinical teams Content analysis of hospital documents
2	-To compare how hospital care and services are experienced by CYP with and without LD and their families -To compare staff experiences of caring for CYP with and without LD and their families -To investigate differences in safety concerns between parents of CYP with and without LD -To investigate differences in access to hospital services between CYP with and without LD	6-8 hospitals in England sampled from Phase 1	CYP: Interviews using creative and digital research techniques PARENTS: Hospital diary, reports of safety concerns, home interview STAFF: Interviews HOSPITAL ACCESS: Retrospective mapping of hospital inpatient and out-patient activity
3	-To compare levels of satisfaction with hospital care between CYP with and without LD -To compare levels of satisfaction with hospital care between parents of CYP with and without LD	6-8 hospitals in England sampled from Phase 1	CYP: Satisfaction questionnaire PARENTS: Satisfaction questionnaire
4	-To synthesise study findings and develop content of a DVD for training staff and students	Suitable workshop setting that is accessible and has good facilities	A workshop involving study participants and experts in the field of LD

The study has been designed to meet the requirements of FINER:

Feasible: Parents of CYP with and without LD have reviewed the research questions and methods of data collection and their feedback has been incorporated into the project design. The research team has the relevant experience and expertise to deliver the project in a timely way. KO, FG, JW and SK have collaborated on grants, including those funded by NIHR. KO (CI) has just completed a single site ethnographic study exploring the needs and experiences of CYP with LD and their families during hospitalisation, and the staff caring for them. One co-applicant (IWT) has recently completed a multi-site HS&DR funded project into healthcare provision for adults with

LD in hospital. AH has extensive experience of LD related issues, multicentre methodologies and expertise in transition and adulthood related issues in those with LD.
Interesting: We will combine traditional, creative and digital methods to make the project interesting and accessible to participants. Hospitals can be very stressful for parents and leaving their child, even briefly, can generate anxiety. A hospital diary will enable parents to upload 'real time' data at their convenience.
Novel: The inclusion of CYP with LD, a vulnerable population who are frequently excluded from research. Our inclusive approach means interviews will be tailored to meet the individual learning, physical, communication and health needs of *all* participants. The research team has extensive skills in working with CYP of all ages, both with and without LD. One of the co-applicants (SK) is a parent of more than one CYP with LD.
Ethical: The team has good understanding of the ethical issues of involving this group of participants in research and has experience of gaining ethical approval for their inclusion. The funds requested will allow the appointment of skilled researchers and the use of appropriate resources to involve CYP in a meaningful way and ensure findings are disseminated in multiple formats. One co-applicant (FG) is alternate chair for an NHS ethics committee.
Relevant: This study supports the HS&DR mission of inclusive research, and bridging the gap between theory and practice. The voice of two groups of CYP with long-term health conditions, those with and without LD, will be heard. Through directly comparing their hospital experience and that of their parents and the staff who care for them, we will identify what facilitates and prevents the provision of an equitable service. A major gap in the evidence base, addressed through this study, is what CYP with LD and their families think about the hospital service they receive.

Background

Definition and prevalence of learning disability in children and young people

The term "learning disability" covers a wide spectrum of impairments. The presence of a low Intelligence Quotient is a defining feature. The definition does not include CYP who have learning *difficulties* that may impair educational attainment, e.g. processing problems, but who are within the average range of intelligence or those with developmental delay who are late in reaching some or all of their developmental milestones. Whilst LD is the preferred term of the Department of Health[2], terminology varies internationally, with 'intellectual disability' and 'mental retardation' also used. In the 10th international classification of diseases (ICD-10)[3] mental retardation is defined as "… a condition of arrested or incomplete development of the mind, which is especially characterised by impairment of skills manifested during the developmental period, which contribute to the overall level of intelligence, i.e. cognitive, language, motor and social abilities". For a diagnosis to be considered '*definite'*, reduced intellectual functioning must result in "diminished ability to adapt to the daily demands of the normal social environment", but notably there can be wide variations in the levels of skills developed by individuals.

Advances in technology have dramatically increased rates of survival and life expectancy for premature babies[4], a significant proportion of whom will subsequently have intellectual impairment[5,6] and life-long health problems[7]. Moreover, technological advancements in equipment have improved life expectancy for those with medical conditions and genetic disorders who previously would not have survived[8]. It is now widely acknowledged that the numbers of those with severe intellectual impairment and multiple/complex long term problems will continue to rise[9]. There are no definitive figures for the prevalence of LD. Whilst estimates indicate a worldwide prevalence similar to one of schizophrenia at 1.0%[10], there are huge variants within that depending on the definition applied. Latest figures indicate that there are 286,000 CYP age 0-17 years with LD in England[11].

Health Inequalities for people with LD

It is widely recognised that people with LD have more health needs that often remain unmet than the general population. In 2007 Mencap published "Death by indifference"[12] detailing case histories of 6 people with LD who died in hospitals from avoidable conditions and calling on the government to take "serious action". An Independent Inquiry into access to healthcare for people with LD followed, revealing significant system failures and reporting that patients with LD were treated less favourably than others, resulting in prolonged suffering and inappropriate care. The report of this Inquiry, "Healthcare for All"[13], identified the invisibility of people with LD within health services, and the lack of priority given to identifying their particular health needs. Training and education about LD was found to be very limited. Combined with ignorance and fear, lack of training was identified as reinforcing "negative attitudes and values towards people with learning disabilities and their carers" and "contributing significantly to a failure to deliver equal treatment, or to treat people with dignity or respect". A need to strengthen the systems for assuring equity and quality of health services for people with LD at all levels was identified.

The degree to which the experiences of CYP with LD or their families were included in this Inquiry is unclear. It was reported only that the 412 responses, included 76 from *family carers* and 100 from *people* with LDs. The limited direct reference to CYP with LD in the report presents a mixed picture. Services were praised for "providing all round care", yet access to general health care was reported as being "as problematic as it appears to be for adults". Accounts were provided of staff suggesting that "disabled children should be allowed to 'slip away' rather than resuscitated or treated", and communication about treatment and options was reported to be particularly poor.

Furthermore, both parents and carers of adults *and* children with LD, *"find their opinions and assessments ignored by healthcare professionals … They struggle to be accepted as effective partners in care by those involved in providing general healthcare; their complaints are not heard; they are expected to do too much of the care that should be provided by the health system and are often required to provide care beyond their personal resources"*.

A Confidential Inquiry into premature deaths of people with LD (CIPOLD)[14] including 14 CYP aged 4-17, followed. It emerged that "more people with LD died from causes that were potentially amenable to change by good quality healthcare". All aspects of care provision, planning, coordination and documentation were found to be significantly less good for people with LD. A plethora of recommendations and guidelines are now available to support hospitals in ensuring that "people with LD are included as "equal citizens, with equal rights of access to equally effective treatment"[13]. Mencap has worked with healthcare professionals and Royal Colleges to develop the 'Getting it Right Charter"[15], highlighting key activities that all healthcare professionals should undertake to ensure that there is equal access to health, including the appointment of a Learning Disability Liaison Nurse (LDLN) in every hospital. Whilst 200 Trusts, hospitals and organisations have signed up to the Mencap Charter demonstrating their commitment to change, a current feasibility audit of adult LD care pathways found that only 56% of the 9 acute trusts that took part had a liaison nurse in place[16]. Providing reasonably adjusted services for people with LD is a legal requirement[17]. Yet, the largest study of its kind to date[18] found that the delivery of reasonable adjustments in the adult hospital setting was haphazard, with a lack of a) effective systems for identifying patients with LD and b) clear lines of responsibility for implementing reasonably adjusted care to individual patients. Dr Tuffrey-Wijne and Baroness Hollins, Emeritus Professor and President of the British Medical Association, subsequently argue that, "the NHS is still failing patients with intellectual disabilities in all departments, all specialties and all services"[19]. Drawing on the findings of the recent Francis inquiry, they argue that those with LD perhaps more than any other patients "need special focus, knowledge and skill to ensure their healthcare needs are met"[20].

The direct relevance that current recommendations (detailed synthesis uploaded separately) about the care of 'people' with LD have to CYP, and guidance on the best way to implement them in the child health setting, is missing. The main thrust of initiatives aimed at reducing health inequalities faced by people with LD has been on improving access to healthcare among *adults* rather than the health inequalities faced by CYP[22]. Hence, we do not know:

- **The extent to which available recommendations *should* be applied to CYP with LD;**
- **To what extent they *are* being applied to CYP with LD;**
- **If they are being applied, what *difference* they are making to patients, parents and staff.**

<u>CYP with LD and their families</u>

CYP with LD routinely experience particularly poor health outcomes. A review of the evidence on the prevalence and determinants of health conditions and impairments among CYP with LD in the UK[21] has found that the risk of children being reported by their main carer (usually their mother) to have fair/poor general health is 2.5-4.5 times greater for those with LD compared to their non-disabled peers[22, 23] a finding only partially accounted for by differences in socio-economic status[11]. As well as having intellectual impairment, these CYP may have sensory impairments and physical impairments e.g. cerebral palsy[24] that adversely affect their speech, feeding and mobility. CYP with LD are also almost twice as likely to report 3 or more health problems and more than 4 times as likely to suffer from a psychiatric disorder than children without LD[23,25] . Increasing numbers are dependent on technological equipment for their survival[26].

Children with disabilities experience more frequent and lengthier hospital admissions than children without disabilities[27] and have contact with numerous professionals, often attending the same hospital many times in a week[28]. They are also more likely than other children to be absent from school. In those with profound multiple learning difficulties, 62% of absences were accounted for by illness and 13% from attending medical/dental appointments[11]. The ability for CYP with LD of *all* ages to understand information about hospital care and treatment will be limited, they may not be able to communicate their needs verbally, and may need additional support with all aspects of hospital life. Whilst many CYP will find it hard to cope emotionally when they are in an unfamiliar hospital environment, those with LD who have challenging behaviour [29] may find it particularly difficult.

The physical, emotional, social and financial impact on parents caring for a child with LD are well documented and have been acknowledged for decades[30-32] They are more likely than other parents to suffer from stress and depression, marriage breakdown, social isolation, poverty, and poor physical health[11]. Whilst parents of all children may find it stressful accompanying their child into hospital, we know that parents of CYP with LD can find it particularly challenging especially when they do not have confidence in the staff caring for them or they feel they have to fight to get the services their child needs (see below). In the CI's most recent study, aimed at understanding the needs and experiences of CYP with LD and their families in hospital, some nursing staff revealed concerns that this population of parents were not given sufficient opportunity to have a break from their care-giving responsibilities. They felt that some staff relied too heavily on them, in part because they did not have the time or confidence to negotiate responsibilities[33]. A nurse working in a local hospital that we consulted about our proposed project expressed similar concerns about this group of mothers “being left to do everything”.

Within the National Service Framework (NSF) for CYP in hospital[34] the distinct service requirements of ‘disabled’ children are recognised, as is their greater need for personalized, child-centred care. However, the NSF framework precedes the latest evidence on the care of people with LD in hospital and may no longer be fit for purpose for meeting the specific intellectual, emotional, social and physical needs of CYP with LD. A number of children's hospitals have introduced nursing posts with a specific focus on improving care for CYP with LD but provision varies geographically and over time and has not been formally evaluated. In one tertiary paediatric hospital for example, there has been sustained commitment and support for improving care to CYP with *autism* and their families. However, this work does not include the wider population of CYP with LD. **This example highlights the repeated issue of CYP with LD falling between the gap in research and provision. Many will have specific needs that are very different to other disabled children, those with autism and adults with LD.**

Expressed Need

Many reports have expressed the need to review NHS services for *disabled* children and their families. The most consistent message is that services need to be tailored to meet the individual needs of these patients and it is imperative that their views are incorporated at every level of service delivery. This message applies equally, if not more so to CYP with LD, whose struggle to get their views heard is widely recognised. The National Children’s Bureau[35] refer to them as *“an invisible group in society,* arguing that, “*the more complex their needs, the more invisible they appear to become”.* A review of children’s services within the NHS[28], identified that care and services for disabled CYP were frequently overlooked and in need of attention. The battle that parents of these children feel they face in getting access to suitable health services was highlighted, as was their frustration at the lack of co-ordination between services. Kennedy (2010) described the “often hostile environment” they had to navigate through to get help for their child. He also drew attention to the lack of streamlining of hospital appointments, in particular them being scheduled on consecutive days at multiple locations at great time and cost to families. **Two recommendations from the Kennedy report have direct relevance to this study – first, that there should be a single criterion for measuring the quality of NHS services for CYP – that being satisfaction with the outcome achieved (Recommendation 29) and second, that NHS services for CYP should be designed, organised and delivered from the perspective of the CYP and parent or carer (Recommendation 33).**

Concerns about inequality in the health system have been recognised by the CYP’s Health Outcomes Forum[36], who draw attention to the fact that disabled children “face even poorer health outcomes” not just in their health, but also in their social and economic potential. It was further identified that these CYP and their families “really struggle to get their voices heard and to be involved in decisions about their own health”. The way services are commissioned and provided to CYP with a disability and their families has also been found to be variable, leading to calls for work “across the range of services … covering the standard of, access to, co-ordination of and continuity of care”. The CQC[37] strongly recommend that commissioners “take the lead in establishing a real, local understanding of the needs of children with disabilities and their families, and respond by ensuring the most suitable provisions are in place”.

In the CMO’s latest annual report[38], it was recommended that a range of measures to reduce barriers to the participation of disabled children and their families were needed to ensure their individual needs were met. In particular, provision for CYP with LD and mental health issues, including challenging behaviour was described as “very unsatisfactory” and evidence of their needs lacking. There was further endorsement that “at every level services should be shaped by the needs, wishes and aspirations of both children and their families”. Few researchers have explored the hospital experience of parents of CYP with LD to see how acceptable and effective services are in meeting their individual needs. Hitherto, the focus has been on improving the organisation and delivery of services in the community. With the introduction of Education, Health and Care Plans there is greater emphasis on looking at *all* the needs a child has across education, health and social care, with hospital Trust’s having a clearly defined role. Our proposed national study will provide evidence of what prevents and supports CYP with LD, including those with mental health issues and challenging behaviour from receiving equal access to high quality hospital care. It will provide an opportunity for CYP with LD and their families to explain how their needs can be met when they are in hospital. **As Professor Dame Sally Davies states[38], “children are different from adults … children need different approaches”. That message applies equally to hospital services and we have a responsibility to ensure that CYP with LD are not facing the same, or different inequalities as have been identified in the adult setting.**

Generating new knowledge

This inclusive study will provide evidence of whether, and what, inequality exists, for whom and why. A key strength is that is has been designed to generate evidence of what issues affect *all* CYP with long-term

conditions and what are particular to those with LD. This evidence is needed to understand the context for making reasonable adjustments for particular populations of CYP on the basis of their specific intellectual, emotional, social and physical needs. This should help address the gap resulting from the focus of policy and research being on two key areas: a) providing safer hospital care for adults with LD and b) providing more integrated healthcare services for disabled CYP. CYP with LD have unique needs that must be taken into account by commissioners and providers of hospital services. Implementing the practical suggestions and guidelines resulting from this study will help to ensure that resources and interventions that promote equality are better targeted to those who need it, when they need it, with the potential for multiple improvements:

1. Fewer unmet needs
2. Reduction in adverse health outcomes and long-term damage to CYP from poor practice
3. Reduction in seriousness of complaints and numbers of clinical incidents
4. Increase in patient & parent satisfaction
5. Improved patient & parent experience
6. Increase in staff confidence & satisfaction
7. Increase parental trust in staff and confidence in care
8. Reduction in numbers of patient hospital visits
9. Reduction in delayed discharge
10. Reduction in financial cost to NHS and families

Paying more attention to the needs of our most vulnerable CYP will help get it right for all.

Increased acceptability and effectiveness of services

Few researchers have focused on how acceptable and effective hospital services are in meeting the needs of CYP with LD and their families. More importantly, the voice of CYP with LD is largely non-existent. Conversely, there has been some research conducted with CYP without LD, including those with long-term conditions, to understand the hospital experience from their perspective[39-43] We know from this body of work the range of fears and anxieties that CYP can have about being in hospital, as well as some understanding of what supports them to feel safer, happier and more positive about their experience. What we do not know is whether CYP with LD have the same needs and experience. A recent review of qualitative studies reporting on the experience of *disabled* children as inpatients[44] led to the conclusion that their experience was “variable and not always optimal" and that providing information … would improve their experience. **Importantly, of the 8 studies included in this review, only 2 focused specifically on the care of children with LD and within these, only two individual CYP were interviewed.** Of significance, is that these 2 CYP, despite talking positively about nursing staff, were reported to be “less positive in general about their hospital stay than their parents”. Similarly, in a small Australian study[45] exploring the views of four children with cerebral palsy about their experience of the medical consultation, it was reported that, “whilst children and mothers had similar views about communication, there were obvious differences in what was perceived to be important.” Children described wanting to be included even if they did not understand what was being said, and expressed a desire to be informed of any tests or procedures before they happened, rather than having things ‘done’ to them. From this small body of evidence we can draw 3 important conclusions:

1. Evidence of what CYP with LD think about hospital and what they want from hospital services is lacking
2. Given opportunity, some CYP with LD are able to share views about hospital and what is important
3. CYP with LD do not necessarily view hospital in the same way as their parents

We will address this major gap in the evidence base by placing CYP with LD at the centre of the study and ensuring, where at all possible, that their voice is heard. We know from our own experience and that of Sharkey et al.[46] that recruiting CYP with LD into research whilst they are in hospital can be challenging. However, this should not preclude their involvement and we have taken steps to enhance recruitment (see table of risk p20).

A small body of qualitative research has been conducted with parents of CYP with LD to understand their own and their child’s experience of hospitalisation[47,48] Avis and Reardon[47] explored parents’ perceptions of nursing care and attitudes and how their child’s experience could be improved. They report parental feelings of stress, anxiety and fear, an expectation to care for their child, a lack of trust and confidence in staff and a lack of information and preparedness. Communication with staff was reported as the biggest issue that needed addressing. More recently Sharkey et al.[46] have reported on the barriers and facilitators to communicating with disabled children when inpatients. Interviews with parents and professionals revealed that, “communication with disabled children on the ward was perceived as less than optimal” and that “staff perceived time pressures and lack of priority given to communicating directly with the child as major barriers”. They found that parents could feel a “weight of responsibility” concerning their child’s communication that could make them reluctant to go home and leave their child alone. A large qualitative study[33] carried out by the CI as part of her PhD supports these findings. Parents described a sense of devoted protection towards their child with LD, which meant they were simply not willing to take any risk by leaving their child in the care of someone they did not have complete confidence in. Moreover, on the rare occasions when they felt they had no option but to leave their child, the occurrence of any problems could devastate trust in the overall system, with some refusing to access those particular services again. Ultimately parent’s felt they had to take complete responsibility for their child’s health and well-being, even in hospital. The general tone was one of apprehension that other care-providers lacked the specialist knowledge they held about their child; anger that their advice was often ignored; and concern that others did not share their dedicated commitment to the child. A feeling that professionals devalued both them and their child with LD was also reported.

Why this research is needed now

1. The numbers of CYP with LD and complex, long-term health needs who are more likely to need hospital services is growing, which means ensuring equitable access to hospital care and services is of ongoing concern.
2. We have the benefit of being able to learn from the costly mistakes that have occurred in the adult hospital setting and have an opportunity to prevent CYP with LD and their families from experiencing the same fate.
3. There is currently a lack of comprehensive evidence of how well *hospital* services are meeting the needs of CYP with LD and their families. They have unique needs that are different from adults with LD and the wider population of disabled children that need investigating.
4. The proposed study builds directly on the methodology and findings of research conducted by the applicants. As discussed earlier, the CI's PhD research revealed that rather than seeing hospitals as a place of safe care, parents tended to view them with trepidation. Her more recent ethnographic research conducted in a single tertiary paediatric hospital setting, revealed that meeting the specific non-medical needs of CYP with LD presented a challenge to hospital staff where the focus was on providing highly specialist, complex medical care for all its patients. Key ‘barriers’ included a lack of clear understanding of a) which CYP had LD b) how much CYP with LD could understand and c) how best to communicate with CYP with LD. Staff identified that having more time, resources and training would help them provide the individualised approach to care that these patients needed[49]. Through this research we learnt how to meaningfully engage CYP with LD in hospital-based research, as well as the practicalities of involving their parents and hospital staff[50]. A co-applicant (ITW) has recently completed an NIHR funded, mixed methods study into hospital provision for people with LD. We have benefitted from knowing what worked well in delivering that project as well as the key challenges[18].
5. The recently launched Children and Families Act[51] places new duties on a) NHS Trusts to inform parents and the local authority if they believe a child under compulsory school age is disabled or has a special educational need and on b) partner commissioning bodies including the NHS commissioning Board to plan and jointly commission the education, *health* and care provision for disabled children and those with special educational needs. It is specified that CYP must receive any health care provision specified in their education, health and care plan.

Aims and objectives

Primary aim:

1. To identify the cross-organisation, organisational and individual factors in NHS hospitals that facilitate CYP with LD and their families receiving equal access to high quality care and services.
2. To identify the cross-organisation, organisational and individual factors in NHS hospitals that prevent CYP with LD and their families receiving equal access to high quality care and services.

Secondary aim: To develop guidance for NHS Trust’s about the implementation for successful and effective measures to promote equal access for CYP with LD and their families.

Research Questions
From the perspectives of the families and clinical staff:

1. Do CYP with and without LD and their families have equal access to high quality hospital care that meets their particular needs?
2. Do CYP with and without LD, assisted by their families, have equal access to hospital appointments, investigations and treatments?
3. Are CYP with and without LD and their families equally involved as active partners, in their treatment, care and services?
4. Are CYP with and without LD and their families equally satisfied with their hospital experience?
5. Are safety concerns for CYP with and without LD the same?
6. What are the examples of effective, replicable good practice for facilitating equal access to high quality care and services for CYP with LD and their families at the study sites?
7. What indicators from the data and the literature suggest the findings may be generalisable to adults with LD and other CYP with long-term conditions in the hospital setting?

Theoretical/conceptual framework

This study takes a systematic approach to an empirical identification of the factors that affect access to high quality hospital care for CYP with LD and their families. Building on Tuffrey-Wijne et al.[52] a theoretical framework for understanding the range of factors at the organisational and individual level that might impact on the delivery of hospital care to CYP with LD and their families has been devised (Figure 1). A synthesis of existing literature and the team’s expertise and research informed its development. Included, are outcomes that might be associated with effective measures for promoting equal access. We intend to re-populate this framework with barriers and facilitators to promoting equal access to safe, high quality hospital care for CYP with LD and their families identified by systematically testing the theoretical and empirical framework throughout this study. From this theoretical framework we will construct a detailed research framework, comprising specific research questions.

Design

A transformative, mixed methods case study design[1] will be used. Case study research provides "a voice to the powerless and voiceless"[53] and so will enable the views of CYP and their parents to be prioritised. Case study design is "an empirical inquiry that investigates contemporary phenomena in depth and within its real-life context" [54]. In this study, a single hospital site represents each case and 4 cases will be included. In each hospital, for every CYP with LD recruited, a CYP without LD will be recruited as a comparator case, thereby allowing the experience of the 2 groups of patients to be compared. This is a complex study, requiring data to be gathered concurrently, over 4 distinct phases (Figure 2). Case study design is characterised by a convergence of diverse sources of quantitative and qualitative data (Figure 3) and is therefore well-suited to evaluating the multiple elements likely to shape and influence whether CYP with and without LD and their families receive equal access to high quality hospital care and services. The production of rich descriptions of the phenomena, through in-depth interviews and digital research methods will allow the many complexities of the situation and factors that can contribute to those complexities to emerge[55]. FG will provide specific expertise in the use of case study design.

Operational definition of LD

The theoretical definition of LD is not always easily operationalised in practice. Among very young children, only severe LD is likely to be apparent[56] and some never receive a formal diagnosis of LD but remain categorised as having 'developmental delay' or a 'syndrome without a name'. Moreover, as highlighted hospital staff do not always know what is meant by LD or which CYP on their ward have this diagnosis. For our purposes, a CYP will be classified as having a LD if any **ONE** of the following is documented in the medical notes:
a) The CYP has a LD
b) The CYP has a condition, which is always accompanied by some degree of LD, e.g. Down syndrome
c) The CYP has global developmental delay (GDD) and they are aged over 10 years old*
d) The CYP attends a special needs school and their parent verbally confirms the child has a LD

*GDD refers to CYP who are late in reaching their developmental milestones, which can be an indication of LD but may also be caused by factors such as inadequate stimulation, malnutrition, psychological and familial situations. In practice, some CYP will remain labelled as having GDD, even when other causative factors have been ruled out and a diagnosis of LD is assumed by process of elimination - we want to make sure these patients are not excluded from taking part solely on the basis of terminology.

A broad approach is taken when recruiting CYP with LD because it is entirely those issues around the identification of this population that need exploring. If a CYP is recruited to the study does not have a LD they will remain in the study as a comparator case. In this way, we maximise the diversity of the sample and minimise sampling bias (see Table of risk p20).

CYP with LD will be broadly matched with another CYP with a long-term condition. CYP with LD will be identified and approached for the study first. Once the CYP with LD has participated in the study i.e. data collection has taken place, then screening can commence for their matched CYP without LD. They will be matched on four criteria:

1) **Age**: 3 age ranges: 5-7, 8-11, 12-15
2) **Reason for admission**: to match e.g. surgical with surgical, medical with medical, tests with tests
3) **Expected length of stay**: 3 lengths of stay: Short: 1-2 nights; Medium: 3-7 nights; 8 nights and over
4) **Co-morbidities**: within a reasonable range if a CYP with LD had e.g. 7 co-morbidities then a match may be around this number.

The aim is to recruit two samples of CYP with and without LD who are of similar age, with equal complexity of health needs and who are admitted to the same hospital during the study period. It is important to include a range of CYP with varying degrees of LD. For those with severe or profound LD who are unable to participate in an interview, parents will be invited to act as a proxy for the CYP data collection methods. Definitions for all of the matching criteria will be agreed prior to data collection.. We will use real life case examples to train staff at the study sites about recruitment and they will have these case examples to refer to.

Selection of hospital sites

Phase 1: Mapping of organisational context and staff questionnaire

24 hospitals in England took part in Phase 1. This ran between January 2016 and May 2016. (IRAS: 193932)

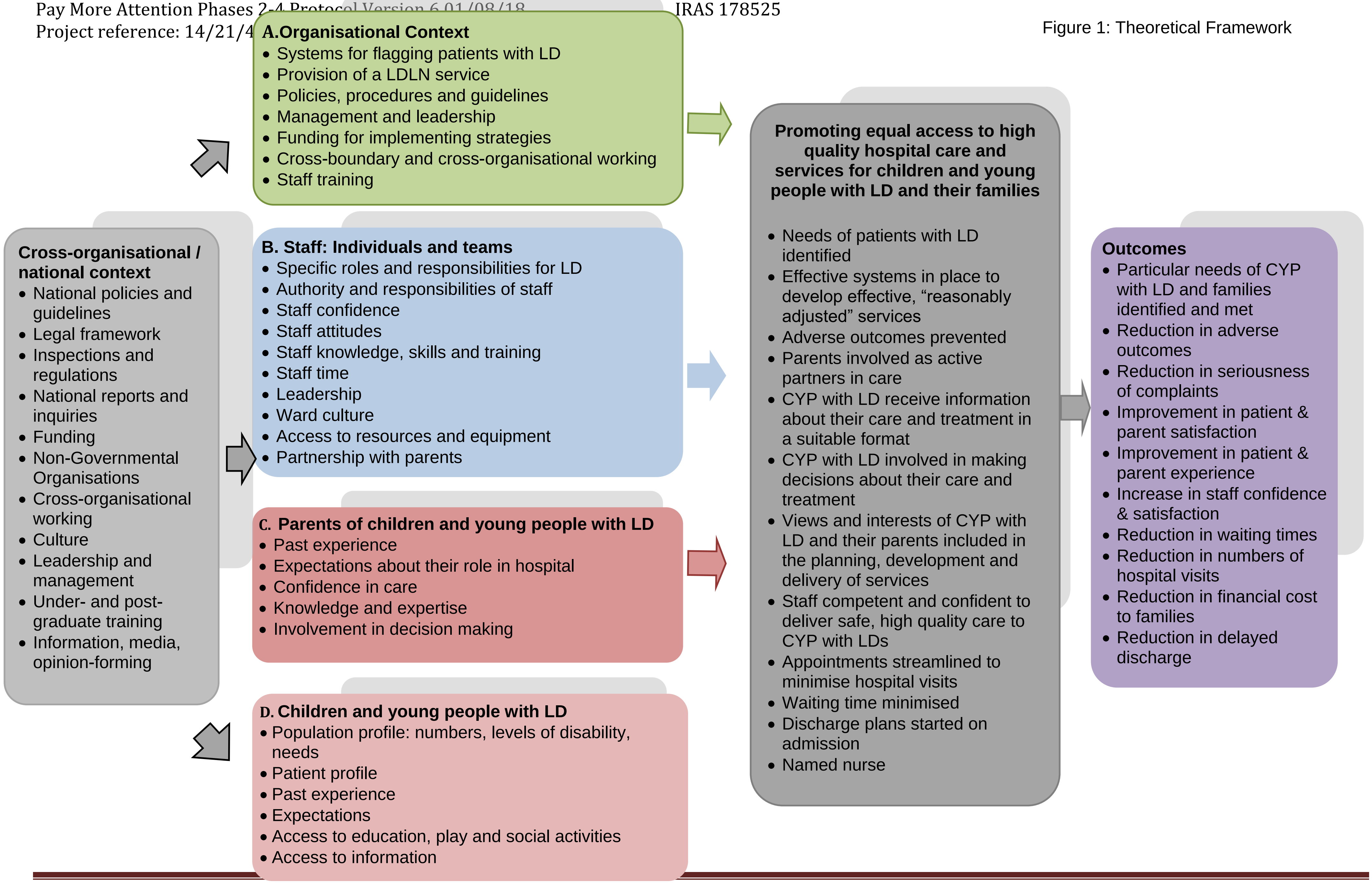
Phase 2: Case studies

To ensure successful study completion, the 24 hospital sites from Phase 1 will become eligible for Phase 2 if they fulfil 2 criteria as ascertained during the mapping exercise: 1) Accessibility to sufficient numbers of CYP with and without LD, 2) Good hospital engagement with Phase 1.
Eligible hospitals will be grouped according to whether they have a lot, a little, or no initiatives/appointments of an LD professional with a remit to improve care for CYP with LD. This important step in the selection process will ensure variability in amount of provision provided. Members of the Study Steering Committee (SSC) will be asked to design scoring criteria to enable objective selection of the 4 sites for phase 2 based on:

1. The strength of organisational context for delivery care to CYP with LD
2. Staff's perceived ability to identify and meet the needs of CYP with LD
3. Initiatives/appointments of an LD professional with a remit to improve care for CYP with LD

Sites will be anonymised to prevent selection bias.

Figure 1: Theoretical Framework



A.Organisational Context
- Systems for flagging patients with LD
- Provision of a LDLN service
- Policies, procedures and guidelines
- Management and leadership
- Funding for implementing strategies
- Cross-boundary and cross-organisational working
- Staff training

Cross-organisational / national context
- National policies and guidelines
- Legal framework
- Inspections and regulations
- National reports and inquiries
- Funding
- Non-Governmental Organisations
- Cross-organisational working
- Culture
- Leadership and management
- Under- and post-graduate training
- Information, media, opinion-forming

B. Staff: Individuals and teams
- Specific roles and responsibilities for LD
- Authority and responsibilities of staff
- Staff confidence
- Staff attitudes
- Staff knowledge, skills and training
- Staff time
- Leadership
- Ward culture
- Access to resources and equipment
- Partnership with parents

C. Parents of children and young people with LD
- Past experience
- Expectations about their role in hospital
- Confidence in care
- Knowledge and expertise
- Involvement in decision making

D. Children and young people with LD
- Population profile: numbers, levels of disability, needs
- Patient profile
- Past experience
- Expectations
- Access to education, play and social activities
- Access to information

Promoting equal access to high quality hospital care and services for children and young people with LD and their families
- Needs of patients with LD identified
- Effective systems in place to develop effective, “reasonably adjusted” services
- Adverse outcomes prevented
- Parents involved as active partners in care
- CYP with LD receive information about their care and treatment in a suitable format
- CYP with LD involved in making decisions about their care and treatment
- Views and interests of CYP with LD and their parents included in the planning, development and delivery of services
- Staff competent and confident to deliver safe, high quality care to CYP with LDs
- Appointments streamlined to minimise hospital visits
- Waiting time minimised
- Discharge plans started on admission
- Named nurse

Outcomes
- Particular needs of CYP with LD and families identified and met
- Reduction in adverse outcomes
- Reduction in seriousness of complaints
- Improvement in patient & parent satisfaction
- Improvement in patient & parent experience
- Increase in staff confidence & satisfaction
- Reduction in waiting times
- Reduction in numbers of hospital visits
- Reduction in financial cost to families
- Reduction in delayed discharge

Figure 2: Mixed Methods Strategy

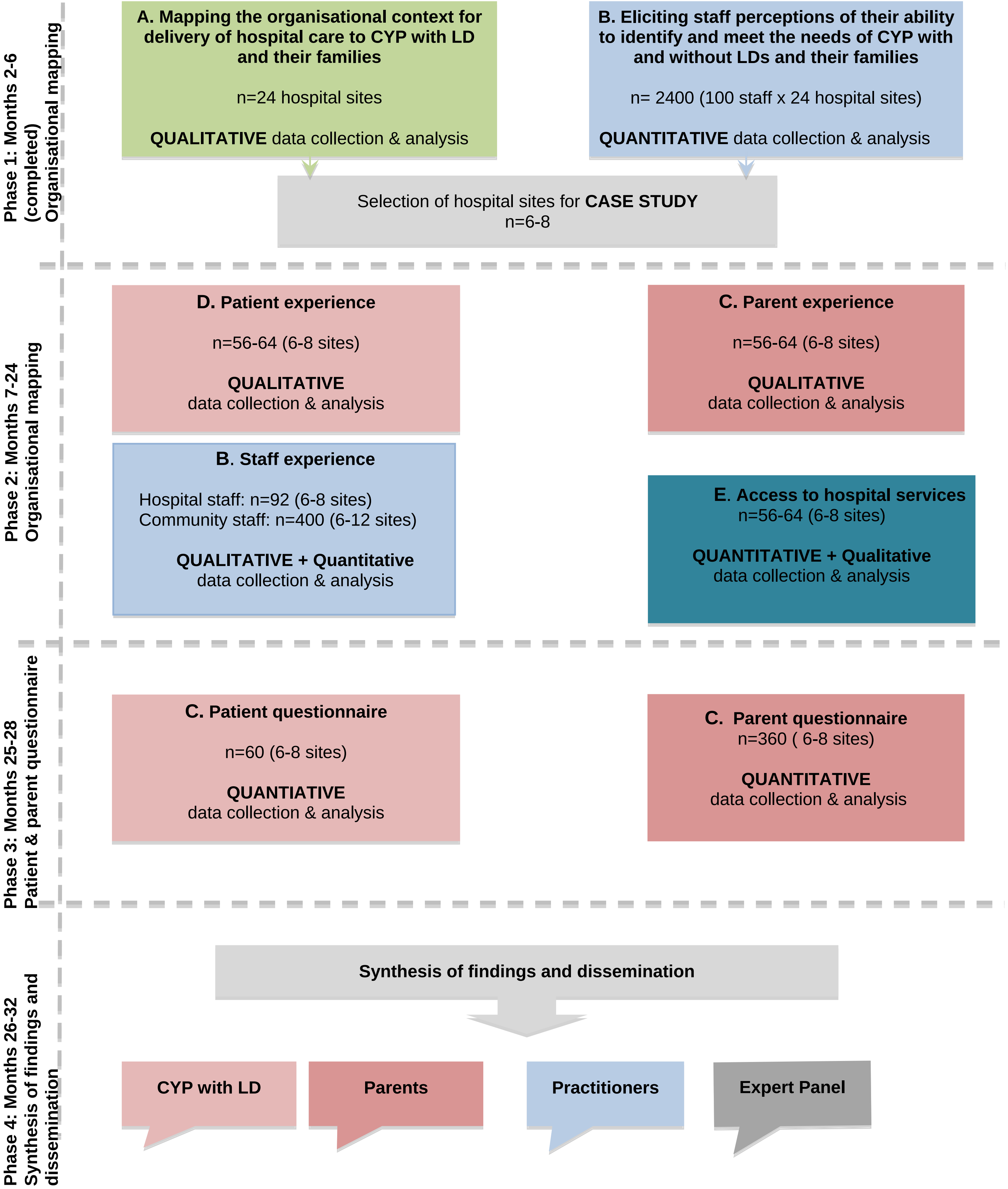
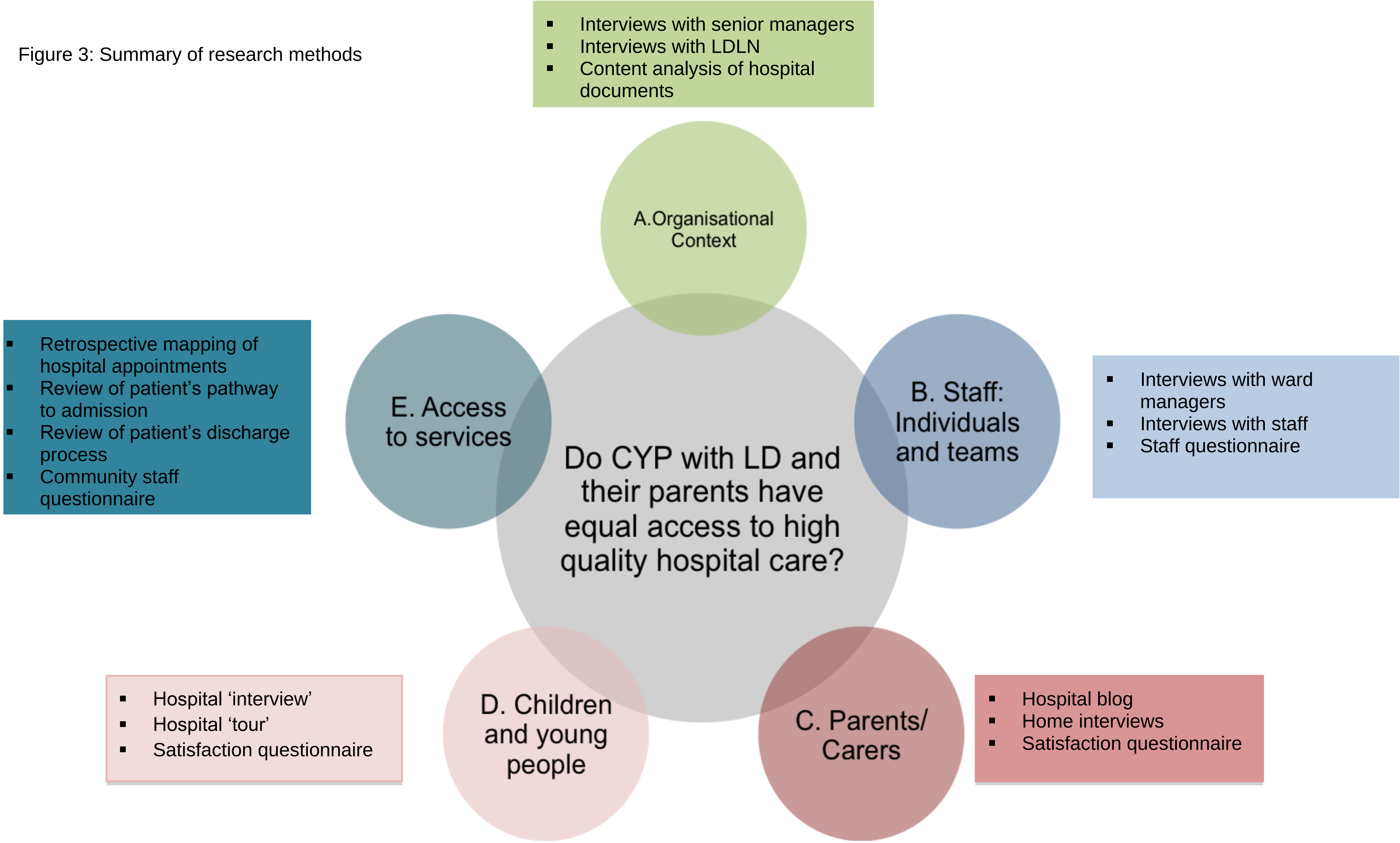


Figure 3: Summary of research methods



- Interviews with senior managers
- Interviews with LDLN
- Content analysis of hospital documents

- Interviews with ward managers
- Interviews with staff
- Staff questionnaire

- Hospital blog
- Home interviews
- Satisfaction questionnaire

- Hospital ‘interview’
- Hospital ‘tour’
- Satisfaction questionnaire

- Retrospective mapping of hospital appointments
- Review of patient’s pathway to admission
- Review of patient’s discharge process
- Community staff questionnaire

PHASE I: MONTHS 2-6 Completed

Aims

1) To describe the organisational context for healthcare delivery to CYP with LD and their families
2) To compare staff perceptions of their ability to identify the needs of CYP with and without LD and their families and provide high quality care to effectively meet these needs.

Setting

24 NHS hospitals in England.

Methods

Semi-structured interviews with senior managers and LDLN (Research questions 1-7)
The interviews focused on the delivery of services to CYP with LD at the organisational level.

Content analysis of hospital documents (Primary aim)
A search and find exercise using predefined terminology (learning disability, special needs, intellectual disability, developmental delay) was conducted to ascertain references to CYP with LD.

Staff questionnaire: (Research Questions 1-3)
All clinical and non-clinical staff with child patient contact at each hospital site were invited to complete a survey focusing on 6 key areas: staff knowledge, skills, training, confidence, time and resources.

PHASE 2: MONTHS 7-24

Aims

This Phase forms the main part of the study and has a number of aims:
1) To compare how hospital care and services are experienced by CYP with and without LD and their families.
2) To compare staff experiences of caring for CYP with and without LD and their families.
3) To investigate differences in safety concerns between parents of CYP with and without LD.
4) To investigate differences in access to hospital services between CYP with and without LD.

Setting

Participants will be recruited from 6 NHS hospitals in England. . Ward managers will be asked to nominate their ward as a site for data collection to increase staff ownership of the study and maximise success. Inclusion criteria to volunteer: the ward would be expected to admit 10 CYP with LD for a stay of **1-3 nights** within a 6 month period (allowing for a 50% recruitment rate).

Sampling

Participants	Sampling strategy	Sample size
CYP and Parents	-A purposive sampling strategy using a sampling matrix to ensure diversity according to level of LD, age, ethnicity	56-64 CYP
Hospital staff	-Ward manager on each study ward -Hospital staff identified by families as being involved in the care of CYP	12 ward managers 112-128 hospital staff
Community staff	-Community health and social care professionals directly involved in the care of CYP	400

Participants	Inclusion criteria	Exclusion criteria
CYP with LD	Aged 5-15 years (including 15 years and 364 days), Known LD (as defined above) Expected minimum in-patient stay of **1-3 nights**	Acute health problem only
CYP without LD	Aged 5-15 years (including 15 years and 364 days) Expected minimum in-patient stay of **1-3 nights**	Acute health problem only
CYP with and without Learning Disability - Only relevant for unplanned admissions where the consent will be taken in	Evidence of the CYP having unplanned admissions within the last two years in their hospital records	No evidence of unplanned admissions in hospital records

advance of admission)		
Parents	Is able to speak English	None
CYP with/out LD and Parents	No current complaints against the Trust or staff or of concern for safeguarding	'Children who are on the Child Protection Register or where the researchers would be at risk of harm by entering the family home'
Hospital staff	Is known to the family as being involved of the care of their child	None
Community staff	To have contact with CYP	Community Staff who do not have contact with CYP

Recruitment and consent

CYP and Parents

A small group of senior staff working on the ward and the local investigator at each site will identify eligible families from admission lists according to inclusion criteria. Eligible participants will be provided with the relevant parent and CYP information sheet in an accessible format, including a purposefully made ‘talking photo album’ (See Figure 3). Study information will be delivered in person or posted. A follow up phone call will be made by a member of the local team to check that the family have received the study information and if they are happy for their contact details to be passed to the research team. Alternatively, parents will be asked to contact the researcher if they are interested in taking part or have any questions. Once contact has been made, the researcher will explain to parents what would be expected of them and their child if they were to participate, including the voluntary nature of the study and the ability for them to withdraw without explanation. Confirmation will be sought that parents have discussed the project with their child. Families will only be recruited where parents and CYP both agree to take part. With permission, the researcher will meet participants prior to their involvement at a place of their choosing, likely to be out-patients or the family home. The CI’s previous experience of recruiting CYP with LD into research underlined the need to allow considerable time to become familiar with each CYP’s level of understanding and usual method of communication to enhance the assent process and ensure their involvement is valuable and meaningful. Once recruited, the researcher will arrange to meet families on the ward during their admission, where consent will be reconfirmed.

In addition to the planned and expected admissions described in IRAS. Input from PPI, the Study Steering Committee and meetings with the hospitals participating in Phases 2-4 have all emphasised the need to try to include unplanned admissions for CYP with and without LD. Two pathways to recruiting unplanned admissions will be established:

1) The first will be where sites are asked to identify CYP with and without LD who they expect will have an unplanned admissions e.g. directly to a ward or via A+E based on their medical history and recent hospital admission patterns. This group of parents will be sent a letter and information sheet sensitively explaining the study and why they have been asked to consider participating in the event that their child requires an unplanned admission within the data collection timeframe of Phase 2 of the study. A follow up phone call will be made by a member of the local team to check that the family have received the study information and if they are happy for their contact details to be passed to the research team. Alternatively, parents can directly contact the researcher via the information on the information sheet if they are interested in taking part or have any questions. Once contact has been made, the researcher will explain to parents what would be expected of them and their child if they were to participate, including the voluntary nature of the study and the ability for them to withdraw without explanation. Confirmation will be sought that parents have discussed the project with their child. Families will only be recruited where parents and CYP both agree to take part. With permission, the researcher will meet participants prior to their potential involvement at a place of their choosing, likely to be out-patients or the family home. Once assented/consented the researcher will ask the family to contact them, should their child have an unplanned admission during the course of the data collection period. As this is unlikely to be at the forefront of a parents mind, the local team will be informed of those who have consented to participate in the event of an unplanned admission and a local process will be established for the PI (or nominated other e.g. Research Nurse) to contact the researcher to inform them of the unplanned admission.
2) The second pathway will be where children who have unplanned admissions and their families are approached at the time of the admission. Their identification and eligibility would be established by the local team who would also make the initial approach about the study at an appropriate juncture in the child’s admission. Parents would be provided with an information sheet about the study and given at least 24 hours to consider if they would like to take part. The local team will then re-contact the family and ask if they are happy for a member of the research team to come to the hospital to see them. A member of the research team would

go to the hospital, meet with the family and CYP (dependent on their health) to discuss the study. Consent/assent would take place at the hospital with hospital based data collection with the child arranged for a time within the expected length of stay. We will only use this pathway when it is expected that the child will be admitted for at least 1 night. We have successfully used this approach before in a previous study.

Site-Specific: One site with few overnight inpatient stays.Justification and inclusion: At one participating site their approach is for the hospital and community to work closely together to limit the need for CYP to have overnight stays in hospitals. This has resulted in few CYP meeting the eligibility criteria of a minimum 1-night stay in hospital. To collect data from this site and to understand what it means for CYP and families *not* to need to stay in hospital overnight an alternative approach to data collection is required. For this one specific site, CYP will not be required to have an overnight admission rather data will be collected from day cases admitted to the participating wards. These CYP will have procedures where it is likely at other participating PMA sites the CYP would normally require an overnight inpatient stay. All CYP and families will need to meet the remaining inclusion criteria: aged between 5 and 15 years and 364 days, have a long term condition and for 8 CYP participants to have a learning disability (out of 16 CYP participants at the site).

Matching: Attempts will be made to 'match' patients with and without LD. However, with this alternative approach the focus will be on collecting child and parent data about their experience at this site.

The researchers will plan to be on site over the course of several days to be agreed in advance based on admission dates.

Data collection: A time for CYP data collection will be agreed with the family with the aim being for at least one CYP activity to be completed by the CYP (or parent proxy). No hospital based parent data will be collected at this site.

Post-discharge: The CYP's parent/carer will be contacted and invited to take part in the parent interview.

Staff interviews: In addition to parents identifying staff, staff working on the participating wards will also be invited to participate in an interview.

Data collected from this site will form a unique case as direct comparisons with the data from the other participating sites will not be feasible.

Figure 3: Resources for assenting CYP

Hospital staff
Staff will be provided with a study information leaflet and invited to take part in an interview. They will be informed that they have been selected because they are involved in the care of a child participating in the study. They will be provided with the researcher's contact details and asked to make contact for more information or to take part. The researcher will meet them at their convenience where written informed consent will be taken.
Community based staff
Community staff from Community Trusts who in their professional role have contact with children and young people that are in close geographical proximity to the Phase 2 participating Hospital Trusts will be sent information about the study (n= minimum of 6 Community Trusts). This will be circulated by a local collaborator at each site via email with a link to an online questionnaire which will be completed and submitted anonymously. Paper versions of the questionnaire can be provided if requested for staff who may have limited access to a computer. The questionnaire will start with a brief summary of the study and will have contact details for the Chief Investigator and Project Manager. The questionnaire will focus on staff perspectives towards caring for CYP with and without LD. Completion of the questionnaire will be taken as consent to participate. This survey is the only part of Phase 2 in which Community

Trusts and their eligible staff will be invited to participate. A separate Statement of Activities and Statement of Events will be provided for Community Trusts for this aspect of the study.

Methods

- **To meet the aim of comparing how hospital care and services are experienced by CYP with and without LD and their families the following methods will be used:**

Interviews with CYP: (Research Questions 1-3)
The Mosaic approach[58,59], combining the "traditional methodology of observation and interviewing with the introduction of participatory tools"[58] will be used to guide interviews with CYP. This multi model approach to data collection is underpinned by a view that CYP are experts on their own lives. The aim is to have a toolkit of creative and digital techniques available that draw on each individual's strengths, thereby enabling them to share their experience and preferences in whatever ways they are able and comfortable with. As Davis et al.[60] suggests, "it is unlikely that any one research activity or tool will be accessible to all young children with different skills, cultural backgrounds and personalities" and this is even more so for CYP with LD.

The primary method of data collection will be 'Talking Mats', a communication symbols tool based on extensive research and designed by Speech and Language Therapists. It consists of a pictorial framework based on 3 sets of picture symbols **-** **issues** relevant to the topic, **factors** relating to each issue and **emotions** to allow participants to indicate feelings about each factor. Talking Mats has been shown to be a powerful tool to allow CYP with different degrees of LD to communicate their views. In a study about making choices at the time of transition for YP with LD, Cameron and Murphy[61] found that participants were able to indicate their likes/dislikes and express views about choices available to them. Some YP with LD raised sensitive issues about transition and revealed opinions that were previously unknown to those caring for them. The method is suitable for CYP of all ages and communication abilities and can therefore be offered to all participants irrespective of whether they have a LD.

Arts-based activities, photography and hospital tour[62-65] are other ways that CYP will be able to share their views. The CI has previously used arts based activities to help CYP with LD share their hospital experience[50]. A 3rd person craft activity enabled them to project their fears and concerns, as well as what they found helpful; choice boxes enabled them to share preferences and a simple questionnaire enabled them to give their opinion (See Figure 4).

Figure 4: Possible methods for collecting data with CYP with LD

Where appropriate, CYP will be offered the opportunity to take the researcher on a 'tour' of the hospital, identifying areas they have accessed during their admission and what those spaces mean to them. They will be offered a digital camera to use during the tour to take photographs of people, pleases and objects of importance to them. Photography has been used in research with CYP with and without LD as a way of giving them control over what is talked about and as a visual prompt for discussion[65]. In a project involving CYP with LD, Cook and Hess[66] reported that the method revealed more complex and in-depth conceptualisations than they had imagined, introduced unexpected topics and offered new suggestions as to what might be meaningful for children in certain situations. Through a process of photo-elicitation, the photographs will be printed and used to facilitate discussion with the researcher about who, what and where is important to them during their hospitalisation. Photo-elicitation is a well-established research method that that uses images to prompt and guide discussion. It allows participants to give meaning to the pictures they take and explain these, thereby giving them an element of control over the process. It is recognised as being non-threatening[11] and as Whiting (2015) suggests the method "can be a positive experience for children, and one that is not only fun and engaging, but that is also empowering and valuing of their contributions". The use of digital photography is therefore particularly useful in a context in which we want to hear from a group of vulnerable children whose opinions about hospital are seldom heard. Moreover, some CYP with LD, such as those with Autism, relate particularly well to photographs and may prefer this method of communication above all others.

Interview questions that specifically focus on the interactions that CYP have with staff and that are not addressed via Talking Mats or hospital tour methods will be asked using a questionnaire style form. CYP will be invited to complete this form to indicate their experience during their admission. The questions listed below will be asked to each participant. However, the exact format will be tailored to the ability of the CYP ranging from a self-complete questionnaire style form through to the researcher reading out the text and asking the CYP to indicate their response.

Depending on the ability of the child CYP they will be asked to indicate either whether this is something that they experience (yes/no), the regularity that this happens (always, sometimes, never) and whether this, from their perspective, is 'a good thing' or 'a bad thing'.

The skill throughout Phase 2 will be on tailoring data collection activities to each individual CYP, whilst ensuring the research questions are addressed. As Mason[67] said in relation to CYP with LD*, "Listening can mean going for walk with someone and noticing what captures their interest. It can mean learning to recognise situations in which a person becomes upset, or becomes animated; it can mean watching a person's movements, or the activities they choose over others".* Our flexible, multi-method approach to data collection is crucial to ensuring that CYP of all ages and abilities are truly listened to.

Data collection sessions will take place in a quiet room on or close to the ward (apart from the hospital tour), depending on each CYP's personal preference and health needs. Some CYP, including those with LD, may find it difficult concentrating for long periods of time and in these circumstances a few short sessions may be preferred to one longer session. The researcher will be guided by CYP and parents as to what would be most appropriate. CYP's preference for their parent(s) to be present or absent during the sessions will be respected.

If children are too unwell to complete all data collection activities during their admission, children (with agreement of their parent(s)) will be given the opportunity to complete these at home after they have been discharged from hospital. Where a CYP/parent have been consented for the study but discharged from hospital earlier than their expected discharge dated, it means that data collection cannot start during the inpatient stay. In this specific circumstance where the CYP is known to have sufficient recall the CYP/parent will be given the opportunity for the CYP data collection to take place at the CYP's home address within one week of discharge.
KO, FG, CJ, SK will provide expertise for this part of the study.

Parent Diary: (Research Questions 1-3)
Parents will be given an android 'tablet' (password protected and security tagged) and invited to complete a hospital diary during their child's in-patient admission using pre-installed applications with templates for written diary entries or a verbal audio diary can be kept on the 'tablet'. . Parents will be encouraged to document their thoughts and feelings in relation to key events during their hospital stay such as admission, discharge and their child's investigations and treatments. Parents will not be sharing their thoughts/feelings with the research team in in real time. Parents will have the option of a paper copy of the diary as an alternative to the 'tablet' and where a child has two parents; both will be invited to make entries into the diary.

The CI's previous experience revealed some parent's reluctance to leave their child to be interviewed, even for short periods. Diaries offer parents flexibility in how they tell their story and can be completed at any time of the day/night. The researcher will spend time showing parents how to use the tablet and applications and will provide a simple crib sheet and FAQ page to refer to. They will also 'check-in' with parents to answer their questions and resolve any technical issues. . Importantly, the content of the parent's diary will be used to facilitate discussion during a follow-up home interview (see below) that has been designed as the primary method for answering the research questions. By incorporating the use of novel, digital research methods we aim to give parents flexibility and enhance the findings through the capturing of 'live data', but the success of the study is not dependent on its use.
CJ will provide expertise for this part of the study

Home interviews with parents: (Research Questions 1-3, 5)
Home interviews will be conducted with parents as soon as possible after the CYP's discharge from hospital, preferably once they have returned to school. The CI previously found home interviews to be more conducive to the collection of rich data. Parents appeared less distracted and more at ease in their own environment and conversations were interrupted less frequently than in the hospital setting. Moreover, conducting home interviews with parents will free the researcher to focus on working closely with CYP during the hospital admission. The CI found that collecting data with parents and patients during the same hospital admission had challenges, sometimes resulting in the voice of CYP being overshadowed. The interview guide will focus on parents' experience of accessing and using hospital care and services for themselves and their child. Data recorded on the parent diary will be used as a further prompt. Questions about the child's pathway to admission and their experience of discharge will be included. Parents will also be asked map the health and social care hospital staff who are or have been involved in the care of their child. This information will be used to decide which staff will be invited for interview. Parent interviews are expected to last one-two hours.

KO, FG, SK will provide expertise for this part of the study

- **To meet the aim of comparing staff experiences of caring for CYP with and without LD and their families the following methods will be used:**

Interviews with hospital staff: (Research Questions 1-3, 5,7)
Interviews with hospital staff will be face-to-face or telephone and semi-structured. They are expected to last 30-60 minutes. Flexibility will be provided as to the timing and location to minimise staff burden. The CI has experience of interviewing hospital staff about their experience of caring for CYP with LD and received a positive response to requests to participate.
KO, FG, ITW, MW, LC will provide expertise for this part of the study

- **To meet the aim of investigating the differences in safety concerns between parents of CYP with and without LD the following methods will be used:**

Completion of the 'Parent Safety Review Form': (Research Question 5)
In light of qualitative evidence that parents of CYP with LD can lack confidence that their child is receiving high quality hospital care and subsequently feel responsible for monitoring their care, parents will be asked to complete the daily safety reporting tool[68]– a six item tool which asks parents to identify their safety concerns from a choice of: medication, communication and information, equipment, unexpected complications of care, hygiene/cleanliness, other safety problems. Completion of the tool will enable perceptions of safety between the two groups of parents to be compared. Information collected will be used as a prompt during home interviews.
LC will provide expertise for this part of the study.

- **To meet the aim of investigating the differences in access to hospital services for CYP with LD, compared with CYP without LD the following methods will be used:**

Retrospective mapping of hospital appointments: (Research Question 2)
For each CYP and with parental consent, a retrospective mapping will be conducted of all in-patient stays and out-patient appointments for the previous 2 years using the electronic hospital appointment system and/or patient records to the researchers with assistance from the site Principal Investigators will retrieve the following data:
In-patient stay – a) age of patient b) diagnosis c) date of admission d) ward e) treating team f) reason for admission g) anticipated date of discharge h) date of discharge i) discharge location j) A+E admissions.
Out-patient appointments – a) age of patient b) diagnosis c) date of appointment d) time of appointment e) admitting team f) 'Did not attend' status and reason given.
Evidence of any of the following documented during the admission for P2 CYP/Parent data collection – a) hospital passport used (or similar document) b) Long term condition c) Learning Disability d) Reasonable adjustments needed/made e) Communication needs/method f) Level of understanding. A note will be made of where this information was recorded e.g. in medical or nursing notes.

After the first set of paired notes have been reviewed at each site, we will ask the SSC to determine whether the data collected is sufficient and any practical considerations such as time taken to collect the data.
JW will provide expertise for this part of the study.

Questionnaires by community-based professionals: (Research Questions 1-3)
Community Trusts in close geographical proximity to the participating Hospital Trusts will become research sites for this aspect of the study. Community professionals with contact with CYP in these Trusts will be invited to complete the survey. The questionnaire will be a modified version of the hospital staff questionnaire from Phase 1 with a particular focus on access to secondary and tertiary care for CYP with and without LD. The survey will be available via an online link with paper copies available for staff with limited access to a computer at work. A Local Collaborator will be identified to assist with the dissemination of the survey to relevant staff and to encourage participation in the survey.

JW and ITW will provide expertise for this part of the study.

PHASE 3: MONTHS 25-28

Aims

To compare levels of satisfaction with hospital care between: a) CYP with and without LD, b) Parents of CYP with and without LD.

Setting and sample

All parents and patients who are discharged from the study wards at each of the 6-8 hospital sites from Phase 2 for a period of 3 months will be eligible for participation. Respondents will be asked to tick whether they/their child has a) LD, b) a long-term condition, c) neither and d) both. A total sample size of 360 parents and 60 CYP has been calculated based on a conservative estimated return rate of 30 questionnaires per hospital per month for parents and 5 questionnaires per hospital per month for CYP. These estimates are based on the research team's experience of collecting this type of data in a hospital setting.

Method
Questionnaire: (Research Question 4)
There is a lack of validated patient/parent satisfaction questionnaires, particularly for CYP and those with LD. Drawing on the best available tools (http://www.chimat.org.uk/default.aspx) a questionnaire will be purposefully designed to answer the research question. Multiple versions of the questionnaire will be developed for CYP across the age range and with differing levels of cognitive functioning. Questionnaires will be piloted with a group of CYP with and without LD and their parents beforehand.

Questionnaires will be given to parents and CYP prior to discharge. Parents will be asked to indicate on the form if their child has a) LD b) a long-term health condition c) both d) neither (Definition of long-term health condition and LD will be provided on the form). A sealed box will be available on the ward for participants to leave their completed questionnaire prior to discharge.
JW, ITW will provide expertise for this part of the study.
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Data analysis
A model for mixed methods data analysis[1] will be used. Qualitative and quantitative data will be analysed within each phase using appropriate methods before merging and connecting them through a period of data synthesis.

Qualitative
A modified version of grounded theory will aim to be used for the majority of the qualitative data analysis, particularly interview data that generates transcripts. This approach uses an inductive approach and intends to generate theory from observations of real life. The strength of this method is that it teases out the depths of an experience and locates this within the wider context of human experience. There have been a number of developments in the application of grounded theory and it is intended to draw upon the Charmaz's (2014) constructivist approach

Quantitative
Separate quantitative analyses will analyse:
(i) Hospital staff questionnaire data (Phase 1). Analyses will follow previous studies of staff questionnaires of patients with LD in hospitals[18]. Descriptive comparisons for each of the 6 key areas of the questionnaire between responses pertaining to CYP with and without LD will be presented (e.g., frequencies, percentages, means and standard deviations, medians and inter-quartile ranges). Comparisons will also be presented for sub-groups of respondents categorised by staff group (e.g., doctors, nurses, professions allied to medicine, non-clinical staff), staff grade and site.
(ii) 'Safety concerns' data using the daily safety reporting tool (Phase 2). Number and type of safety concerns will be compared and analysed descriptively.
(iii) Community-based staff questionnaire (Phase 2). Responses to this questionnaire will be compared descriptively and analysed in the same way as for the hospital staff data, described above in (i).
(iv) Parent and patient satisfaction with hospital care (Phase 3). Responses to this questionnaire will be compared descriptively and analysed in the same way as for the hospital staff data, described above in (i).
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PHASE 4: MONTHS 26-32

Aim
To synthesise and disseminate findings and develop the content of a DVD and/or training package for students/staff

Method
The amount of qualitative and quantitative data generated by the study will be large. During the stage of data synthesis, the research team will use quantitative data to explain and illustrate qualitative findings, and look for congruence and incongruence between qualitative and quantitative findings. In particular, the team will look for instances where there is incongruence between policy and practice, using specific queries within the NVivo programme to address these issues and explain any incongruence. It is at the stage of data synthesis that barriers and facilitators to ensuring CYP with LD and their families receive equal access to high quality hospital care and services, will be highlighted, looking for specific examples of successful and effective measures that promote equal

access. The final analytical framework will be compared with our Theoretical Framework and the initial common analytical framework, in order to generate a final Empirical Framework of factors that affect the promoting of equal access to high quality hospital care for CYP with LD and their families.

Two workshops will be held towards the end of the study with parents and professionals. One will be for parents of CYP with LD and the other for parent of CYP without LD. The workshops will be held at Great Ormond Street Hospital in London. The workshops will be led by the CI who will be supported by members of the Core and Executive study teams. In addition members of the Study Steering Committee will be invited to attend and participate in their professional capacities.

The aim of these workshops will be to disseminate findings back to participants and professionals to promote discussion regarding the content of a DVD and/or training package that will be used in practice to guide students and staff of the barriers and facilitators to the delivery of high quality care for CYP with LD and their families. **It is only the content of this training package that will be decided during the project duration. Additional funding will be sought elsewhere for its development and implementation.** In the adult setting, a similar training initiative has been used to ensure staff have a good understanding of the issues faced by patients with dementia so they can recognise and support care for these vulnerable patients. So far, 12,500 staff at Guys and St Thomas' hospital have attended a training session, the main focus of which is a short film about Barbara and her experiences during a hospital visit.

The discussions will produce rich data in the form of ideas to improve CYP care and the workshops and will be used as an additional data collection opportunity. PPI advice has been sought from the Pay More Attention Parents' Advisory Group and integrated into the design and conduct of the workshops. Phase 4 Participant Information Sheets and consent forms have been developed for parents and professionals.

The workshops will be run in the style of a 'World Café' (www.theworldcafe.com), which is a flexible method for large group discussion and modified to meet the aims of Phase 4. The World Café encourages interaction between participants based on the environment of a café with multiple tables.

At the start a facilitator will outline the World Café process, aims and etiquette for the discussions. The facilitator will oversee the 'World Café', keep to timings and at the end summarise the workshop. The specific issues and questions for the discussions will be developed from the findings of Phase 1-3 of the Pay More Attention study. Parents and professional participants will sit at tables in small groups of 4-5 people with a host and discuss a specified issue for a set period of time; this may be through direct conversation or by using a creative method to stimulate discussion. At the end of this time the participants move to different tables to discuss another issue thus allowing interaction with a range of participants and their views and opinions. New participants arriving at a table will be provided with a summary of the previous discussion by the host, who will then encourage the new participants to build on this conversation adding a layer of greater depth so the content of the progressive discussion builds over time as participants move between tables. At the end each host will provide a brief feedback about their tables' discussions to the wider group. Each workshop will last 2-3 hours.

For the purposes of the study the discussions will audio recorded, with the consent of the participants, to capture the richness and more nuanced aspects of the discussion, which may not be covered in the brief feedback. The recordings will be deleted at the end of the study. Consent will also be sought from participants for photographs to be taken of the World Café in action to be used in the presentation and dissemination of the findings.

Participant Recruitment: Parents

We will invite parents who participated in Phase 2 of the study to the relevant workshop. As Phase 2 participants, at the time of their original participation, did not consent to be re-contacted, each of the Phase 2-4 sites will be asked to forward a letter of invitation including an information sheet and a request for parents to contact the research team at GOSH if they would like to participate. After this point all contact for those who would like to participate will be with the GOSH team.

Additional parents of CYP with and without LD with experience of having stayed in hospital who attend schools local to GOSH will be identified and invited to take part in the workshops.

Participant Recruitment: Professionals

We will invite professionals who participated in Phase 2 and those who worked on the wards where Phase 2 and Phase 3 ran to take part in the workshops. This will include ward staff as well as allied health professionals who work with inpatients. We will ask each site to forward the Phase 4 staff information via email and request that those wishing to take part contact the research team at GOSH if they would like to take part in a workshop.

We will directly invite members of the Pay More Attention Study Steering Group to take part in the workshops in their professional capacities.

As Pay More Attention is a national study parents and professional participants will be reimbursed for subsistence and standard return travels costs between their home and GOSH.

FG, ITW, MW and CJ will provide expertise for this part of the study.

Patient and public involvement
This study has directly evolved from 4 substantial pieces of work involving the CI and/or other co-applicants to understand: 1) how to manage pain in non-verbal children with LD, 2) parental perspectives of caring for a child with LD, 3) the needs and experiences of CYP with LD, their families and staff in hospital, 4) how to create a safe hospital environment for adults with LD. Key to each of these was hearing what matters most to participants. We believe strongly that it is crucial to involve patients and public in all aspects of the research process, particularly when the research involves marginalised groups of people, such as those with LD, whose views are rarely heard. One of the co-applicants is a parent (SK) with nearly 20 years' experience of using NHS hospitals within and outside London for her children with LD. SK has previously worked with the CI on a related LD project. Our aim is to ensure that any data collection involving parents is carried out in an appropriate, accessible, sensitive and ethical manner. SK will play a crucial role in advising on data collection so that methods are acceptable to parents and likely to be completed.

Parents, hospital and community based clinicians and LD experts have been consulted about the research questions, design, methods and dissemination to ensure they are acceptable, feasible and of importance. Parents identified the need to consider raised parental stress, illness of child, possibility of parents having a LD and benefit of collecting data online, using a LD nurse to support CYP involvement in the project and collecting data from multiple hospital sites. 3 parents of CYP with LD and 3 parents of CYP without LD have agreed to sit on the Parents Advisory Group (PAG). We will offer relevant training in the form of a workshop to all parents who are part of this group. This will include a session on roles and responsibilities, providing parents with clear guidance of what is expected of them and who to contact with any questions or concerns. Two members of the research team with particular expertise in PPI, FG and MW, will act as mentors to parents, acting as a point of contact for them throughout the study and providing relevant advice and support as needed. We will also offer SK the opportunity to attend an external training session on qualitative data analysis in preparation for her involvement in this element of the project.

A CYP Advisory Group will be established via local schools and we will also utilise existing young people's groups at GOSH to act as reference groups. Members of the research team will work with these groups of young people who will be asked to trial data collection activities, review and discuss information materials, ethical issues and study designs to ensure they are appropriate for young people. The researchers will meet with the CYP in schools as this is an environment with which they are familiar.
MW, FG will provide expertise in PPI

Outputs and dissemination
There will be at least one output from each part of the study, led by different team members with relevant expertise:

What	When
Phase 1:Comprehensive mapping of the systems and structures in place for the delivery of hospital care to CYP with LD and their families across 24 hospitals in England	December 2016
Phase 1: Comparison of staff's perception of their ability to meet the needs of CYP with and without LD and their families	December 2016
The hospital experience of CYP with and without LD	November 2018
The hospital experience of parents of CYP with and without LD	November 2018
Hospital staff's experience of caring for CYP with and without LD	November 2018
Community staff's perception of hospital access for CYP with and without LD	November 2018
In-patient and out-patient activity for CYP with and without LD	November 2018
Comparison of safety concerns of parents of CYP with and without LD	November 2018
Patient satisfaction with hospital care and services	November 2018
Parent satisfaction with hospital care and services	November 2018

The primary output will be guidance for commissioners and providers of NHS hospital services for CYP with LD and their families. Other outputs include:

- Report to CYP/parent participants in a range of accessible formats, accounting for the needs of those with LD.
- On-line report for lay audiences, including those with LD, at GOSH and Contact a Family, BILD,
- Presentation back to staff participants at each Phase 2 site to share examples of good practice
- Published abstracts and papers in international peer-reviewed journals including an e-journal for healthcare students and professionals 'www.intellectualdisability.info' which is accessed by more than 500 people daily
- Presentations through national and international conferences, including those that reach different professional groups such as nurses (Royal College of Nursing International Research Conference), doctors (Royal College of Paediatrics and Child Health) and Allied Health Professions (Royal College of Speech and Language Therapists) as well as the International Association of Scientific Study of Intellectual Disabilities (IASSIDD).
- Separate funding will be sought, from a 3rd sector organisation, for the making of the DVD and disseminating it widely into practice. The aim is to replicate the impact that Barbara's Story has had at Guys and St Thomas' Hospital in changing care and attitudes towards patients with dementia. 12,500 staff have currently seen the film and been trained. Once made the training DVD will also be available for trained staff. **http://www.youtube.com/watch?v=DtA2sMAjU_Y&feature=share&list=UUBJBH2MFKrX6lf8bJ7_ZGW Q**

Working in partnership with the Foundation for People with Learning Disabilities, the British Institute of Learning Disabilities, Contact a Family and NHS England will strengthen our dissemination strategy, ensuring the findings are shared in a variety of accessible formats and reach a wide range of parents, professionals and CYP with and without LD, as well as academics and policy makers helping drive support for change. Through inclusive dissemination this research will bridge the gap between NHS management and users.

We will develop a social media strategy to ensure ongoing dissemination of findings and user engagement throughout the project, and to build a network/community of interested users/stakeholders. We will set up a project website, blog, and twitter for the project. This will enable key research narratives to be disseminated and will provide a creative way to engage directly with potential users and testing ideas and contributing to relevant debates as they evolve. The web site will be maintained by the research team and will be a 'read only' site. The research team will provide an accessibly written project blog entry on a key issue related to the project on a regular basis. Only the project team will be able to post information. A project dedicated email address will enable people to directly contact the research team. Twitter will be used as necessary to connect with major stakeholders, policy makers and key players about ongoing debates and ideas. All participants will be provided with links to the study's social media presence.

Expected Impact

The NHS is striving to improve quality of care while also increasing the efficiency of resource use. This study has the capacity to meet both these goals through identifying inequalities in hospital care for CYP with LD and their parents and determining how resources and interventions that enable equality can be targeted more effectively to the right patients at the right time and be reconfigured to ensure care is preventative rather than reactive, thereby increasing efficiency. The guidance produced will enable commissioners and service providers to improve the way that hospital services are organised and delivered to CYP with and without LD and their families ensuring equality for all. There is potential to see improvements in staff awareness of what matters to these CYP and their families as well as their ability and confidence to identify and meet their needs, leading to improved satisfaction for everyone.

Through identifying inequality and offering strategies to eradicate it, there is the potential to see a reduction in:

1) Financial costs to the NHS and families through the delivery of a more efficient and effective service
2) Health costs to CYP and parents through better understanding of their physical needs and focus on risk prevention, leading to improved quality of care
3) Emotional costs to CYP and parents as a result of reduced stress and greater confidence in care
4) Social costs to CYP and parents through reduced hospital contact

It is key that we make findings accessible and they are widely disseminated so that the relevant people are given the opportunity to 'take notice' of them and respond.

Failure to identify inequalities in hospital care for adults with LD has led to preventable deaths and damage to the reputation of the NHS, as well as on-going costs from reviews and inquiries. Through identifying inequalities in access to high quality hospital care for CYP with LD and what action is needed to remove them, these extensive human and financial costs can be avoided, thereby making the proposed study extremely good value for money.

Success Criteria (Phases 2-4)

1. 100% of CYP with LD will be matched with a comparator case
2. 50% of parents complete a hospital diary
3. 75% of parents complete the daily safety reporting tool

4. 100% of parents complete the home interview
5. 40% response rate of the parent satisfaction questionnaire
6. Workshops will have representatives from each of the four stakeholder groups.

Research Management

Dr Kate Oulton (KO) is the CI and will act as project manage. KO has experience of project managing a 4 site randomised controlled trial, including supporting clinical nurses to recruit and deliver an intervention to parents in the neonatal intensive care setting. KO is currently leading or collaborating on a number of projects, each one involving CYP and/or their families in the hospital setting. She has recently led and completed a study to understand the needs and experiences of CYP with LD in hospital, the results of which have reinforced the need for the proposed research. KO is a Visiting Fellow at South Bank University and is linked to the Department of Children's Nursing Team. She also has an NIHR funded post as the clinical academic programme lead for nursing and allied health professions research at Great Ormond Street Hospital NHS Foundation Trust (GOSH). KO will provide day to day supervision for the research team and lead regular team meetings, assist with day to day management of the project as needed, monitor progress, guide data analysis, and communicate with the joint applicants. All research management will be based at GOSH; researchers will travel to the research sites for data collection as needed.

In addition to being a core member of the research team (see below) Prof Gibson (FG) will provide mentoring support for KO through the provision of weekly face to face 'meetings' which provide regular opportunity to listen, share experiences, offer support and encouragement, as well as provide a framework to explore options for career development; including identifying and supporting opportunities for teaching and training and drawing on existing networks to maximise opportunities for KO to reach out into the research community in child health. Mentoring in terms of a research career already features in the relationship between the CI and FG, both are based at GOSH, thus regular contact is possible. FG currently mentors a number of nurse academics, and offers at a distance guidance to two NIHR post doctoral fellows. It is this process of getting information to people, through people that will lead to changes in practice. Professor David Richards, a nurse and Professor of Mental Health Services Research at University of Exeter Medical School, will provide additional mentoring to KO externally. Prof Richards is CI on NIHR-HTA and MRC/EME funded projects, a NIHR Senior Investigator and the NIHR Academic Training Advocate for Nursing. He will be an excellent role-model for KO and will support her leadership development.

The research team comprises individuals from different professional backgrounds that have extensive experience of researching the field of child and adult health and significant expertise in the delivery of time-limited projects, involving hard-to-reach participants. As a team we are aware of the challenges and burdens for families for participating in research and have the necessary skills to minimise and manage these. The core research team and researchers will meet weekly to discuss and address current issues and progress. In all cases, face to face meetings will be encouraged where there is no significant time or travel cost, but teleconferencing will be used to minimise time and funding required. The wider research team will meet monthly to oversee the study at an operational level.

Name	Professional background	Research expertise	Primary population
Dr Oulton (KO)	Paediatric nurse	Qualitative research methods	CYP with LD
Prof Gibson (FG)	Paediatric nurse	Qualitative research methods/ case study design	CYP
Dr Whiting(MW)	Paediatric nurse	Patient and public involvement	CYP Complex long term health conditions
Dr Wray (JW)	Health psychologist	Quantitative research methods	CYP
Dr Tuffrey-Wijne (ITW)	Adult nurse	Health services delivery research	Adults with LD
Dr Hassiotis (AH)	Consultant psychiatrist	Applied health services research	LD/mental health
Dr Carr (LC)	Consultant neurologist	Expert clinician	CYP with epilepsy
Prof Jewitt (CJ)	Education and technology	Digital research methods	CYP
Mrs Kerry (SK)	Expert Parent	Expert parent	CYP with LD

A SSC has been set up to provide overall supervision and ensure the project is conducted to the standards set out in the "Department of Health's Research Governance Framework for Health and Social Care and the Guidelines for Good Clinical Practice" (NIHR). The SSC is necessarily large, as this study involves multiple research sites and strands, requiring wide-ranging expertise and viewpoints from a range of academics, clinicians and lay people.

Ethical issues

This study includes data collection involving vulnerable CYP. The research team has longstanding expertise in conducting research in sensitive areas. The research team feels strongly that ethical considerations for this study need to be given attention above and beyond any requirements of Research Ethics Committees. Therefore, a range of steps will be taken in order to safe guard all informants from undue harm in accordance with the principal of beneficence. We will pay particular attention to obtaining assent from research participants with LD, using a range of accessible study information materials, including a talking photo album, and ensuring sensitivity to the various ways in which people with LD may express withdrawal of consent. Ethical approval for the study will be obtained via IRAS. . We propose to commence the study in July 2016.

Summary of study risks

Potential risk	**Action to limit or resolve risk**
Delayed approval from NHS Ethics committee	We are well aware that our application may present to the ethics committee challenges in relation to the population and research methods. We will ensure clarity and offer fuller explanations to assist the members to fully understand: • The need for the research • The sample and sampling framework used • The need to be flexible in our approach with CYP that will best reveal a yes or no to participation • The range of methods of data collection used with CYP and evidence of our expertise in using them • The challenges associated with using digital photography in a hospital setting and evidence of our expertise in this area • What the burden of being in a study can mean to families, our aim will be to sensitive to this and keep it to a minimum • Our approach to PPI that ensures we have a robust and achievable study • Our skills in managing large and complex data sets • Our relationships within the child healthcare settings that enable multi-site working
Protracted period for local R&D approvals	Our experience to date would suggest we might be delayed, similar to NHS ethics, as we are presenting local approval teams with a challenge in terms of population and data collection. We have experience of multiple conversations to provide additional information about the creative approaches to data collection we use with CYP to R&D departments. Similar to ethics we will ensure fuller descriptions of our work in the Statement of Activities, making clear the role of local PI, and their role in identifying families, approaching and recruiting. The role of their staff, and time to be given on the study will be made very clear: anticipating questions in advance based on our experience to date in multi-site complex studies like this.
Low recruitment rate: CYP	-Experienced network of parents and professionals on SSC -Parent of CYP with LD as a co-applicant -Small sample size for Case Study during Phase 2 -Mixed methods allows other sources of data to add to the voice of CYP -Costs included for local investigator time at each site -Costs included to allow for suitably trained researchers -Tailored assent information and recruitment materials -Home visit to facilitate rapport and understand CYP's individual needs -Ongoing feedback to sites through project Newsletter and support for local investigators
Low recruitment rate: staff	Telephone interviews will be offered as an alternative to face-to-face interviews
Identifying CYP with LD	A broad definition of LD will maximise potential success of recruiting families and minimise risk of patient identification problems disrupting recruitment or study processes. Accommodating and including families where the child has an uncertain diagnosis, which by definition may change over time, is an important aspect of the inclusivity of this study.
Site reluctance to include 'Parent Safety Review tool'	We will liaise closely with study sites to ensure they have complete understanding of the reasons for including the tool and how we will use the data collected. Maintaining confidentiality of study sites will be a crucial element of the study.
Responding to patient safety concerns	As part of the ethics approval process, the research team, SSC and local investigators will agree an appropriate plan of action in response to the

	retrospective parent's reporting of safety concerns and the research team's reporting of these to the relevant hospital. Parents will be informed that any concerns should be reported directly to staff at the hospital. .
Maintaining patient and site confidentiality	Information pertaining to participants and sites will be kept strictly confidential and used solely for the purpose of the study. Strict coding procedures will be adhered to at all times.
Dissemination of findings	We are well aware of the complexities of disseminating to families and professionals sensitive information. We are mindful of published work that offers suggestions about how to approach this sensitively, whilst still ensuring we inform study participants about what we found out. Having a parent of children with LD on the research team will help us here. In addition, the research team have significant experience in producing dissemination materials, the development of the DVD will ensure we reach as many people as need to know.

References

1. J.W. Creswell, V.L. Plano Clark. Designing and Conducting Mixed Methods Research. 2nd ed. London: Sage; 2010.
2. Department of Health. Valuing People: A New Strategy for Learning Disability for the 21st Century. London: DH, 2001.
3. World Health Organisation. International Statistical Classification of Diseases and Related Health Problems - 10th Revision. Geneva, Switzerland: WHO; 2011.
4. BLISS. Special Care for Sick Babies - Choice or Chance? London: BLISS; 2005.
5. N. Marlow, E. M. Hennessy, M. A. Bracewell, D. Wolke, E. PICure Study Group. Motor and Executive Function at 6 Years of Age after Extremely Preterm Birth. Pediatrics. 2007;120(4):793-804.
6. S. Johnson, J. Fawke, E. Hennessy, V. Rowell, S. Thomas, D. Wolke, N. Marlow. Neurodevelopmental Disability through 11 Years of Age in Children Born before 26 Weeks of Gestation. Pediatrics. 2009;124(2):e249-57.
7. S. Saigal, L. W. Doyle. An Overview of Mortality and Sequelae of Preterm Birth from Infancy to Adulthood. Lancet. 2008;371(9608):261-9.
8. Nuffield Council on Bioethics. Critical Care Decisions in Fetal and Neonatal Medicine: Ethical Issues. London: Nuffield Council on Bioethics, 2006.
9. Prime Ministers Strategy Unit. Improving the Life Chances of Disabled People. London:Strategy Unit, 2005.
10. V. A. Morgan, H. Leonard, J. Bourke, A. Jablensky. Intellectual Disability Co-Occurring with Schizophrenia and Other Psychiatric Illness: Population-Based Study. Br J Psychiatry. 2008;193(5):364-72.
11. E. Emerson, C. Hatton, J. Robertson, H. Roberts, S. Baines, F. Evison, G. Glover. People with Learning Disabilities in England 2011. Improving Health and Lives Learning Disability Public Health Observatory, 2012.
12. Mencap. Death by Indifference. London: Mencap; 2007.
13. J. Michael. Healthcare for All: Report of the Independent Inquiry into Access to Healthcare for People with Learning Disabilities. London: DH; 2008.
14. Improving Health and Lives Learning Disabilities Observatory. Confidential Inquiry into Premature Death of People with Learning Disabilities. Bristol: IHAL; 2012.
15. Mencap. Getting It Right Charter. 2010.
16. The Joint Health and Social Care Learning Disability Self Assessment Framework: How Local Learning Disability Communities Rate Themselves [press release]. Learning disabilities observatory 2014.
17. Disability Discrimination Act 2005. London: DH; 2005.
18. I. Tuffrey-Wijne, L. Goulding, N. Giatras, E. Abraham, S. Gillard, S. White, C. Edwards, S. Hollins. The Barriers to and Enablers of Providing Reasonably Adjusted Health Services to People with Intellectual Disabilities in Acute Hospitals: Evidence from a Mixed-Methods Study. BMJ Open. 2014;4(4):e004606.
19. S. Hollins, I. Tuffrey-Wijne. Improving Hospital Care for Patients with Intellectual Disabilities. Br J Hosp Med (Lond). 2014;75(6):304-5.
20. S. Hollins, I. Tuffrey-Wijne. Meeting the Needs of Patients with Learning Disabilities. BMJ. 2013;346:f3421.
21. L. A. Allerton, V. Welch, E. Emerson. Health Inequalities Experienced by Children and Young People with Intellectual Disabilities: A Review of Literature from the United Kingdom. J Intellect Disabil. 2011;15(4):269-78.
22. E. Emerson, C. Hatton. Contribution of Socioeconomic Position to Health Inequalities of British Children and Adolescents with Intellectual Disabilities. Am J Ment Retard. 2007;112(2):140-50.
23. E. Emerson, C. Hatton. Poverty, Socio-Economic Position, Social Capital and the Health of Children and Adolescents with Intellectual Disabilities in Britain: A Replication. J Intellect Disabil Res. 2007;51(Pt 11):866-74.
24. E. Emerson, C. Hatton, D. Felce, G. Murphy. Learning Disabilities: The Fundamental Facts. London: The Foundation for People with Learning Disabilities, 2001.
25. E. Emerson. Prevalence of Psychiatric Disorders in Children and Adolescents with and without Intellectual Disability. J Intellect Disabil Res. 2003;47(Pt 1):51-8.
26. C. Glendinning, S. Kirk, A. Guiffrida, D. Lawton. Technology-Dependent Children in the Community: Definitions, Numbers and Costs. Child Care Health Dev. 2001;27(4):321-34.
27. M. Mahon, M. S. Kibirige. Patterns of Admissions for Children with Special Needs to the Paediatric Assessment Unit. Arch Dis Child. 2004;89(2):165-9.
28. I. Kennedy. Getting It Right for Children and Young People: Overcoming Cultural Barriers in the NHS So as to Meet Their Needs. London: DH; 2010.
29. E. Emerson, C. Kiernan, A. Alborz, D. Reeves, H. Mason, R. Swarbrick, L. Mason, C. Hatton. Predicting the Persistence of Severe Self-Injurious Behavior. Res Dev Disabil. 2001;22(1):67-75.
30. C. Glendinning. Unshared Care: Parents and Their Disabled Children. London: Routledge Kegan Paul; 1983.
31. S. Baldwin, J. Carlisle. Social Support for Disabled Children and Families. A Review of the Literature. Edinburgh: HMSO; 1994.
32. J. Read. Disability, the Family and Society. Listening to Mothers. Buckingham: Open University Press; 2000.
33. K. Oulton, B. Heyman. Devoted Protection: How Parents of Children with Severe Learning Disabilities Manage Risk. Health Risk Soc. 2009;11:303-19.
34. Department of Health. National Service Framework for Children, Young People and Maternity Service. Disabled Children and Young People and Those with Complex Health Needs. London: HMSO; 2004.

35. K. Martin. Making Ourselves Heard: Exploring Disabled Children's Participation. London: National Children's Bureau Enterprises Ltd 2009.
36. Department of Health. The Children's and Young People's Health Outcomes Forum: Report of the Long Term Conditions, Disability and Palliative Care Subgroup. London: DH; 2013.
37. Care Quality Commission. Health Care for Disabled Children and Young People. Newcastle: CQC; 2012.
38. Annual Report of the Chief Medical Officer 2012. Our Children Deserve Better: Prevention Pays. London: DH; 2013.
39. M. Brady. Hospitalized Children's Views of the Good Nurse. Nurs Ethics. 2009;16(5):543-60.
40. I. Coyne. Children's Experiences of Hospitalization. J Child Health Care. 2006;10(4):326-36.
41. I. Coyne. Consultation with Children in Hospital: Children, Parents' and Nurses' Perspectives. J Clin Nurs. 2006;15(1):61-71.
42. T. Carney, S. Murphy, J. McClure, E. Bishop, C. Kerr, J. Parker, F. Scott, C. Shields, L. Wilson. Children's Views of Hospitalization: An Exploratory Study of Data Collection. J Child Health Care. 2003;7(1):27-40.
43. S. A. Sartain, C. L. Clarke, R. Heyman. Hearing the Voices of Children with Chronic Illness. J Adv Nurs. 2000;32(4):913-21.
44. V. Shilling, V. Edwards, M. Rogers, C. Morris. The Experience of Disabled Children as Inpatients: A Structured Review and Synthesis of Qualitative Studies Reporting the Views of Children, Parents and Professionals. Child Care Health Dev. 2012;38(6):778-88.
45. B Garth, R. Aroni. "I Value What You Have to Say". Seeking the Perspective of Children with a Disability, Not Just Their Parents. Disability and Society. 2003;18(5):561-76.
46. S. Sharkey, C. Lloyd, R. Tomlinson, E. Thomas, A. Martin, S. Logan, C. Morris. Communicating with Disabled Children When Inpatients: Barriers and Facilitators Identified by Parents and Professionals in a Qualitative Study. Health Expect. 2014.
47. M. Avis, R. Reardon. Understanding the Views of Parents of Children with Special Needs About the Nursing Care Their Child Receives When in Hospital: A Qualitative Study. J Child Health Care. 2008;12(1):7-17.
48. F. Jackson-Brown, J. Guvenir. The Experiences of Children with Learning Disabilities, Their Carers and Staff During a Hospital Admission. British Journal of Learning Disabilities. 2009;37:110-5.
49. K. Oulton, S. Kerry, L. May, D. Sell, C. DeSousa, F. Gibson. How to Meaningfully Include Children and Young People with learning disabilities in Research. Journal of Applied Research in Intellectual Disabilities. 2014;27(4):361.
50. K. Oulton, S. Kerry, L. May, D. Sell, C. DeSousa, F. Gibson. Hospital Staff Experiences of Caring for Children and Youth with Id and Their Families: It’s the Little Things That Make the Difference! Journal of Applied Research in Intellectual Disabilities. 2014;27(4):316.
51. Children and Families Act 2014. In: Office Stationery, editor. London2014.
52. I. Tuffrey-Wijne, N. Giatras, L. Goulding, E. Abraham, L. Fenwick, C. Edwards, S. Hollins. Identifying the Factors Affecting the Implementation of Strategies to Promote a Safer Environment for Patients with Learning Disabilities in Nhs Hospitals: A Mixed-Methods Study. London: NIHR, 2013.
53. W. Tellis. Application of a Case Study Methodology. The Qualitative Report. 1997;3(3).
54. R. K. Yin. Enhancing the Quality of Case Studies in Health Services Research. Health Serv Res.1999;34(5 Pt 2):1209-24.
55. D. E. Howard, C. Lothen-Kline, B. O. Boekeloo. Using the Case-Study Methodology to Teach Ethics to Public Health Students. Health Promot Pract. 2004;5(2):151-9.
56. Health and Social Care Information Centre. Learning Disabilities Census Report. England: hscic, 2013.
57. J. Blair. Staff Experience, Knowledge and Skills When Working with Children and Young People with Learning Disabilities in Hospital. 2013.
58. A. Clark, P. Moss. Listening to Young Children. The Mosaic Approach. London: National Children's Bureau, 2011.
59. A. Clark. The Mosaic Approach and Research with Young Children. In: Lewis V., Kellet M., Robinson C., Fruser S., Ding S., editors. The Reality of Research with Children and Young People. London: Sage; 2003. p. 157-61.
60. J. Davis, N. Watson, S. Cunningham-Burley. Disabled Children, Ethnography and Unspoken Understandings: The Collaborative Construction of Diverse Identities. In: Christensen P.M., James A., editors. Research with Children: Perspectives and Practices. New York, USA: Routledge; 2008. p.220-38.
61. L. Cameron, J. Murphy. Making Choices at the Time of Transition. Scotland: University of Stirling, 2000.
62. J. Coad, G. Plumridge, A. Metcalfe. Involving Children and Young People in the Development of Art-Based Research Tools. Nurse Res. 2009;16(4):56-64.
63. M. Horstman, S. Aldiss, A. Richardson, F. Gibson. Methodological Issues When Using the Draw and Write Technique with Children Aged 6 to 12 Years. Qual Health Res. 2008;18(7):1001-11.
64. K. Knighting, N. Rowa-Dewar, C. Malcolm, N. Kearney, F. Gibson. Children's Understanding of Cancer and Views on Health-Related Behaviour: A 'Draw and Write' Study. Child Care Health Dev. 2011;37(2):289-99.
65. B. Carter, K. Ford. How Arts-Based Approaches Can Put the Fun into Child-Focused Research.Nurs Child Young People. 2014;26(3):9.
66. T. Cook, E. Hess. What the Camera Sees and from Whose Perspective. Fun Methodologies for Engaging Children in Enlightening Adults. Childhood. 2007;14:29-45.

67. M. Mason. Incurably Human. London: Working Press, 2000.
68. H. Clausen. Doctors in Training Leading Quality Improvement: Families Reporting Critical Incidents and near Misses in a Children’s Hospital. London: The Health Foundation, 2014.
69. J. Ritchie, J. Lewis. Qualitative Research Practice - a Guide for Social Science Students and Researchers. London: Sage; 2003.