



## **Protocol**

### **Long Title:**

How do different neurodisability services meet the psychosocial support needs of children/young people with feeding disabilities and their families: a national survey and case study approach to mapping and costing service models, care pathways and the child and family experience?

### **Short study title:**

G-PATH SUPPORT: Gastrostomy feeding and psychosocial support

**This protocol has regard for the HRA guidance and order of content**

**Protocol version: 1:5 4-1-16**

**Research reference (IRAS): 178366**

**SPONSORS: City University London**

**FUNDERS REFERENCE NUMBER:14/04/40**

### **Representative of the Sponsor:**

Professor Jill Francis  
Associate Dean Research and Enterprise  
School of Health Sciences  
City University London  
Northampton Square, London, EC1V 0HB  
E: Jill.Francis.1@city.ac.uk  
T: +44 (0)20 7040 4084

**REC Reference:** (TBC)

### **Chief Investigator:**

Dr Gillian M Craig  
Senior Lecturer  
School of Health Sciences  
City University London  
Northampton Square  
London, EC1V 0HB, UK  
020 7040 5843  
[gill.craig.1@city.ac.uk](mailto:gill.craig.1@city.ac.uk)

### **Co-Investigators**

Harding C<sup>1</sup>, Flood C<sup>1</sup>, McCourt C<sup>1</sup>, Ayers S<sup>1</sup>, Sellers D<sup>2</sup>, Townsend J<sup>3</sup>, Dhandapani B<sup>4</sup>, Donaldson B<sup>5</sup>, Moss D<sup>6</sup>, Protheroe S<sup>7</sup>, Tuffrey C<sup>8</sup>, Cole M<sup>9</sup>, Gill A (Lay Chair)

<sup>1</sup>City University London, <sup>2</sup>Sussex Community NHS Trust, <sup>3</sup>The London School of Hygiene and Tropical Medicine, <sup>4</sup>Lewisham and Greenwich NHS Trust, <sup>5</sup>Kent Community Health Foundation NHS



Trust, <sup>6</sup>NHS Borders, <sup>7</sup>Birmingham Children's Hospital NHS Foundation Trust, <sup>8</sup>Solent NHS Trust  
<sup>9</sup>Triangle

## 2. SUMMARY/SYNOPSIS

<b>Short Title</b>	G-PATH SUPPORT: Gastrostomy feeding and psychosocial support
<b>Methodology</b>	Collective case study/mixed methods design involving three interrelated phases. Phase 1:national e-survey to identify exemplar models Phase 2:case study methodology, in-depth study of four contrasting service models Phase 3:resource utilisation and costs of service models
<b>Objectives/Aims</b>	To map how neurodisability services have responded to evidence reviews to embed structured support in care pathways: to  a) identify different support models nationally b) compare the implementation and operation of contrasting models of support and key resource differences c) provide an estimate of their costs, sources of funding and cost variations.
<b>Research Sites:</b>	The case is a local system of health and social care. Case study site 1 has been pre-selected and three more will be identified purposively following the national survey and the learning we garner from in-depth study of the first study site
<b>Inclusion criteria of case study sites</b>	<ul style="list-style-type: none"> <li>i) Evidence of written care pathway/best practice statement which includes psychosocial support</li> <li>ii) Evidence of linkages with local health and social care systems (eg. user groups/parent groups/voluntary sector and education)</li> <li>iii) Evidence of integrated care pathways (health and social care)</li> <li>iv) Evidence of designated staff who can provide psychosocial support</li> </ul>
<b>Number of Participants: Parents,</b>	Approximately 275 including:  131 in a national e-survey of child development teams.



<b>staff, children</b>	80 interviews across four sites;  8 focus groups (approx. 64 participants)
<b>Main inclusion criteria: interviews, focus groups, observation</b>	<p>Children with neurodisability and feeding difficulties and their families/HCP and other staff who provide care</p> <ol style="list-style-type: none"> <li>1. Professionals who work with families of children with neurodisability and feeding difficulties or manage services or who have a role for commissioning services,</li> <li>2. Families/caregivers of children with neurodisability and feeding difficulties, who may also receive nutrition via their gastrostomy tube or where a tube has been recommended,</li> <li>3. Children and young people who have gastrostomy tube feeds or have been recommended a tube, or have been identified as having complex feeding needs</li> <li>4. Children aged 5 and over with a reasonable level of communication using AAC for example</li> <li>5. Services that have indicated a willingness to participate in the research and have capacity to participate.</li> </ol> <p><b>Exclusion criteria</b></p> <ol style="list-style-type: none"> <li>1. Children/adults who lack capacity,</li> <li>2. Children under 5 years of age,</li> <li>3. Staff, children, families who are already involved in research studies which would suggest participation in this research would be too onerous,</li> <li>4. Children who have a tube as a result of cystic fibrosis or severe skin disorders.</li> </ol>
<b>Proposed Start Date</b>	01-01-16
<b>Proposed End Date</b>	31-12-17

### Summary

Children with neurological impairment (e.g. severe cerebral palsy) experience poor nutrition and growth due to difficulties chewing and swallowing. This can affect their ability to achieve an adequate nutritional intake by mouth (Reilly et al 1996). Mothers spend lengthy periods trying to feed a child (more than 3 hours a day) which can be experienced as stressful. Maternal depression therefore is not uncommon (Sullivan, 2004). A gastrostomy feeding tube (surgically placed in the stomach) is



recommended and prescription feeds. However the suggestion of a gastrostomy can generate parental opposition. This can result in conflicts between families and professionals around the child's feeding management particularly where oral feeding, which parents view as important, is considered unsafe and the child is severely malnourished. Evidence reviews recommend providing care pathways with structured support for families to improve informed decision making about children's feeding and the need for a gastrostomy feeding tube (Ferluga, 2013; Mahant, 2011).

**Aims:** This research aims to map how neurodisability<sup>1</sup> services have responded to these recommendations: to a) identify different support models nationally b) compare the implementation and operation of contrasting models of support and key resource differences c) provide an estimate of their costs, sources of funding and cost variations.

**Methods:** We will adopt a three-staged, mixed methods design involving:

1. An online national survey of policy and practice, support models, staff deployment and costings distributed through the British Academy of Childhood Disability national database of child development teams (n=240) and Nationally Managed Clinical Network for Children with Exceptional Health Care Needs (NHS Scotland);
2. A multi-site 'collective case study' (Stake, 2005) of four service models sampled purposively from the results of our survey and criteria developed in a familiarisation study (see figures 1 and 2) to reflect differences in levels of integration, organisational structure and insight into how support is embedded in a range of contrasting contexts. Figure 1 describes our initial sampling criteria in a 2 x 2 matrix describing levels of integration in care pathways (high/low) and levels of support offered (high/low). We plan to conduct interviews (n=20) and group discussions (n=2) per case with a theoretical sample of managers, staff, children, parents, teachers and others involved in children's care to analyse their experiences of feeding and support, and observations to examine how support, guidelines and protocols are implemented in practice, allowing triangulation of data;
3. A study of cost of support and associated resource use within different services will provide economic descriptive data using Personal Social Services Research Unit data (Curtis, 2011) and Willingness to Pay Methodology with service managers and carers.

**Analysis:** Numerical data (frequencies and summary statistics) from the national survey and costs will be analysed using SPSS. Qualitative interview data will be analysed using framework analysis (Ritchie & Spencer, 1994) informed by: Mahant et al.'s (2011) model of decisional conflict and support and Greenhalgh et al.'s (2004) organisational theory of factors influencing diffusion of innovation and barriers to implementation.

**Outcomes:** This research programme will produce evidence based guidance on models of support to inform commissioners of future services and make recommendations for change on how to embed support within the care pathway. We will identify the training and support needs of professionals and develop minimum standards of support.



Benefits to NHS: Estimating the key characteristics and costs of providing different models of psychosocial care will ensure a minimum standard of support for all families, equity across services and efficient use of staff time and resources. This, in turn, will help improve the quality of informed decision making so children and families achieve better quality of life, health and psychological outcomes.

## **Background and Rationale**

Neurodevelopmental conditions, such as cerebral palsy (CP), are the most common cause of disability, estimated to affect 3–4% of children in the UK (Emerson, 2012). This population of children are also the most frequent users of health services (Allard et al, 2014). A high proportion of children with CP experience disorders of swallowing and feeding with implications for their nutrition, growth and overall health (Reilly et al 1996). Dysphagia, although often under-recognized in this population of children is common (Reilly, 2000), with a reported prevalence ranging from about one-fifth of children with CP of any degree (Parks 2010) to 99% in children with severe CP and intellectual disability (Calis, 2008). Pulmonary aspiration is also common (where food or fluid enters the airway) resulting in poor respiratory health and is a cause of premature death in this group of children (Glover & Ayub, 2010). A gastrostomy feeding tube is usually recommended, surgically placed in the child's stomach, and meals are replaced with prescription feeds. Approximately 6,000 children in the UK are technology-dependent for their feeding (Kennedy, 2010) and the largest group of tube fed children are those with neurodisability.

Research suggests that parents of disabled children are vulnerable to stress due to the demands of care: 70% of mothers and 40% of fathers of severely disabled children which can, in turn, impact negatively on children's wellbeing and family functioning (Sloper, 1999). Indeed there is research to suggest that chronic stress, associated with caring for children with multiple and complex needs, can impact negatively on caregivers' quality of sleep, physical health and immune function (Gallagher et al. 2010; 2009). The demands of care are particularly high in children with feeding disabilities. Caregivers, usually mothers, report prolonged and stressful mealtimes (Sullivan, 2004). It has been estimated that children can take up to 15 times longer to chew and swallow their food, and that mothers would have to spend in excess of waking hours to ensure their child received an adequate nutritional intake by mouth (Johnson & Dietz, 1985). Significant maternal psychological morbidity has been reported in a community sample (Sullivan, 2004).

Research on the effectiveness of gastrostomy has tended to focus on the more biomedical aspects, including weight gain, although there is an increasing body of research that has highlighted the non-clinical aspects of tube feeding and the symbolic meanings parents attach to feeding by mouth (Craig & Scambler 2006; 2003; Thorne et al 1997). Although research reports improvements following gastrostomy, for example improvements in children's health, weight gain and maternal mental health (Pemberton et al 2013; Craig et al 2006; Sullivan et al 2005), there can be parental opposition to the procedure (Sleigh, 2004) creating conflicts about the feeding management of children. Conflicts may also arise in schools and other institutional settings where surrogate feeders may refuse to feed



children orally, sometimes against the wishes of parents, because of concerns about choking and an unsafe swallow (Craig & Higgs, 2012). Following gastrostomy insertion stress may also increase in the short term for some families due to the aftercare and complications of the procedure, the need to learn new nursing techniques (Pederson, 2004) and the grief associated with coming to terms with a medicalised way of feeding and other psychosocial sequelae (Calderon 2010; Enrione et al 2005; Thorne et al 1997). The loss of oral feeding that some children experience post gastrostomy may impact on dental and oral hygiene (FDS, 2013) and raise quality of life issues for parents. Support to re-establish oral feeding however is not consistent. The disciplinary background of health care professionals (HCPs), funding arrangements and type of service configuration may also impact on the ability of services to support families (Morrow et al 2007; Spalding & McKeever, 1998). Those services that are well integrated may also struggle to support families because there are no psychological services to refer to, or designated professionals who can perform this role within the remit of their job. This can create a disproportionate burden of time on team members. Although some children with specific conditions, e.g. Rett syndrome and cystic fibrosis, can access support from specialist nurses, children with neurodisability may only access specialist care post-surgery leaving gaps in pre-surgery support. Some families may be able to access support through third sector organisations or services may have developed links with the voluntary sector to perform this role. We therefore plan to map these relations as part of this study.

Where children are severely malnourished, and parents refuse a gastrostomy, clinicians may consider child protection proceedings but there are inconsistencies in the use of safeguarding legislation and it is unclear whether this could be avoided if adequate support were in place or guidance governing its use. This is particularly pertinent given the recent high media attention some cases have received where professionals and parents have disagreed on the best care for their children, for example Aysha King (Craft, 2014).

To summarise, there is an established literature which has highlighted the stressors parents of disabled children experience in response to the demands of caring for a child with feeding disabilities. Significant psychological morbidity has been demonstrated in mothers of children with consequences for their own health and the wellbeing of families. Stress may be further impacted because of the decisional conflict (Mahant et al 2011) parents experience when a gastrostomy is recommended and post-surgery, due to the complications of the procedure and the practical management of the tube both medically and in social arenas.

### **Evidence for need of study: why now?**

A number of factors suggest this research is needed and timely including evidence from reviews, the lack of existing guidelines and, the new NHS outcomes framework and commissioning agenda.

#### **1. Evidence reviews**

In addition to the service user involvement work with parents and consultations with professionals, conducted during the familiarisation study (funded by City University London in preparation for this





grant proposal), which identified gaps in service delivery and the need for support, there is a growing body of research that has highlighted the importance of the psychosocial aspects of feeding a disabled child and the need to improve the quality of decision making and emotional support.

a. A systematic review of 11 qualitative research studies conducted in the UK, Canada and Australia (Mahant et al 2011) highlighted the decisional conflicts parents experience when considering a gastrostomy and the role of services in shaping parental experiences of care and ameliorating stress. Eight of the eleven studies reported that HCPs did not provide enough support to families. Four studies reported that families felt pressured to accept a gastrostomy and six reported that professionals failed to appreciate the difficulty of the decision making process for parents and the impact of tube feeding on child and family life. Lack of information, conflicting information and lack of opportunity to meet with other families, in addition to concerns about the operative procedure, were also reported. The authors concluded that: 'Care pathways are needed to provide consistent and structured processes around decision making' (Mahant et al, 2011:1480).

b. A systematic review of the evidence of gastrostomy feeding by the American Agency for Health Care Research and Quality also emphasised the importance of non-clinical factors influencing parental decision making. The review highlighted the role of support in ameliorating stress, and concluded that: "Stressors associated with caring for a severely disabled child and the potential impact of feeding interventions on the relationship with the child should not be underestimated" (Ferluga et al., 2013:16).

c. A Children's and Families' Services knowledge review of care pathways for children with complex health care needs (not feeding) also highlighted gaps in the provision of emotional support for families (Marchant et al 2007). The study, which involved a consultation with children and families and a survey of professionals, highlighted what the authors termed, the problem of 'divided thinking' which dominates service provision whereby social care and health needs are deemed to be separable and therefore met by different agencies or professionals. The authors reported on a system of 'rule breaking' by professionals in order to meet children's needs which were seen to be outside of their remit or contract arrangements. The authors suggested that a blurring of role boundaries was often necessary to meet child and family needs but highlighted the role of commissioning arrangements as a potential barrier to delivering best practice in terms of governing what professionals / services could or could not do. The recent Children and Young People's Health Outcomes Forum, established to develop health outcomes, has also called for better integration of children's health and social care, including models of funding which 'encompass whole pathway provision' (RCYPHOF, 2014:6).

## **2. The need for guidelines**

We know that parents who are stressed or experience depression interact differently with their children which, in turn, can affect child outcomes or impact on relationships with siblings (Allen, 2011; Hastings & Beck, 2004). The evidence also suggests psychosocial support can ameliorate caregiver stress (Lazarus & Folkman, 1984). Responsive services can therefore play a pivotal role in



supporting families with positive impact on both child and family wellbeing (McConachie, 1999). A literature search of best practice in the UK however found only one of two sets of guidance that referred to the need to provide families with: 'practical, emotional and social support' (NHS Quality Improvement Scotland, 2007:1).

### **3. NHS Outcomes framework and commissioning agenda**

Enhancing the quality of life for people with long-term conditions and, in particular, ensuring people feel supported to manage their condition and have a positive experience of care have been identified as outcome performance indicators in the National Health Service Outcomes Framework (DH, 2013). Supporting parents to feel more confident in the management of their children's feeding and improving the quality of life for children and their caregivers is in keeping with the framework. Recent research also suggests that children and young people consider quality of life as key to their health outcomes (Allard et al, 2014), including: emotional wellbeing, community and social life and the ability to participate in social activities. Given the recent changes in the commissioning landscape (The Health & Social Care Act 2012) a better understanding of the influence of commissioning arrangements in relation to service delivery models and supportive practices for children with feeding disabilities will help to demonstrate how support is accomplished in different service models in relation to these broader definitions of health outcomes.

### **4. Need for evidence on operation of services**

There is a heterogeneous approach to the delivery of services to children with neurodisability including regional, and cross country variations in the use of gastrostomy. Such variation can also make it difficult to embed evidence-based packages of support. Variation has been attributed to differential access to services, parental choice, differences in clinical practice and general lack of guidelines (Dahlseng et al. 2012). Parental and provider delay in recommending a gastrostomy may have implications for children's nutritional status, growth and development as well as the psychological wellbeing of parents; for example 83% of children presenting for surgery for gastrostomy are under the third centile for weight (Stringel et al 1989). Services may fall short of providing structured support because: 1) they lack capacity; 2) care pathways are complex and not integrated; 3) although there are multidisciplinary teams there is no agreed consensus on what teams should consist of; and, 4) there is no national guidance on service delivery models, including support.

There is evidence from wider studies in health care that organisational and service delivery factors may impact on the quality of care (Mannion et al. 2005, Nuffield Trust 2008). A series of fifteen US-based case-studies reported in a Commonwealth Fund Report on healthcare organisation, identified six key attributes of high-performing systems as: information, continuity, patient engagement, care coordination, team-oriented care delivery, continuous innovation and learning, and convenient access to care. These attributes were supported by values-driven





leadership, interdisciplinary teamwork, integration and aligned incentives (both at the organisational and provider level), accountability and transparency (McCarthy & Mueller 2005). Inter and intra-professional relationships, communication and information channels, staffing models, skill mix and service ethos have been highlighted as potential factors in quality (Kings Fund 2008, Rowe et al. 2001). Little is known about the impact of aligning service delivery models, philosophy of care, support packages and the child and family experience. In particular the role of peer support or third sector organisations in delivering support is under researched. Although services may informally put families in contact with each other, more information on the workings of these informal, or formalised, relations and their interface with services is needed given the evidence that peer support can have a positive impact on the mental health status of mothers of children with chronic illness (Ireys, 2001).

This research aims to respond to these policy streams and evidence reviews. It will map national practice and, in particular, different approaches to support in four contrasting service delivery contexts using a case study approach. The findings from identified exemplar services will assist others to integrate the clinical and support pathways in a more structured way to improve the quality of support, decision making and health outcomes for children and their families.

### **Aims & objectives**

This research aims to map how different neurodisability services have responded to the recommendations of evidence reviews on the need to provide families considering a gastrostomy feeding tube for their disabled child with consistent and structured support within care pathways to improve the quality of informed decision making. We aim to: a) identify different service support models nationally; b) compare the implementation and operation of contrasting models of support and key resource differences; and, c) provide an estimate of their costs, sources of funding and cost variations.

#### **The objectives are to:**

1. Establish a national (UK) picture of models and practice (on line survey of 240 child development teams disseminated through a national research database of disability leads),
2. Compare the implementation and operation of four contrasting service models and practice, selected purposively in relation to criteria identified in our familiarisation study and national survey (see figures 1 and 2), using a collective case study research methodology,
3. Analyse how support is accomplished in different organisational structures including health and social care sectors and the role of schools (visual mapping of care pathways, interviews with staff, families and young people, document analysis),
4. Provide rich description of contrasting supportive practices and the interface between services and other key agencies (interviews, document analysis, and observations of key meetings),



5. Identify different features of service configurations and supportive practices and analyse their sources of funding and cost variations (questionnaires, clinical records). More specifically we will: a) assess the resources required for each of the models considered and how these costs impinge on the NHS and Social Care Services and, b) estimate the costs of providing different models of psychosocial care nationally (WTP methodology),
6. Identify if any models of support utilise service user involvement or third sector organisations (i.e. families with experience of gastrostomy feeding or voluntary organisations) and analyse how service users/ family support organisations interface with the formal health and social care sectors and how they are resourced (surveys, interviews, document analysis),
7. Analyse professionals', young people's and their families' experiences of support (analysis of patient audits, interviews, observation),
8. Identify contextual issues that influence variations in practice including commissioning arrangements (interviews, document analysis),
9. Derive best practice guidance on support issues from an explication of the service models identified in the study and service specifications and, make recommendations for change to inform commissioning decisions and set standards of support for children and families,
10. Make recommendations for embedding support for a wider constituency of children with complex health needs and their families, including recommendations for training and skilling the wider children's workforce, by deriving general principles of how services can provide support and where in the care pathways this should happen (case study of exemplar services).

**Information in relation to management and organisation of services.**

Additionally we aim to answer the following questions where possible:

- a) How do referral pathways operate in different models of service organisation
- b) How does variation in service delivery impact on staff/children/family experiences of support
- c) How do these different service configurations compare in terms of value for money
- d) What is the staff deployment pattern and grade mix and how do variations in staffing impact in terms of equity and access of service provision
- e) What models of psychosocial/practical support exist for children, parents, and families more generally (including siblings of children, fathers and other family members) where and when is it offered and by what type of professional/service (e.g. designated or ad-hoc). Are there opportunities for flexible appointments for fathers, other family members etc.)
- f) Are service users/peer led support models involved in supporting children, parents and how are relations with formal services configured, supported, resourced, accessed by children/parents and appraised by staff and families



- g) How are protocols and guidelines used in practice, particularly with regard to the care of the gastrostomy, the safety of oral feeding and the reintroduction of oral feeding post gastrostomy where applicable
- h) How is shared care accomplished across community, secondary and tertiary sectors and conflicts around children's feeding managed including child protection policy and guidance
- i) What kind of support is available for staff e.g. clinical supervision when working in challenging contexts
- j) How are outcomes of care monitored and measured in relation to care plans?

## Methods

### Design and conceptual framework

The research design is a three-staged, mixed methods exploration of service delivery support through a national survey, and a case study illustration of four exemplar services and their costs. Primary research of this nature is needed before a formal evaluation of support contingencies is possible as recommended in the Medical Research Council Guidance (2008) which emphasises the need for researchers to have conducted appropriate development work and gained a better understanding of contextual factors that could inform the design of a future formal evaluation of different support models.

#### ***PHASE 1. Health Care Professional's survey: Child Development/Feeding Teams***

We will conduct a UK wide, web-based e-survey of all disability leads and other key professionals in Child Development Centres (CDCs) (n=240) identified through the British Academy of Childhood Disability (BACD) database of disability leads. We estimate that 131/240 child development centres have feeding teams. In keeping with the purposes of the survey there will be no sub-sampling: all CDTs nationally will be invited to participate. Previous surveys using the BACD database have obtained response rates of 94% (Parr et al. 2013). Based on these estimates we would therefore expect information on a 121 feeding teams. The BACD is a multidisciplinary organisation and sub group of the Royal College of Paediatrics and Child Health. The BACD research group supports high quality disability research and has given permission to use the database (see supporting letter). The aim of the database is research utilisation in order to improve child health and service development initiatives. On our behalf the BACD will email all leads nationally with our electronic survey; they, in turn, will disseminate to other professionals involved in feeding. We will ensure wide and inclusive coverage of all practitioners involved in children's care through our networks including gastroenterology, speech and language therapy, and dietetics identified through the project co-investigators and through the Royal Colleges. In Scotland we will distribute the questionnaire through the National Managed Clinical Network for Children with Exceptional Healthcare Needs (CEN), NHS National Services Scotland.



Our survey will provide a picture of established practice, processes and support delivery mechanisms, service specifications detailing entry gates to support, information about integrated care pathways and commissioning arrangements, staff deployment in core teams, information about catchment areas, preliminary costing information, the use of guidelines and protocols and particular issues for professionals in supporting families, including perceptions of service user needs and gaps in provision. The role of third sector organisations and service users in offering support will be a core theme of the survey. We will maximise response rates through follow-up, telephone reminders and advanced publicity about our study through our partner organisations. Where possible we aim to check information provided in surveys against information publicly available through websites and via follow up telephone calls (Pilger et al 2012). From the national survey we aim to derive typologies of service delivery and psychosocial support and further refine the criteria for the selection of our case studies for in-depth investigation.

### ***PHASE 2. A collective case study***

We aim to conduct a multi-site, 'collective case study' (Stake, 2005) of four exemplar services, selected purposively based on preliminary criteria developed in our familiarisation study (see figure 1) and from the data provided in our national survey to analyse how support is accomplished in different contexts. Figure 1 describes the rationale for our sample size using the criteria: levels of integration in care pathways (high/low) and levels of support in a 2 x 2 matrix. Additional criteria will include different geographical areas (as catchment area and commissioning arrangements are likely to determine service delivery to some extent), service user involvement and, services with designated staff with specialist roles (or enhanced roles). Relationships with schools were also suggested in our service user consultation. The cases are intended to provide exemplars of different service models and four cases will be selected to balance the need for a small sample to facilitate in-depth data collection and sufficient scope to explore variation in levels of integration and support within different local systems. A 'case' covers the local health and social care system rather than a single service in order that these issues and linkages can be explored.

Robson (1993:52) describes case study as: "a strategy for doing research which involves an empirical investigation of a particular contemporary phenomenon within its real life context using multiple sources of evidence." This is important when analysing complex health care organisations and systems of care as it allows for both depth and breadth of analysis through multiple data sources and the differing perspectives of key players (Greenhalgh, Kushner, Russell 2014; Yin, 2003). For example we aim to map best practice but as this itself is a contested term, and is likely to differ from the family, provider and commissioner perspective, we aim to use the case study to bring into relief the discursive construction of good care in a variety of settings through interviews with a range of stakeholders, observations of key meetings and a review of strategic documents and protocols. Greenhalgh (2007) aptly describes the contested nature of practice with reference to a range of frameworks traditionally used to assess quality, including: The Donabedian Three (i.e. the design and organisation of the service, process and outcome); or, The Maxwell Six (i.e. effectiveness, efficiency,



acceptability, equity, access and relevance). Family centred care (FCC) is also seen as a marker of quality (BACCH, 1994). FCC defines best practice in terms of those services that deliver responsive and flexible care, work in partnership with families treating them as experts and where care plans attend to the families' wider social networks. Signposting, advocacy and key working are also features of FCC (Marchant, 2007). Our case study aims to bring into relief these different understandings and will identify philosophies of care and best practice to see how these are operationalised and delivered.

Our case study will focus on the different perspectives of families, HCPs and commissioners to gain a better understanding of the notion of good care with reference to local service configurations.

### ***Initial selection criteria case study sites: integration and support***

Integrated care has been operationalised as a care pathway approach and defined as: "structured multidisciplinary care plans which detail essential steps in the care of patients with a specific clinical problem" (Campbell & Hotchkiss 1998:133). The European Pathway Association, adopting Vanhaecht et al's (2007) definition of a pathway suggests: "A care pathway is a complex intervention for the mutual decision making and organisation of care processes for a well-defined group of patients during a well-defined period". The following steps have been identified as the defining characteristics, and hence standards of integrated care pathways:

- evidenced based
- multidisciplinary with identified roles and responsibilities
- involving patients/carers
- crossing boundaries
- recorded in single documentation and standard format
- outcomes orientated
- audited on a regular basis as part of quality monitoring
- deviations from the care pathway are recorded as part of variance analysis (National Leadership and Innovation Agency for Healthcare, 2005, p.8).

We acknowledge that services may use additional criteria to describe integrated care pathways or different terminology (e.g. best practice statement) or operate according to a different definition and we aim to reflect this in our questionnaire survey. Discussions of integrated care will form part of the qualitative interviews to gain staff and parents' understandings and experiences of integrated care pathways and structured support. We expect to find that some services are not integrated in these terms, but engage in joint working which may also have merits. We also recognise that care may be integrated within a service but not across services and we will ask staff and parents to identify where these gaps are and the impact on children's care. We aim to identify reasons for the gaps and invite suggestions for strengthening these areas.



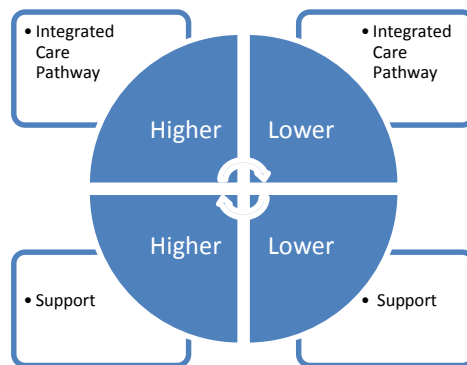
We will define 'high' levels of support in terms of the availability of designated professionals with specific remits and training (e.g. psychologists, counsellors, family support workers). We aim to use Mahant's model of decisional conflict as a tool to identify the nature of support parents are deemed to require and how services address these needs. Based on a qualitative synthesis of the evidence, this model outlined three concepts and 28 themes that can contribute to conflict between HCPs and families, including: the values parents attach to the meaning of feeding and tubes; the child and family context including unique family characteristics; and, a service's ability to share information and support families (process issues). We will use this framework as a focus for discussion with HCPs and parents in interviews and group discussions to identify examples of structured support in the care pathway, good practice and any gaps in services. The framework has been used in interviews with HCPs to examine their awareness of decisional conflict and strategies for support (Morgan et al. 2014). It will allow professionals and parents to reflect on, and discuss, the different meanings of support and how support is accomplished in different organisations.

We will select our cases in line with our initial selection criteria and willingness to participate is an additional criterion (Pope, 2005). However we are also mindful of the time constraints of conducting a questionnaire survey and four cases within a 2-year timescale and the need to build in time for R&D permissions. Pragmatically therefore, we aim to frontload the selection of our first case in advance of the survey analysis, using the criteria developed from our familiarisation study (see figures 1 and 2). Case study 1 [anonymised] for example, would meet the criteria of higher levels of integration and support, including integrated care pathways with schools. It is likely that a case study site will be in Scotland because of the low volume of children who are managed by a small number of highly specialised and trained staff within nationally managed clinical networks (see <http://www.cen.scot.nhs.uk/about-nmcm>). Unlike the BACD, which is a professional network, dissemination through the CEN (Children with exceptional health care needs) will require R&D approvals as it is a network which comes under NHS Scotland.

Given conversations about children's feeding can be initiated in schools and other arenas involving surrogate feeders, and it can take several months for a referral to specialist centre, we aim to identify opportunities for early intervention and support outside of the formal health care sectors. The role of schools and peers in supporting children and their families is a key focus. We aim to map the feeding pathway/family journey within local care systems as the preliminary unit of analysis. We are aware that much parental interaction happens through schools where there may be a critical mass of children with feeding difficulties and a gastrostomy. Luncheon clubs, tea party groups, after school clubs for children and families provide opportunities for parental interaction and support which is more difficult to access for pre-school children. Case study 1 [anonymised] has been selected because of: i) the links with schools and schools have been identified as having an important role in health and wellbeing agendas in the NHS outcomes framework (DH, 2013); and, ii) the potential role of schools in supporting families was identified in our familiarisation study with service users.

Figure 1: Preliminary Selection Criteria





### Interviews with key stakeholders

We aim to conduct up to 20 semi-structured interviews and two group discussions per case with a theoretical sample of service managers, staff, families and commissioners where appropriate. Given the composition of teams will vary in different services, key informants will be identified through snowball sampling, a recognised approach in case study research (Pope & Mays, 2013). Interviews with professionals will identify their perceptions of needs of parents/families and how services meet these needs.

We aim to interview up to ten parents and five young people per case to explore their experiences of care pathways, how needs were met, any gaps and recommendations for change. Child Development Teams and other services usually have parent groups associated with their services and will provide an opportunity to access the parental viewpoint. Where feasible group discussions with inter-professional teams will be conducted instead of individual interviews, since the interaction and discussion is likely to generate useful data and triangulation of opinion. The interviews with parents and children will use a semi-structured approach, as they will seek open views as well as responses to more focused questions. We will use process mapping (NHS, 2008) and visual methods to map care pathways, including comparison of formal pathways in documentation with maps constructed by service providers and users (McCourt et al. 2014). We will seek out information on history of service and support developments, models of support and how support is delivered in practice and any gaps. We will also identify the barriers to providing support and the training needs of staff to deliver support. More detailed questions to be addressed in the case study include the following:

It will be difficult to have standardised interview schedules for children as children's ability to respond will depend on a range of factors including: their individual experiences of feeding/tube feeding, cognitive ability and communication preferences. We will therefore aim to adopt a personalised approach using different types of questions (open/closed) and media (talking mats with symbols, story boards, dolls ) to enable children of different ages and abilities to participate in line with guidance on involving children and young people with communication impairments (see Morris, 1998 for example).

### **Observations of key nodes in service delivery**

We plan detailed observation of selected aspects of services at key junctures including care planning meetings, review meetings, case conferences, and meetings with family groups, referrals for investigations and for surgical opinion. Due to time restrictions however these non-participant, observations will be focussed and time-limited involving up to 20 hours of observation per case. The aims of the observations will be threefold and in addition to document review will: 1) provide a baseline understanding of the context of care and sensitise us to the influences of this on care delivery; 2) highlight issues of relevance that can be explored in interviews; and, 3) triangulate data obtained from interviews with different groups to enhance the depth of analysis of interview data.

Observation can be a valuable complement to interview data, since it can illuminate gaps between intentions or ideals and practices and can enhance understanding of the contextual constraints and influences on practice. It can highlight areas of tension that may be addressed sensitively in interviews and can help to illuminate why interview accounts from a range of respondent groups may offer differing accounts and explanations of care pathways and support.

### **Document review**

We will obtain key documents relevant to the case study for analysis (e.g. service documents, service specifications, audits and protocols, parent satisfaction surveys, care plans, care pathways, mission statements, protocols on eating and drinking and psychosocial support, school meal policies, dysphagia policies) and, where possible, we will obtain documents prior to site visits. The aim of the review is threefold; to provide information that will: a) sensitise us to the context of service delivery; b) inform interview schedules and, c) allow us to triangulate data obtained in the e-survey and subsequent interviews with staff and parents.

### ***PHASE 3. Resource utilisation and costs***

We will adopt a three stage approach to the study of costs of support and associated resource use of four contrasting service models as follows:

a) The main cost analysis will include cost of resources as reported in the clinical records (paper based/electronic) of our cases, together with results from the survey of staff involved in delivering support, whether performing a designated role or an enhanced role or informally. Staff time will be captured from clinical records and from their own estimates of the time they spend delivering non-clinical support. This approach has been tested in a questionnaire survey with a special interest dysphagia group as part of the familiarisation study. Data on staff deployment, grade and staffing model will be ascertained from both sources. Staff time will be costed using prices from the Personal Social Services Research publication (Curtis, 2011) to obtain mean costs and variability of costs. Parents of children who had a gastrostomy in the previous 12 month period (to allow for sufficient



throughput and control for recall bias) will also complete questionnaires on the support they have received to allow for triangulation of data where possible.

b) Secondly there will be more detailed analysis of resource data and practice with in-depth costings of each service delivery model, to estimate differences in the costs of each alternative, in a descriptive cost-consequences analysis. Costs and service use will be presented separately and described in detail, so decision makers will be able to judge what they consider to be good value for money.

c) Willingness to Pay (WTP): Key respondents (providers, budget holders and service users) in each of our four cases will be given questionnaires where the models of care we have identified in our case study will be presented as hypothetical services with detailed descriptions of what would be provided, the level of resource that would be incurred and the professional support that would be offered to families. We will then evaluate what financial value families' and providers' would attribute to each of the identified models in our case study using WTP methodology. Questionnaires will be administered with the ultimate aim of producing monetary valuations of what respondents would be willing to pay in practice for each service model described. Respondents will be asked to rate and rank each of the different psychological support models in order to determine a preference structure.

This direct approach for ascertaining 'preference' and 'acceptable price' is suited to the proposed research question as the anticipated number of care models will not be large. The approach to the respondents described above will capture the expert judgements of commissioners, clinicians and provider managers. Carer perspectives will be sought using the same approach to get a carer monetary value to compare with those of professionals and decision makers. We will pilot the scenarios with service users to ensure they are presented in ways which are legible to families. Respondents will be given the option of a range of prices to assess what they would be willing to pay (a) to see the described service provided or (b) what they would be willing to pay to receive the care model if they are carers. The average WTP will be determined for decision makers and for carers and will allow comparison with the results of other WTP studies for treatments in health care.

Willingness to Pay methods can provide decision-makers with a broad range of information (Tambor et al. 2014) and are conceptually easier to understand than other economic measures and the information elicited can therefore be easier to explain to policy makers and other stakeholders. Additionally as a methodology it is rooted in sound economic theory (Bridges, 2003). Criticisms of WTP concern the results being affected in biased responses by a perceived ability to pay when used with health consumers. This can be addressed in analyses by weighting responses according to income and using regression methods (Bridges, 2003). Table 1 illustrates each activity the different groups will be asked to participate in.



<b>Table 1 Research Participant Activities</b>			
<b>Activity</b>	<b>Staff</b>	<b>Parents</b>	<b>Children</b>
<b>PHASE 1: Survey</b>			
National Survey Child Development Teams/equivalent disseminated through the BACD (and clinically managed networks in Scotland) and completed by paediatric community leads	1	0	0
<b>PHASE 2: Case study</b>			
Interviews (participants drawn from across local health and social care teams)	1	1	2
Focus groups (participants drawn from across local health and social care teams) NB. Staff and parents will participate in either an interview or focus group, not both	1	1	0
Observations of care processes in multi-disciplinary team meetings	1	1	1
<b>PHASE 3: Resource utilisation and costs</b>			
Completing a questionnaire about appointments & support	0	1	0
Completing a questionnaire about time spent supporting families	1	0	0
A short questionnaire demonstrating preference for four service models & their costs (WTP methodology)	1	1	0

**Data Analysis:**

We will analyse numerical data, including information about costs, with SPSS using frequencies and summary statistics for the national e-survey to develop service profiles and typologies. Case study interviews and focus groups will be professionally transcribed and analysed using framework (Ritchie & Spencer 1994) which allows a team approach to both inductive and deductive coding informed by our theoretical frameworks. The stages include: familiarization with data; development of a thematic framework through emerging themes; indexing and labelling of themes within and across interviews; charting themes under categories; and, mapping and interpretation. Comprehensive field notes will be uploaded alongside interview transcriptions and codes into the qualitative software (NVivo10) to aid analysis. We will record the process of research and analysis in a reflexive log (Ortlipp, 2008). The reflexive log will be dual purposed, providing an audit trail of key decisions taken throughout the research and a learning resource which is likely to inform articles which will contribute to the case study literature. Two researchers will conduct field work supervised by the Chief Investigator. The framework described by Greenhalgh (2004) will be used to structure how systems of support have been implemented allowing for comparisons across our four selected cases. However we aim to be flexible in our analysis, so in addition to framework we will use a narrative approach to provide rich illustrations and storied accounts to convey multiple perspectives. Following immersion in the data common themes will be mapped across multiple data sources and compared across the four cases using cross-case comparison and negative case analysis. Findings will be synthesised using a thematic synthesis approach in line with our case study protocol (see figure 2) and theoretical resources.

**Plan of investigation and timetable**

Ethical approval will be obtained within the first four months and the process of obtaining R&D approvals will be initiated immediately in conversation with the CRN. We will recruit project staff. PPI involvement will develop project information sheets and input into the survey tool, which we will finalise and disseminate within the first three months, with analysis taking place in the following three months. We aim to recruit the Research Assistant by the six month. We will conduct a further PPI consultation to input into the development of interview schedules and prepare to conduct the first case study in months 7-9 with concurrent preliminary analysis taking place. Data gathering will take place with case studies conducted serially, each taking approximately three months of field work and analysis. There will be PPI input into the Willingness to Pay questionnaires in months 19, which will be distributed month 21. Final analysis of all data, report writing, PPI involvement in dissemination events will take place in the final three months where we will run dissemination workshops with each of the four sites and we will gather input into standards of support. Here we will invite service user input. Dissemination is likely to continue beyond the project timeline.

**Ethical issues**



We will obtain ethical approval from the REC. Given that children with neurodisability are likely to be viewed as vulnerable we will apply for full ethical approval. In conducting the study, we will be particularly mindful of the ongoing ethical challenges of conducting small-scale qualitative research, including the use of observation. The key ethical issues posed by such an approach are the need to obtain and regularly reaffirm consent with a range of participants, to ensure that privacy and confidentiality are respected and protected in a small setting, and to take measures to ensure that care of people who are not directly involved in the study, and who have not given consent, is not inadvertently recorded. In addition to individual information and consent procedures (which will involve written information sheets and verbal explanations, signed and verbally reconfirmed consent), information sheets and posters about the study will be actively circulated and displayed in the relevant service areas. We will ensure comments reported in the public domain cannot be attributed to single individuals or services.

Our approach is based on general ethical principles of maintaining the dignity, privacy and confidentiality of participants, and avoidance of any potential harm. A study of this type has no direct impact on the care or treatment that children receive, or the work process of staff, and all participants will be assured of this. They will also be assured of their full rights to decline to participate or to withdraw their consent at any point, without need to give explanation and without their care being affected. Observation has the potential to indirectly affect care processes, as those involved may be conscious of being observed, although many observation-based studies affirm that the effects of observation tend to be very limited in practice. The research observers will adopt an unobtrusive approach; observations will take place in public areas. Any observations of care processes will involve full information and consent for parents as well as the health professionals involved. Interview participants will be given a verbal explanation of the study in addition to the information sheets and consent form. Codes will be used for confidentiality and care will be taken to ensure full compliance with data protection and security.

Guarantees of confidentiality and anonymity given to research participants will be honoured, unless there are clear and overriding reasons to do otherwise, for example or in relation to the abuse of children or criminal acts (BSA 2004). Where the observation or interviews reveal details of overt system failures, the nature of these will be fed back to the appropriate authority after discussion with the project investigators and clinical leads. Interviews with young people and families may reveal a safeguarding issue and we will devise protocols to deal with this. In the event that children or parents become distressed we will devise distress protocols and follow local custom and practice.

Ground rules will be negotiated with children on how we should conduct the interview, including pace, pauses, breaks and withdrawal from the interview and how these should be indicated (eg through the use of symbols) to the researcher.

Triangle, who will co-ordinate our PPI for the service user group (not interviews), also build in a contingency fund within their charges for safeguarding.





## **Consent**

1. Parents will give their consent for children under the age of 16 to participate in an interview and children will also consent. Additionally we will re-consent children verbally in the first of two interviews using symbols and talking mats which will be recorded by photograph.
2. We aim to consent all activities with families as one process as far as possible so that families are aware of all the activities they will be asked to participate in:
  - a) for the interviews and focus groups forms will be sent by post or distributed at clinics by the local PI or research team. Consent will take place at the parental home and forms returned to the university or local PI
  - b) in the case of observations of care processes, because these may be identified opportunistically, we aim to consent/re-consent in the clinic with the help of the local PI or Clinical Nurse Specialist. Posters will be advertised in the clinics advising parents that researchers are observing care processes, with permission (written consent) and that if they don't wish to participate they can indicate to the clinical staff without prejudice,
  - c) with respect to the economic costings, all families with contact with services in the last 12 months will be sent a questionnaire asking them about their contacts with services in relation to psychosocial support. Completion of the form assumes consent (for those families who haven't consented already in advance). Additionally we will ask for consent to review children's clinical records to further ascertain costs.
  - d) all staff will be asked to provide written consent in advance of interviews, focus groups and observations and we will re-consent verbally.

Parents will therefore be able to opt in/out of the different aspects of the study. They will be advised that if they initially consent, but then change their minds about participating in any activity, this is an option as they will always be asked verbally to re-consent. Children will be able to withdraw at any point by indicating with a symbol (negotiated as part of ground rules) their desire to pause, stop or withdraw completely.

## **Patient and Public Involvement (PPI)**

We have developed the proposal with service user input and we have designed a model of working with our PPI reference group (parents and young people) which allows their active involvement in all stages of the project including the development of research tools and dissemination activities.



Our research study will be conducted with the active involvement of parents and young people in addition to a range of health care professionals and commissioners. The project advisory group will include a parent and caregiver of a disabled child. The other arm of our PPI strand will be managed by [anonymised] an independent organisation that works to reposition children in society as competent communicators and active agents in their own lives. The organisation will arrange a PPI reference group including: a parent reference group (three mothers) and a young people's reference group (four young people) to be involved in our study. We have proposed 12 meetings with parents and young people over the course of the project. Input into research instruments will ensure that the questionnaire survey and interview schedule contain questions that are relevant to the experiences of service users based on their interactions with health and social care providers. We will construct and pilot our WTP questionnaires with parents to check that they are easy to understand and to complete. We will ask for input into project information sheets to ensure all information is user friendly. We aim to discuss our findings and get service user input to help us make sense of the data that we find. We will involve them in dissemination activities, for example, inputting into materials and assisting with the development of standards through the dissemination workshops. This will ensure that information is appropriate, user friendly and accessible to others, including those with alternative communication needs.

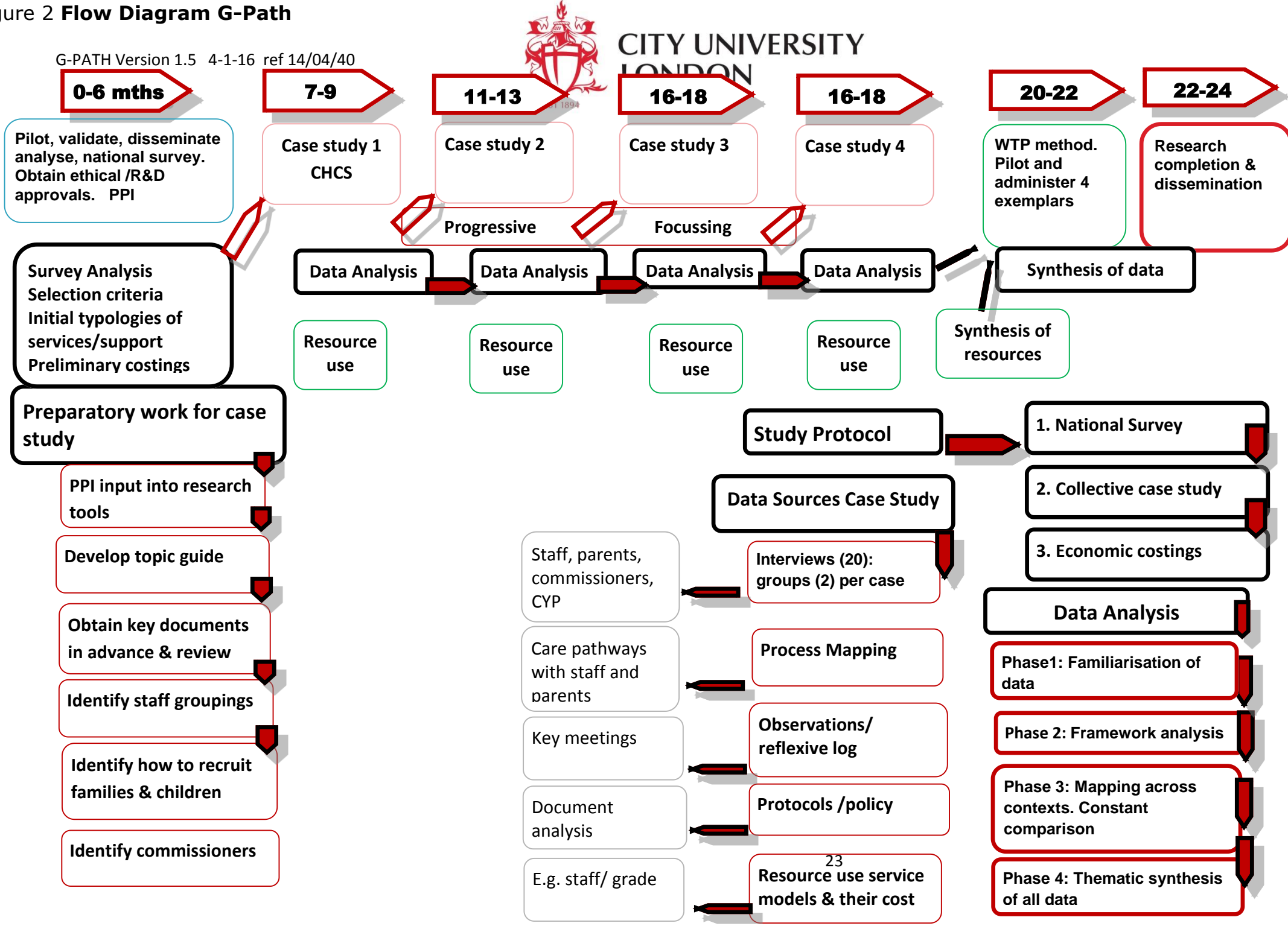
### **Project Advisory Group**

We have established a project advisory group of clinicians, therapists, service users and commissioners, in addition to the co-investigators, to input into all aspects of the research and address any challenges to help achieve milestones.

### **Dissemination of Research**

Dissemination outputs will include: a) service standards of support ; b) four exemplar models of support and “know- how” on the implementation of different best practice models in a range of contexts; and, c) an estimate of costs of each exemplar. In addition we will make recommendations for change and staff training. Dissemination will work on a number of levels including: dissemination workshops associated with the four study sites; service user fora; professional associations; academic articles in peer reviewed journals; and, academic conferences.

Figure 2 **Flow Diagram G-Path**



## NOTES

*‘Neurodisability describes a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations. A specific diagnosis may not be identified. Conditions may vary over time, occur alone or in combination, and include a broad range of severity and complexity. The impact may include difficulties with movement, cognition, hearing and vision, communication, emotion, and behaviour’ [Morris et al 2013  
<http://onlinelibrary.wiley.com/doi/10.1111/dmcn.12218/pdf>]*

## REFERENCES

- Allard A, Fellowes A, Shilling V, et al Key health outcomes for children and young people with neurodisability: qualitative research with young people and parents. *BMJ Open* 2014; 4:e004611. doi:10.1136/bmjopen-2013-004611
- Bridges, J.F Stated preference methods in health care evaluation: an emerging methodological paradigm in health economics. *Applied health economics and health policy*. (2003). 213-224. 2(4).
- Brotherton AM, Abbott J, Aggett PJ: The impact of percutaneous endoscopic gastrostomy feeding in children; the parental perspective. *Child Care Health Dev* 2007, 33:539–546.
- BSA. Statement of Ethical Practice for the British Sociological Association 2004. Available <http://www.britisoc.co.uk/media/27107/StatementofEthicalPractice.pdf?1410863413137> Accessed 14-08-2014
- Calderón C, Gómez-López L, Martínez-Costa C, Borraz S, Moreno-Villares JM, Pedrón-Giner C Feeling of burden, psychological distress, and anxiety among primary caregivers of children with home enteral nutrition. *J Pediatr Psychol*. 2011 Mar ;36(2):188-95. doi: 10.1093/jpepsy/jsq069. Epub 2010 Aug 18 .
- Calis EA, Veugelers R, Sheppard JJ, Tibboel D, Evenhuis HM, Penning C. Dysphagia in children with severe generalized cerebral palsy and intellectual disability. *Dev Med Child Neurol* 2008; **50**: 625–630.
- Campbell H , Hotchkiss R (1998) Integrated Care Pathways *BMJ* 1998;316:133
- Children and Young People’s Health Outcomes Strategy Report of the Children and Young People’s Health Outcomes Forum. (2012) Available [https://www.gov.uk/government/uploads/system/uploads/attachment\\_data/file/216852/CYP-report.pdf](https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/216852/CYP-report.pdf). Accessed 15-09-13
- Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M Developing and evaluating complex interventions: the new Medical Research Council guidance *BMJ*. 2008; 337: a1655. Published online 2008 September 29. doi: 10.1136/bmj.a1655
- Craig G, Higgs P. Risk owners and risk managers: dealing with the complexity of feeding children with neurodevelopmental disability. *Health Risk Soc* 2012; **14**: 627–637.
- Craig GM, Carr LJ, Cass H, Hastings RP, Lawson M, Reilly S *et al*. Medical, surgical, and health outcomes of gastrostomy feeding. *Dev Med Child Neurol* 2006; **48**: 353–360.
- Craig GM, Scambler G. Negotiating mothering against the odds: gastrostomy tube feeding, stigma, governmentality and disabled children. *Soc Sci Med* 2006; **62**: 1115–1125.
- Craig GM, Scambler G, Spitz L. Why parents of children with neurodevelopmental disabilities requiring gastrostomy feeding need more support. *Dev Med Child Neurol* 2003; **45**: 183–188.
- Craft A (2014) What exactly happened in the hospital treating Aysha King? *The Telegraph* 12 September
- Curtis L. Unit Costs of Health and Social Care. Personal Social Services Research Unit, University of Kent 2011
- Dahlseng, M.O., Andersen, G.L., DA Graca Andrada, M., Arnaud, C., Balu, R., DE LA Cruz, J., Folha, T., Himmelmann, K., Horridge, K., Juliusson, P.B., Pahlman, M., Rackauskaite, G., Sigurdardottir, S., Uldall, P., Vik, T. & on behalf of the Surveillance of Cerebral Palsy in Europe Network, Gastrostomy tube feeding of children with cerebral palsy: variation across six European countries, *Developmental Medicine & Child Neurology*, 2012, 54, (10):938-944.
- DFES Aiming high for disabled children: Better Support for families HM Treasury and Department for Education and Skills. 2007
- DH, The NHS Outcomes Framework 2013-14 (2013) Available [http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH\\_131700](http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_131700) Accessed 20-04-14
- DH. National service framework: children, young people and maternity services, 2004 Available <https://www.gov.uk/government/publications/national-service-framework-children-young-people-and-maternity-services> Accessed 12-09-13
- Emerson E Deprivation, ethnicity and the prevalence of intellectual and developmental disabilities. *J Epidemiol Community Health* (2012) 66:218–44.
- Enrione EB, Tomlinson B, Rubin A Medical and Psychosocial experiences of family caregivers with children enteral fed at home *Journal of Parenteral & Enteral Nutrition*, 2005 29 413-419



Faculty of Dental Surgery. Clinical Guidelines and Integrated Care Pathways for the Oral Health Care of People with Learning Disabilities. FDS:London 2012.

Ferluga ED, Archer KR, Sathe NA, Krishnaswami S, Klint A, Lindegren ML, McPheeters ML. Interventions for Feeding and Nutrition in Cerebral Palsy Comparative Effectiveness Review 2013, No. 94 AHRQ Publication No.13 Available. [www.effectivehealthcare.ahrq.gov/reports/final.cfm](http://www.effectivehealthcare.ahrq.gov/reports/final.cfm). Accessed 01-04-13

Gallagher S, Phillips AC, Carroll D.J Parental stress is associated with poor sleep quality in parents caring for children with developmental disabilities. *Pediatr Psychol*. 2010 Aug; 35(7):728-37. Epub 2009 Oct 29.

Gallagher S, Phillips AC, Drayson MT, Carroll D. Parental caregivers of children with developmental disabilities mount a poor antibody response to pneumococcal vaccination *Brain Behav Immun*. 2009 Mar; 23(3):338-46. Epub 2008 Jul 1.

Glover G and Ayub M (2010) How people with learning disabilities die. Published by Improving Health and Lives: Learning Disabilities Observatory [http://www.improvinghealthandlives.org.uk/uploads/doc/vid\\_9033\\_IHAL2010-06%20Mortality.pdf](http://www.improvinghealthandlives.org.uk/uploads/doc/vid_9033_IHAL2010-06%20Mortality.pdf)

Greenhalgh T, Kushner S and Russell J (eds) Case Study Evaluation: past, present and future challenges. *Advances in Program Evaluation*, Emerald. (forthcoming)

Greenhalgh T, & Swinglehurst D. Studying technology use as social practice: the untapped potential of ethnography *BMC medicine*, 2011 9:45 Available at <http://www.biomedcentral.com/1741-7015/9/45> Accessed 15-09-14

Greenhalgh. T. Primary Health Care, Theory & Practice (2007) Blackwell. Chapter 11

Greenhalgh T, Robert G, Macfarlane F, Bate P, Kyriakidou O, Diffusion of Innovations in Service Organizations: Systematic Review and Recommendations. *Millbank Quarterly* 2004:82

Ireys H, Chernoff R, DeVet KA, Kim Y. Maternal outcomes of a randomized controlled trial of a community-based support program for families of children with chronic illnesses. *Arch Pediatr Adolesc Med*.2001;155 :771– 777

Johnson, C.B. & Deitz, J.C. 1985, "Time use of mothers with preschool children: a pilot study", *The American Journal of Occupational Therapy : Official Publication of the American Occupational Therapy Association*, vol. 39, no. 9, pp. 578-583.

Kennedy. I Getting it right for children and young people: Overcoming cultural barriers in the NHS so as to meet their needs. 2010 Available <https://www.gov.uk/government/publications/getting-it-right-for-children-and-young-peopleovercoming-cultural-barriers-in-the-nhs-so-as-to-meet-their-needs>. Accessed 21-04-14

King's Fund (2008) Safer Births: Everybody's Business. An Independent Enquiry into the Safety of Maternity Services in England. London, Kings Fund. [[http://www.kingsfund.org.uk/research/publications/safe\\_births.html](http://www.kingsfund.org.uk/research/publications/safe_births.html)].

Kurien M, White S, Simpson G, Grant J, Sanders DS, McAlindon ME. Managing patients with gastrostomy tubes in the community: can a dedicated enteral feed dietetic service reduce hospital readmissions? *Eur J Clin Nutr* 2012 Jun;66(6):757-760. [Abstract]

Mahant S, Jovcevska V, Cohen E. Decision-making around gastrostomy-feeding in children with neurologic disabilities. *Pediatrics*, 2011, 127(6):e1471-81.

Mannion R, Davies H, Marshall MN, (2005) Cultural characteristics of "high" and "low" performing hospitals *Journal of Health Organization and Management* Vol. 19 No. 6, 2005 pp. 431-439

Marchant R, Lefevre M, Jones M, Luckock B (2007) SCIE Knowledge review 18: 'Necessary stuff' - The social care needs of children with complex health care needs and their families. Available <http://www.scie.org.uk/publications/knowledgereviews/kr18.asp> Accessed 08-01-14

Marmot M. Fair Society, Healthy Lives: Strategic Review of Health Inequalities in England post 2010. The Marmot Review, 2010.

[http://www.instituteofhealthequity.org/\(accessed 20.6.12\)](http://www.instituteofhealthequity.org/(accessed 20.6.12)).

McCarthy D, Mueller K. (2005) Organizing for Higher Performance: Case Studies of Organized Delivery Systems *Series Overview, Findings, and Methods* • July 2009. The Commonwealth Fund Pub. 1288, Vol. 21. Accessible from: [www.commonwealthfund.org](http://www.commonwealthfund.org)

McConachie, H.R. Conceptual frameworks in evaluation of multidisciplinary services for children with disabilities. *Child: Care, Health & Development*. (1999) 25: 2

McCourt C, Rayment J, Rance S, Sandall J. An ethnographic organisational study of alongside midwifery units: a follow-on study from the Birthplace in England programme. *Health Serv Deliv Res* 2014;2(7)

National Leadership and Innovation Agency for Healthcare (2005) Integrated care pathways: a guide to good practice :NLIAH.

Nuffield Trust 2008. Organising for Quality. A briefing paper. Nuffield Trust, London. Accessible from [www.nuffieldtrust.org.uk](http://www.nuffieldtrust.org.uk)

NHS Institute for Innovation and Improvement (2008) Process Mapping An Overview. Quality and Service Improvement Tools. [Available]

[http://www.institute.nhs.uk/quality\\_and\\_service\\_improvement\\_tools/quality\\_and\\_service\\_improvement\\_tools/process\\_mapping\\_-\\_an\\_overview.html](http://www.institute.nhs.uk/quality_and_service_improvement_tools/quality_and_service_improvement_tools/process_mapping_-_an_overview.html)

NHS Quality Improvement Scotland. Best Practice Statement (2007) Caring for children and young people in the community receiving enteral tube feeding Available

[http://www.healthcareimprovementscotland.org/previous\\_resources/best\\_practice\\_statement/caring\\_for\\_children\\_and\\_young\\_.aspx](http://www.healthcareimprovementscotland.org/previous_resources/best_practice_statement/caring_for_children_and_young_.aspx) Accessed 15-09-14

NHS Institute for Innovation and Improvement (2008) Process Mapping An Overview. Quality and Service Improvement Tools. [Available]





[http://www.institute.nhs.uk/quality\\_and\\_service\\_improvement\\_tools/quality\\_and\\_service\\_improvement\\_tools/process\\_mapping\\_-\\_an\\_overview.html](http://www.institute.nhs.uk/quality_and_service_improvement_tools/quality_and_service_improvement_tools/process_mapping_-_an_overview.html)

NICE. Cerebral palsy: the diagnosis and management of cerebral palsy draft scope for consultation

7 July to 4 August 2014 Available <http://www.nice.org.uk/guidance/gid-cgwave0687/resources/cerebral-palsy-sc-draft-scope2> Accessed 15-09-14

Ortlipp, M. Keeping and using reflective journals in the qualitative research process. *The Qualitative Report*, (2008) 13(4), 695-705. Retrieved from <http://www.nova.edu/ssss/QR/QR13-4/ortlipp.pdf>

Parkes J, Hill N, Platt MJ, Donnelly C. Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study. *Dev Med Child Neurol* 2010; **52**: 1113–1119.

Parlett, M., & Hamilton, D. Evaluation as illumination: A new approach to the study of innovatory programs: in *Occasional paper*, Centre for Research in the Educational Sciences, 1972. University of Edinburgh. Edinburgh.

Parr JR, Jolleff N, Gray L, Gibbs J, Williams J, McConachie H. Twenty years of research shows UK child development team provision still varies widely for children with disability. *Child Care Health Dev* 2013 Nov; **39**(6):903-907.

Pederson SD, Parsons HG, Dewey D. Stress levels experienced by the parents of enterally fed children. *Child Care Health Dev* 2004; **2**,30:507–513.

Pemberton J, Frankfurter C, Bailey K, Jones L, Walton JM Gastrostomy matters--the impact of pediatric surgery on caregiver quality of life. *J Pediatr Surg*. 2013 May; **48**(5):963-70.

Pilger D, Nguipdop Djomo P, Abubakar I, et al. BCG vaccination in England since 2005: a survey of policy and practice. *BMJ Open* 2012; **2**:e001303. doi:10.1136/bmjopen-2012-001303

Pope C and Mays N *Qualitative Research in Health Care*, John Wiley & Sons, 2013

Pope C. Conducting ethnography in medical settings. *Med Educ* 2005; **39**:1180-7.

Reason, P. & Bradbury, H., (Ed.) *The SAGE Handbook of Action Research. Participative Inquiry and Practice*. 1st Edition. London: Sage, 2001.

Reilly S, Wisbeach A, Carr L. Assessing feeding in children with neurological problems. In: Southall A, Schwartz A, (eds) *Feeding Problems In Children a Practical Guide*. Radcliffe Medical Press: Abingdon, Oxon, UK, 2000, pp 153–170.

Reilly, S., Skuse, D. & Poblete, X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: a community survey, *The Journal of Pediatrics*, 1996. **129**, (6): 877-882

Ritchie, J. & Spencer, L. Qualitative data analysis for applied policy research

In Bryman A. & Burgess R. G eds. *Analyzing Qualitative Data*. London: Routledge 1994:173-194.

Robson, C. *Real World Research: A Resource for Social Scientists and Practitioner Researchers*. (1993). Oxford, Blackwell.

Rowe, R. E., Garcia, J., Macfarlane, A. J. & Davidson, L. L. (2001) Does poor communication contribute to stillbirths and infant deaths? A review. *J Public Health*, **23**: 23-24.

Segar J, Checkland K, Coleman A, McDermott I. Thinking about case studies in 3-D: researching the NHS clinical commissioning landscape in England in Greenhalgh T, Kushner S and Russell J (eds) *Case Study Evaluation: past, present and future challenges*. *Advances in Program Evaluation*, Emerald.

Sharpe R Benfield G , Lessner E & DeCicco E. From: Scoping Study for the Pedagogy strand of the JISC e-Learning Programme. Available [www.jisc.ac.uk/.../scoping%20study%20final%20report%20v4.1.doc](http://www.jisc.ac.uk/.../scoping%20study%20final%20report%20v4.1.doc) Accessed 01-08-14

Sleigh G, Brocklehurst P. Gastrostomy feeding in cerebral palsy: a systematic review. *Arch Dis Child* 2004; **89**: 534–539.

Sloper, P. Models of service support for parents of disabled children. What do we know? What do we need to know? *Child: Care, Health & Development*, Mar1999, Vol. 25 Issue 2, p85, 15p

Spalding, K. & McKeever, P., "Mothers' experiences caring for children with disabilities who require a gastrostomy tube", *Journal of pediatric nursing*, 1998. **13**, 4: 234-243.

Stake RE. Qualitative case studies. In: Denzin NK, Lincoln YS, eds. *The Sage Handbook of Qualitative Research*. London: Sage, 2005:443-66.

Stake, R. E. (1981). The art of progressive focusing: in 65th Annual Meeting of the American Educational Research Association. Los Angeles.

Stake, R. E. *The art of case study research* (1995) Thousand Oaks: Sage Publications.

Stringel G, Delgado M, Guertin L, Cook JD, Maravilla A, Worthen H. Gastrostomy and Nissen fundoplication in neurologically impaired children. *J Pediatr Surg* 1989 Oct; **24**(10):1044-1048.

Sullivan PB, Juszczak E, Bachlet AM, Lambert B, Vernon-Roberts A, Grant HW. *et al*. Gastrostomy tube feeding in children with cerebral palsy: a prospective, longitudinal study. *Dev Med Child Neurol* 2005; **47**: 77–85.

Sullivan, P Impact of gastrostomy tube feeding on the quality of life of carers of children with cerebral palsy, *Developmental Medicine & Child Neurology*, 2004, **46**, (12):796-800.

Swinglehurst D, Greenhalgh T, Russell J, Myall M. Receptionist input to quality and safety in repeat prescribing in UK general practice: ethnographic case study. *BMJ* 2011 Nov **3**;343:d6788.

Symon G & Cassell C . *Qualitative Organizational Research: Core Methods and Current Challenges*. London Sage 2012

Tambor M, Pavlova M, Rechel B, Golinowska S, Sowada C, Groot W. (2014)

*SocSci Med*. Sep; **116**:193-201. doi: 10.1016/j.socscimed.2014.07.009. Epub 2014 Jul 5.





Willingness to pay for publicly financed health care services in Central and Eastern Europe: Evidence from six countries based on a contingent valuation method.

Thorne SE, Radford MJ, McCormick J. The multiple meanings of long-term gastrostomy in children with severe disability. *J Pediatr Nurs* 1997 Apr;12(2):89-99

Vanhaecht, K., De Witte, K. Sermeus, W. (2007). The impact of clinical pathways on the organisation of care processes. PhD dissertation KULeuven, 154pp, Katholieke Universiteit Leuven.

Vitae/Careers Research and Advisory Centre (CRAC) (2010). Introducing the Researcher Development Framework. [Available] <https://www.vitae.ac.uk/vitae-publications/rdf-related/introducing-the-vitae-researcher-development-framework-rdf-to-employers-2011.pdf> Accessed 11-03-15.

Yin, R.K. 2003 *Case Study Research, Design and Methods*. London: Sage