DOCTOR OF PHILOSOPHY

Sleep hygiene education and children with developmental disabilities

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Award date:
2017

Awarding institution:
Bangor University

[Link to publication](#)

Sleep hygiene education and children with developmental disabilities.

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2017.

Thesis submitted in fulfilment of the requirements for the Degree of Doctor of

Philosophy in the University of Wales, (12.6.17)

DECLARATION

This work has not been previously been accepted in substance for any degree and is not being concurrently submitted in candidature for any degree.

Signed .... (candidate)

Date   ....

STATEMENT 1

This thesis is the result of my own investigations, except where otherwise stated.

Other sources are acknowledged by footnotes giving explicit references. A bibliography is appended.

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DEDICATION

I dedicate this study to families of children with developmental disabilities who can struggle every day and night with sleep deprivation. I hope these findings help in some way to improve other people's understanding of your experiences and the quality of support you receive.

ABSTRACT

Sleep hygiene education (SHE) is a first-line treatment for behavioural sleep

problems that affect up to 80% of children with developmental disabilities (DD).

SHE involves advising parents on behaviours which promote quality sleep in

children; however, it is an inadequately conceptualised intervention currently

supported by theories based on popular wisdom. Professionals who support families

of children with DD and sleep problems have routinely implemented SHE for years

without explicit explanation of what the intervention is supposed to achieve. For

these families, severe and enduring sleep problems are a daily reality, placing

increased impetus on the need for evidence based and transparent SHE interventions.

This is also reflected in the current prudent healthcare agenda in the UK, which

obliges professionals to only deliver care which is relevant to individual need. The

aim of this thesis is therefore to develop a systematic understanding about what SHE

does, how it is delivered and how it is supposed to work to improve sleep. Through a

hybrid of deductive and inductive approaches, an evidence based SHE tool and

nuanced programme theory is developed, which helps communicate the essentials of

SHE. Firstly, a scoping review investigates what is known about SHE and findings

inform the design of an inductive programme theory development phase underpinned

by a participatory methodology. This incorporates an exploratory study examining

the perceptions of parents and professionals about SHE through qualitative

interviewing. Stakeholders then deliberate emerging themes in a co-design study and

cumulative findings are synthesised to develop the main outputs of this thesis: a SHE

tool underpinned by a programme theory. An abstraction to mid-range change

theories demonstrates how the programme theory presents plausible solutions to sleep problems and the original contribution of an explicit understanding of the nature of the complexity embedded in a SHE intervention for children with DD is presented. The findings indicate how the legitimation of sleep problems is a foundation on which SHE implementation should be based. It also demonstrates a greater understanding of the nature of customisation, knowledge exchange, health expectation and impact of rationing and gaming on implementation success. Implications for policy and practice include a more explicit understanding of what SHE is and how best to deliver it and for research, a theory-driven framework for evaluating SHE.

CHAPTER 1: INTRODUCTION

1.1 Introduction

This thesis aims to develop the evidence base supporting sleep hygiene education (SHE) as a treatment for behavioural sleep problems in children with developmental disabilities (DD), and presents findings from three studies. The first study was undertaken to identify the body of literature pertaining to SHE by conducting a scoping review. The second study was an exploratory study which aimed to address gaps in qualitative evidence identified in the review. The exploratory study aimed to explore the experiences of parents and sleep practitioners (professionals who support families with children's sleep problems) of SHE through a series of qualitative interviews. Data were thematically analysed into parent and sleep practitioner (stakeholder) themes. The third study was a co-design study which synthesised scoping review findings and exploratory study stakeholder themes into overarching discussion themes. These discussion themes were used to guide debate between parents and practitioners as equal research partners in a series of three co-design events. Co-designers were also invited to explore the acceptability of SHE advice identified in the scoping review. The co-design study culminated with a synthesis of review, exploratory and co-design study findings and development of a SHE tool for children with DD underpinned by programme theory. Figure 1 shows an outline of the studies presented in this thesis.

Figure 1-Overview of studies.


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                    ┌─────────────────────┐
                    │   Scoping review    │
                    └─────────────────────┘
                       ↙             ↘
┌─────────────────────────┐   ┌─────────────────────────┐
│   Exploratory study     │   │   Exploratory study     │
│ 1st round of parent     │   │ 1st practitioner focus  │
│ interviews              │   │ group                   │
└─────────────────────────┘   └─────────────────────────┘
            ↓                            ↓
┌─────────────────────────┐   ┌─────────────────────────┐
│   Exploratory study     │   │   Exploratory study     │
│ 2nd round of parent     │   │ 2nd practitioner focus  │
│ interviews              │   │ group                   │
└─────────────────────────┘   └─────────────────────────┘
              ↘                  ↙
            ┌─────────────────────────┐
            │  Co-design study event 1│
            │     (parent only)       │
            └─────────────────────────┘
                        ↓
            ┌─────────────────────────┐
            │  Co-design study event 2│
            │ (Joint parent /practitioner)│
            └─────────────────────────┘
                        ↓
            ┌─────────────────────────┐
            │  Co-design study event 3│
            │ (Joint parent /practitioner)│
            └─────────────────────────┘
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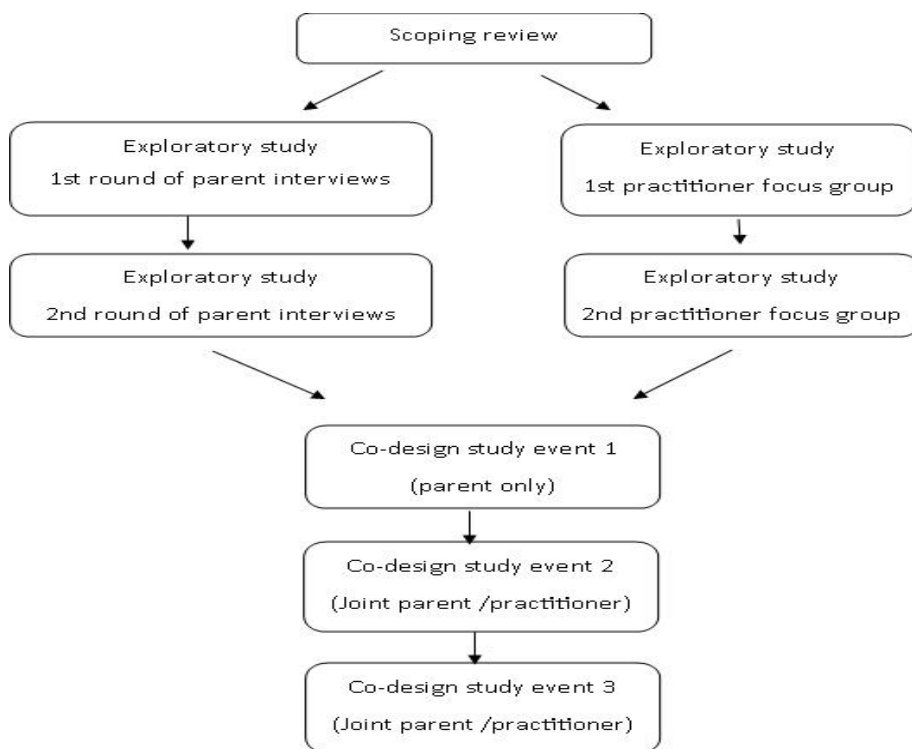
The Introduction chapter will introduce the background to the thesis and present a rationale for the need to study SHE and children with DD. SHE and DD are conceptually defined, and the researcher's professional background in relation to SHE is outlined. The role of sleep assessment in identifying behavioural sleep problems is described and existing theoretical underpinnings of SHE are explored. What we need to know to advance the topic of SHE is summarised and the research aim and objectives are defined. The policy landscape around disability and sleep is reviewed to clarify the political arena in which SHE is currently delivered. The chapter concludes by outlining subsequent chapters.

1.2 Sleep Problems in Children with DD

The physical, social and psychological effects of sleep problems on children's well-being are well documented and include compromised growth, immune system health, cognitive performance and behaviour problems (Bonuck & Grant, 2012; Durand, 2013b; Galland & Mitchell, 2010). Childhood sleep problems typically include difficulties with sleeplessness, sleeping too much or bizarre experiences related to sleep (Stores, 2009). Such problems are common in children with estimates of between 25-50% of the general population being affected (Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2012; Goldman et al., 2009; Schreck & Richdale, 2011). This figure rises to 80% in children with developmental disabilities (DD) (Table 1) (Jan et al., 2008; Richdale & Wiggs, 2005) who are more prone to developing severe and persistent sleep problems for reasons such as neurological damage, associated respiratory conditions and communication problems (which can prevent children from learning bedtime routines and how to sleep independently) (Bounck & Grant, 2012; McDaid & Sloper, 2008). Such an increased incidence of sleep problems highlights double jeopardy for children with DD who already face daily challenges in reaching their developmental potential (Wiggs & Stores, 2001). In addition, of further concern is the impact that sleep problems has on the health and well-being of parents and siblings; these problems include heightened stress, relationship breakdown and employment difficulties (Contact a Family, 2009; Family Fund, 2013; Handsel Trust, 2007).

1.3 Operational Definition of Developmental Disabilites (DD)

It is acknowledged that various developmental disability definitions exist such as Neurodevelopmental Disorder (The American Psychiatric Association, 2013),

Intellectual Disabilities (American Association on Intellectual and Developmental Disabilities, 2013) and the International Classification of Disease (ICD) version 10 (2016) Code for Disorders of Psychological Development. However, on the basis that some of these originate from America or offer narrower classifications which eliminate specific conditions, the following DD definition tailored to the needs of this UK based study will be used (Table 1). This has been specifically developed for the purposes of this study in collaboration with the supervisory team and encompasses a variety of conditions.

Table 1. Operational definition of developmental disabilities (DD).

Conditions originating in the developmental period (0-18 years) including specific learning difficulties, global developmental delay and disorders of psychological functioning such as Autistic Spectrum Disorder (ASD) (encompassing Autism and Asperger syndrome). Excludes children with a sole diagnosis of physical disabilities.

1.4. Interventions for Sleep Problems

A variety of interventions are used to treat behavioural sleep problems that are linked to parental management issues or poor boundary setting (McDaid & Sloper, 2009) in children with DD. These include first-line pharmacological treatments such as melatonin, based on the sleep inducing neuro-hormone produced at night which also regulates sleep/wake circadian rhythms (Phillips & Appleton, 2004) (biological changes that follow a 24-hour cycle). Behavioural interventions are also available such as sleep hygiene education (SHE), graduated extinction and sleep restriction. SHE is typically used as a first-line intervention and aims to improve children's sleep, through educating parents about positive sleep behaviours and habits that promote healthy

sleep. (Vriend, Corkum, Moon & Smith, 2011). SHE is recommended prior to implementing more specific interventions such as graduated extinction and sleep restriction, because SHE aims to create fundamental, optimal environmental conditions conducive to sleep (Durand, 2013b). Graduated extinction is a strategy whereby the child's protestations at bedtime are ignored and interspersed with intervals of parental checking (Durand, 2013b). Sleep restriction involves restricting the child's sleep to 90% of normal sleep length and altering the timing of bedtimes, to make the child tired enough for sleep (Richdale & Wiggs, 2005). The common first-line interventions for sleep problems; pharmacological treatments and SHE, will now be outlined in 1.4.1 and 1.4.2.

1.4.1. Pharmacological treatments.

Pharmacological treatments that improve sleep include hypnotic medications such as Benzodiazepines or Antihistamines which have a sedative effect on children's sleep. Synthetically produced, exogenous melatonin is, however, the most commonly used drug to treat chronic sleep problems in children with DD (DeLeersnyder, Zisapel, & Laudon, 2011) and short term use (< 3 months) is well evaluated in recent evidence reviews (Blackmer & Feinstein, 2016; Hollway & Aman, 2012; Sajith & Clarke, 2007). Melatonin acts through regulating the sleep-wake cycle and is given at bedtimes to boost natural levels of melatonin secretion at night (Blackmer & Feinstein, 2016). It is acknowledged that melatonin has a relatively small side effect profile, is effective in improving sleep onset latency (time taken to fall asleep) in children with DD and is widely used to treat sleep problems in children (Appleton et al., 2012). Although melatonin is a commonly used intervention to treat sleep problems in children with DD; this will not be the focus of this present research. The author's

interest lies in developing the first-line behavioural intervention of SHE, which ultimately aims to ensure the child has optimal conditions for sleep before pharmacotherapy is tried.

1.4.2. Sleep hygiene education (SHE).

The focus of this research is on the behavioural intervention of sleep hygiene education (SHE) which advises parents on creating optimal sleeping conditions for their child. SHE is defined as "*A set of sleep-related behaviours that expose persons to activities and cues that prepare them for and promote appropriately timed and effective sleep*" (Jan et al., 2008, p.1344). Typical SHE advice components include creating consistent bedtime routines, avoiding caffeine, altering sleep timings and encouraging daytime exercise. SHE is accepted as an essential first step in the treatment of behavioural sleep problems in children with DD (Blackmer & Feinstein, 2016; Durand, 2013b). It has also been highlighted as a first approach to sleep problems in children with ASD in best practice guidance issued by the UK's National Institute for Health and Clinical Excellence (NICE, 2013). However, Jan et al. (2008) and Vriend et al. (2011) emphasise that SHE is commonly implemented in combination with other behavioural strategies, implying that although SHE is a necessary primary step, it is insufficient as a stand alone intervention. This is reflected in the majority of sleep intervention evaluative research studies, which are largely restricted to multi-intervention studies that incorporate SHE as an adjunct treatment for children with DD (Vriend et al., 2011; Stores & Stores, 2004). Notwithstanding, if SHE is implemented inadequately, subsequent interventions often fail or are less effective (because the basic conditions for sleep such as a darkened room or comfortable bed have been overlooked) (Vriend et al., 2011). Furthermore Jan et al.

(2008) claim that practitioners (professionals who support families with children's sleep problems) often lack the expertise to advise on SHE and call for more evidence based studies exploring the nature of this intervention with DD. The next section explores the researcher's professional background in relation to SHE and children with DD.

1.5. Researcher's Professional Background

The concept for this thesis emanated from the researcher's professional background as an NHS Community Children's Learning Disability Nurse and Sleep Practitioner trainer for the Handsel Project/ Team Around the Child (TAC) Interconnections[1]. The researcher's previous Community Nursing role involved assessing the sleep problems of children with DD's and for those identified as behavioural in cause, supporting parents to implement SHE in their own home. Advice took the form of a standard SHE leaflet for children with DD (developed by the clinical team) based on best practice principles (Durand, 1998). However, families commonly reported the advice to be ineffectual and simplistic, and further behavioural interventions such as graduated extinction (section 1.4.) were often necessary to improve the child's sleep. This experience prompted the researcher to further question SHE as a credible first-line intervention for behavioural sleep problems in children with DD.

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[1] The Handsel Project/ Team Around the Child (TAC) Interconnections is an organisation working to build knowledge around children with disabilities and hosts 'Sleep Practitioner' training for health and social care professionals.

Questions about the impact of SHE were also raised through the researcher's current Sleep Practitioner Trainer role, which involves training health, social care and voluntary sector professionals to support families of children with DD and sleep problems. This highlighted the existence of a myriad of ways in which SHE is delivered across the UK, ranging from referring parents to freely downloadable SHE leaflets on the internet, to delivering advice in group-based parenting workshops. Sleep assessment preceding this advice is usually minimal and often non-existent, which brings into question the appropriateness of the SHE advice being given. Furthermore, in these training events, anecdotal evidence suggests that the support parents receive to implement SHE is commonly perceived as being superficial. Such a notable mix in delivery method, concerns around intervention effectiveness and widely accepted importance of SHE as a first-line intervention, evidence the need for further inquiry and enhanced understanding of SHE.

1.6. Recent Reviews of SHE and Children with DD

This research began with a search for evidence based reviews performed on the 11/10/2012. This revealed three studies which focused on behavioural sleep interventions in general and described SHE as an adjunct intervention for children with DD (McDaid & Sloper, 2009; Richdale & Wiggs, 2005; Vriend et al., 2011).

In Richdale and Wiggs's (2005) review of 22 studies, the importance was highlighted of sleep assessment in identifying sleep problem causes, to ensure the application of appropriate interventions. Extinction (like graduated extinction (section 1.4.) minus the checking intervals) had the most empirical support, and the SHE elements of advising on a regular bedtime routine and setting a fixed bedtime featured in most studies. Authors concluded that more research was needed comparing bedtime routine

advice with other interventions, clearly described and supported by treatment manuals. These findings support the view that sleep assessment is an integral part to any intervention and evaluative research should be explicit in describing the interventions being tested. However, this review only focused on the bedtime routine and fixed bedtime elements of SHE and did not present a more comprehensive range of possible SHE advice components (such as exercise, diet and sleep environment, for example). In addition, reference was not made to the need to develop a deeper understanding or theory of how interventions work to improve sleep. This indicates the need for further research which establishes the component parts of SHE and systematically develops an understanding or theory of how SHE works to enhance sleep.

McDaid and Sloper's (2009) rapid review of 19 studies, also reported sleep assessment as a prerequisite to any intervention and how empirical research largely evaluated behavioural interventions combined with the positive bedtime routine advice component of SHE. It was not always clear from evaluating multi-component interventions, which aspects of the intervention in terms of specific SHE advice, and contextual factors such as level of practitioner support, resulted in improved sleep. To develop this understanding, authors called for further research that examines parents' and practitioners' real life experience of sleep interventions. This evidences the need to develop an explicit understanding or theory of how SHE is supposed to work to achieve the desired outcome of improved sleep.

Vriend et al's. (2011) review of 15 ASD studies, reported more detailed SHE interventions including advice on bedtime routines, daytime and night-time habits and behaviour management. SHE was largely evaluated in combination with graduated extinction or sleep restriction and reported as a necessary but insufficient component

to any sleep intervention. Authors emphasised that SHE advice should be adapted to meet children's individual needs and how inadequate implementation could lead to unsuccessful or less effective subsequent interventions. This builds the evidence base in support of the development of a SHE tool for children with DD and underpinning theory explaining how correct implementation leads to improved sleep. Furthermore, whilst review findings are significant for ASD this prompts the need for a broader review of SHE and with children who have other disability conditions.

In summary, reviews to date have highlighted the importance of sleep assessment prior to the implementation of behavioural sleep strategies. They have focused on behavioural interventions in general for treating sleep problems in children with DD, but have only mentioned SHE as a stand-alone intervention in brief. Reviews have been limited in their definitions of SHE and descriptions of SHE components. Furthermore, there has been no exploration of underlying theory describing how SHE is supposed to work. These gaps in the evidence base support the need for further inquiry into SHE as a first-line sleep intervention.

1.7. Sleep Assessment

As these evidence based reviews highlight, the importance of assessing sleep problems prior to sleep intervention should not be under-estimated. This is also emphasised by Malow et al. (2013) and Stores and Wiggs (2001). This evidence advises sleep practitioners to assess sleep problems and identify possible causes to ensure that the most appropriate advice is given. An example taken from the researcher's professional experience illustrates this further: if SHE is applied to sleep problems triggered by breathing difficulties (which have been overlooked), it is unlikely that the advice will work to improve sleep, because surgical intervention to remove any obstruction may

have been the most appropriate treatment. However, the researcher's practice experience has shown that sleep assessment is rarely completed well and its fundamental function of correct treatment selection is commonly overlooked.

According to Stores (2009), the journey towards an improved understanding of a child's sleep problem and effective intervention, commonly begins with parents reporting a set of sleep symptoms: sleeplessness (problems falling asleep, night waking and early waking), sleeping too much or disturbed episodes interfering with sleep. When the process of sleep assessment is initiated by a professional; sleep history questionnaires such as the Modified Simmonds and Parraga Sleep questionnaire (MSPSQ) (Maas et al., 2011) and various other sleep outcome measures can be used. Such measures include, for example, parent completed sleep dairies, the Child Sleep Habits Questionnaire (CSHQ) (Johnson et al., 2012) or Actigraphy which is a mechanical device worn on the child's wrist to monitor basic sleep patterns (Sitnick, Goodlin-Jones & Anders, 2008). Sleep history questionnaires enquire about the nature and longevity of the sleep problem and establish what has already been tried to address it. Sleep outcome measures quantify the sleep problem at the outset and can also be used to measure effectiveness post intervention. According to the researcher's practice experience, when used in combination, these assessment techniques can contribute to an in-depth understanding of the sleep problem and conceivable causes.

1.7.1 Identifying behavioural sleep problems.

Sleep problem causes are often multiple and include everyday social or psychological factors which may warrant the involvement of additional support services, or medical conditions known to affect sleep such as epilepsy or asthma which require appropriate treatment (Richdale & Wiggs, 2005). It is important that all possible causes are

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identified prior to designing an appropriate sleep intervention (Malow et al., 2013). Where sleep problems are linked to parental management issues or poor boundary setting, these are sometimes informally referred to as behavioural sleep problems (McDaid & Sloper, 2009) and are best treated with behavioural interventions (Richdale & Wiggs, 2005) such as SHE, graduated extinction or sleep restriction (Section 1.4.)

In some instances, behavioural sleep problems will be given a sleep disorder diagnosis of insomnia by a medical professional (Durand, 2013b). The International Classification of Sleep Disorders (3rd edition) produced by the American Academy of Sleep Medicine (AASM) (2014)) defines insomnia as "*A persistent difficulty with sleep initiation, duration, consolidation, or quality that occurs despite adequate opportunity and circumstances for sleep, and results in some form of daytime impairment*" (p 19)

The AASM (2014) nosology describes how insomnia in children is often due to behavioural issues such as negative sleep associations or poor behavioural limit setting. Problems with sleep associations arise when a child depends on certain circumstances or special objects to help them fall asleep and return to sleep after a night-time awakening. When the child is denied these sleep associations or special conditions, this produces anxiety and problems with falling asleep at the scheduled time. Limit setting problems occur when the child persistently stalls bedtimes and refuses to fall asleep, which is reinforced by the parent's poor boundary setting.

It is important to be aware that over 80 types of sleep disorder exist including circadian rhythm sleep disorders (sleeping well but at the wrong times) and hypersomnolence (sleeping too much) (AASM, 2014). The symptoms of these

differing conditions are complex and wide ranging (Richdale & Wiggs, 2005). Furthermore, they all require different interventions, evidencing why a comprehensive sleep assessment must be completed prior to any sleep intervention, to ensure they are not overlooked and remain untreated. It is also important to note that in the literature, the terms sleep problem and sleep disorder are commonly used interchangeably (Stores 2009) and clear distinctions are rarely made. Therefore, for the purposes of clarity, the term 'behavioural sleep problems' will be used throughout this thesis. This will to refer to sleep problems which have been assessed and evidence found of limited parental boundary setting and the child's autonomous control over behaviour at sleep times (McDaid & Sloper, 2009).

1.8. Theoretical Underpinnings of SHE

As the evidence based reviews (Section 1.6.) have shown, existing theory supporting SHE is scarcely quoted in the literature and attempts at defining it could be described as generic and poorly articulated. In their practitioner review Jan et al. (2008), propose that SHE might work by entraining intrinsic circadian rhythms, behavioural conditioning, reducing anxiety and environmental stimulation and relaxation. Similar theories exist explaining why SHE improves sleep in typically developing children (Galland & Mitchell, 2010). The assumptions on which these are based are not made explicit and nor are any links made between programme activities and outcomes. Furthermore, the way in which these theories have been developed and tested is not described. This indicates the need to explore the evidence base around SHE theory in a scoping review and systematically develop a more explicit understanding of how the intervention is supposed to work.

1.8.1. Programme theory development.

The importance of establishing theory underpinning interventions or programmes of work is well documented (Astbury & Leeuw, 2010; Chen, 2015). Scholars in the field of programme evaluation define programme theory as the articulation of how and why interventions work (or fail) in varying contexts and identification of causal links between programme activities and outcomes. Such an explicit understanding of interventions is needed, so that a distinction can be drawn between implementation failure (the intervention is performed incorrectly), or theory failure (intervention performed correctly, but was still unsuccessful) (Funnell & Rogers, 2011).Without this, interventions risk being ineffective in achieving outcomes without stakeholders really understanding why. Similarly, the reasons why interventions work well may elude stakeholders in the absence of supporting theory, making it difficult to pinpoint and repeat elements of success. Furthermore, as Funnell and Rogers point out, it is common for intervention providers to have a poor understanding of how interventions bring about change and to engage in uninformed implementation for years.

The evidence thus far describes SHE as an important but poorly conceptualised intervention in need of further development. In essence what we need to know to advance this topic is an explicit understanding of what SHE is, how and why it is supposed to work and the best way to deliver it, to ensure it can be replicated effectively and evaluated reliably in a theory-driven manner. This describes the development of an explicit programme theory supported by a SHE intervention tool for children with DD.

Funnell and Rogers (2011) suggest that a more detailed understanding of interventions can be developed and presented in the form of a purposeful programme theory which

consists of a 'theory of change' and *'theory of action'*. A theory of change identifies the core processes by which change occurs and can derive from tacit beliefs about how interventions work or more evidence based change theories such as empowerment theory (Perkins & Zimmerman, 1995) which purports that change occurs through a process of people gaining mastery and control over their lives. A theory of action focuses on how an intervention's activities stimulates these theories of change, such as how parent buddies might help a parent change their perceptions of their child's sleep problems and help them accept SHE advice. The researcher felt that purposeful programme theory development would facilitate a fresh consideration of the familiar intervention of SHE and provide a useful framework to improve understanding about what makes it work to improve sleep.

1.8.2. Programme theory and the UK's Medical Research Council.

Adopting programme theory development as a framework to guide this research thesis is also aligned with Medical Research Council (MRC) guidance for evaluating complex interventions (2000, 2008). The framework advises researchers to systematically attend to several phases of study: development, feasibility, evaluation and implementation prior to intervention evaluation. Furthermore, it recommends that the development phase should include a commitment to identifying the evidence base and developing a sound theoretical comprehension of how an intervention can effect change (Craig et al., 2008). Therefore the present research begins with a scoping review to explore current SHE literature, followed by an exploratory study and finally a co-design study to systematically develop a SHE tool and underpinning programme theory as advised by Funnell and Rogers (2011) and the MRC (2000 , 2008).

1.9. Research Aim and Objectives

The initial search for evidence underpinning SHE, indicated the need for further specific inquiry into this as a stand-alone intervention for treating behavioural sleep problems in children with DD. Primarily it showed how there is a need to clearly define SHE and establish advice components. In addition the search called for a better understanding of how SHE works to improve sleep, through exploring parents' and practitioners' experiences and views on how the intervention should be delivered. This leads on to the research aim and objectives:

Research aim:

The aim of this research is to advance the knowledge base underpinning SHE as a treatment for behavioural sleep problems in children with DD.

Research objectives:

- To design an evidence based, systematically developed SHE tool to treat behavioural sleep problems in children with DD. The tool will be developed through performing a scoping review of the evidence and exploring stakeholder views and levels of agreement on SHE advice components in a co-design study.
- To develop a systematic and nuanced understanding or programme theory of how SHE is supposed to work to improve sleep in children with DD. This will be developed through performing a scoping review, exploring stakeholder perceptions of SHE in an exploratory study and co-design process involving parents and practitioners. Cumulative study findings will be finally synthesised in a co-design study to produce the programme theory.

1.10. The Policy Landscape

It is also important to explore the policy landscape in which SHE is delivered to place the need for this research in context. Ensuring children with DD receive early interventions leading to better health outcomes is cited as a priority in NHS England's (2016) business plan and similar commitments have been made in recent national policy documents: *National Service Framework for Children* (Department of Health, 2004), *Aiming High for Disabled Children* (Department for Education and Skills, 2007) and *Transforming Care- Next Steps* (Department of Health, 2015). In Wales, broad commitments have been made to improve the health of all children in *Together for Health* (Welsh Assembly Government (WAG), 2011), *Building a Brighter Future* (WAG, 2013) and more specifically for DD: *National Service Framework for Children, Young People and Maternity Services in Wales*, (WAG ,2005), *The Journey So Far,* (WAG, 2011) and *Transforming Care in Wales for People with Learning Disabilities and Challenging Behaviour* (All Wales Challenging Behaviour Community of Practice, 2013). It is posited that this broad commitment to enhancing children's care incorporates the appropriate treatment of sleep problems, which supports the need for this research.

In specific relation to sleep and children with DD, the voluntary sector has produced various reports. These have been seminal in raising awareness of sleep problems and evidence the need for further inquiry into effective interventions. Publications such as *Sleep. What's That?* (Handsel Trust, 2007), *What Makes My Family Stronger* (Contact a Family, 2009) and *Tired all The Time* (Family Fund, 2013) highlight the prevalence and severity of sleep problems and describe the negative impact on parents' mental health, relationships and employment. Sleep problems in children with ASD are also

emphasised in *The NICE Guideline On The Management and Support of Children and Young People on the Autistic Spectrum* (NICE, 2013), which makes specific research recommendations for randomised controlled trials to compare "*sleep hygiene interventions*" (p732) with the use of melatonin. However, the NICE guidance does not make it clear what the SHE intervention looks like or how it should be delivered. Being explicit about what an intervention involves prior to evaluative research, is necessary to ensure the correct interpretation of results (Funnell & Rogers, 2011) and illustrates a further reason for improving understanding around SHE and the need for this research project.

1.10.1. The prudent healthcare agenda.

When considering SHE as a clinical intervention potentially delivered by health practitioners in the NHS, it is important to acknowledge the political backdrop of prudent healthcare. This is espoused by Bradley and Willson (2014), *The 1000 lives +* initiative (NHS Wales. Public Health Wales, 2015) and UK policy which acknowledges the current financial strain of the NHS: *the Five Year Forward View* (NHS England, 2014). In a cost-conscious economy the principles of prudent healthcare extol the virtues of understanding what makes interventions work, being more conscious of the care that is delivered and ensuring patients receive the minimum appropriate intervention to achieve health outcomes. This may involve doing some things less and avoiding unnecessary interventions, which will help extract greater value from resources spent on healthcare. The researcher's professional experience has shown that although SHE is universally delivered, there is a lack of awareness and agreement about how it is supposed to work and how best to implement it. In line with

the principles of prudent healthcare, further inquiry is therefore needed to ensure practitioners are using SHE in the most effective way.

1.10.2. Economic costs.

More generally the current economic costs to the NHS of supporting families with children who have sleep problems have been found to be considerable. In a cost burden analysis Morris, St James-Roberts, Sleep, and Gilham (2001) investigated how much is invested in health professional contact time (home visits, telephone support) related to infant crying and sleeping problems in the first 12 weeks of life. The study estimated £65 million per annum NHS spend on total health practitioner contacts in relation to these early childhood sleep problems. Although this study only pertains to infant sleep difficulties, it is helpful in giving some indication of the costly nature of supporting families of children with DD whose sleep problems can often be severe and life-long.

It is also noted that pharmacological interventions require substantial financial investment. In relation to the commonly used hypnotic drug: melatonin, cost data specific to age and type of use was not available. However, cost analysis data shows an annual spend on dispensed melatonin in Wales of £345,693.42 in 2015 (NHS Wales, 2015). Comparable figures on dispensed melatonin in England were not available, however £22.6 million was spent on prescribed melatonin in England in 2015 (Prescquipp, 2015). Although these figures do not provide a precise breakdown of actual melatonin spend for sleep problems in children with DD, they do give some indication of the financial costs of utilising a pharmacological approach. Therefore, the costs of supporting with families with sleep problems are significant and it is important to ensure interventions are both effective and prudent.

1.11. Structure of the Thesis

<u>Chapter 2.</u> This reports on the developmental phase of study: a scoping review to map the literature pertaining to SHE. It highlights what is already known about SHE to inform the development of a SHE tool and underpinning programme theory. Findings also inform the design of stakeholder questioning in the exploratory study.

<u>Chapter 3.</u> This reports the methodological underpinnings of the exploratory and co-design studies. It explores qualitative design issues, data collection methods and data analysis approaches. It concludes with a summary of the design of the studies presented in this thesis.

<u>Chapter 4</u> This reports the methods and findings from the exploratory study. This study aims to build on scoping review findings and explore stakeholder (parent and practitioner) perceptions of SHE through a series of qualitative interviews. It concludes with a summary of the themes developed and synthesis of data sets, which inform the subsequent co-design study.

<u>Chapter 5</u> This reports the methods and findings of the co-design study. This study invites stakeholders to consider discussion areas developed from the exploratory study themes, in a series of three events and co-create a SHE tool underpinned by programme theory. Preliminary analytical links are made with mid-range theories of change and novel explanatory themes which make explicit the complexity of SHE and this chapter concludes with a presentation of a co-designed SHE tool for children with DD.

<u>Chapter 6.</u> This chapter describes the synthesis of findings from the scoping review, exploratory and co-design studies and methods employed to develop a programme

theory and logic model which explains how SHE is supposed to work to improve sleep. This chapter concludes with the presentation of a logic or conceptual model of the combined theory of change and action (programme theory) which also represents emerging mid-range and novel theoretical underpinnings.

Chapter 7.

This chapter begins with a discussion of how the systematically developed SHE programme theory relates to mid-range theories of change. A discussion follows of how the developing SHE programme theory links to the underpinning novel analytical themes which represent this study's original contribution to knowledge: an explicit understanding of the complexity embedded in an SHE intervention. Most importantly, this study shows how the legitimation of sleep problems is a foundation on which SHE implementation should be based. Findings also demonstrate a greater understanding of the nature of customisation, health expectation, knowledge exchange and impact of rationing and gaming on implementation success. These themes are discussed in relation to existing evidence and broader theoretical perspectives. Implications for practice, policy and research are also outlined. Presented next is a section on reflexivity to show how the researcher's learning developed throughout the study and PhD process.This is followed by study limitations and a critical appraisal of thesis findings. This chapter then concludes with some final remarks on this study's overall findings and original contribution to knowledge.

CHAPTER 2: SCOPING REVIEW

2.1. Introduction

The Medical Research Council (MRC) framework for evaluating complex interventions (2000, 2008) recommend that intervention development should begin with an understanding of the existing evidence ideally collated in a literature review. A review is also suggested by Funnell and Rogers (2011) as a preliminary stage in programme theory development to identify the nature and contributing factors of the problem the intervention seeks to address. This chapter reports the findings of a scoping review which explores what is known about SHE as a treatment for behavioural sleep problems in children with DD and begins with a rationale for this choice of review method. Arksey and O'Malley's (2005) methodological framework is presented which was used to broadly structure this review. An explication is given of how broad and specific review questions were developed, how the literature search was organised and appropriate studies selected, how extracted data were charted, analysed and results reported. This chapter concludes with an interpretation of the meaning of scoping review results, how they inform the development of a SHE tool and underpinning programme theory and design of subsequent studies presented in this thesis.

2.1.1. Why a scoping review?

A scoping study is a type of literature review which aims to map the existing evidence in a field of interest in relation to the volume, nature and features of primary research (Pham et al., 2014). It offers a rigorous and clear method of summarising research evidence and is becoming a well-established approach (Anderson, Allen, Peckham &

Goodwin, 2008; Daudt, van Mosel & Scott, 2013; Davis, Drey & Gould, 2009). Scoping reviews have some commonalities with systematic reviews, but differ in their aims and purposes. For example, a systematic review will aim to answer a well-defined research question with quality assessed studies, whereas a scoping review will present an overview of a diverse body of evidence relevant to a broad topic (Arksey & O'Malley, 2005). Scoping reviews also aim to provide a descriptive summary of the literature; and systematic reviews in contrast aim to critically analyse and synthesise available evidence from studies (Pham et al., 2014). The decision to perform a scoping review was based on its well established methods and usefulness in mapping topics which have not yet been extensively reviewed (Levac, Colquhoun and O'Brien, 2010) which, as established earlier is applicable to SHE and children with DD.

Arksey and O'Malley (2005) have proposed four common aims of scoping reviews:

1. To examine the extent, range and nature of research activity in a particular area
2. To determine the value of undertaking a full systematic review
3. To summarise and disseminate research findings
4. To identify research gaps in the existing literature

Levac et al. (2010) acknowledge that reviews can commonly have multiple objectives and recommend researchers begin by reflecting on these aims whilst simultaneously defining the review questions. Levac et al suggest linking these steps will help provide a clear rationale for undertaking the review and inform subsequent methodological decision making about study selection and data charting. This review was conceived with the aims of gauging the extent of research activity and envisaging where gaps and innovative methods may lie, to inform the inductive development of a SHE tool and programme theory.

2.2. Scoping Review Framework Stages

Arksey and O'Malley's (2005) five stage model for structuring scoping reviews was followed, which incorporates: identifying the research question, identifying relevant studies, study selection, charting the data and finally collating, summarising and reporting results. Each stage will now be described sequentially.

2.3. Framework Stage 1: Identifying the Review Question

This scoping review was guided by the broad question- what is known about SHE as a treatment for behavioural sleep problems in children with DD? Three specific purposes were defined:

1. To identify the content of SHE interventions reported in the literature.
2. To identify effectiveness evidence on SHE.
3. To identify what is known about how SHE works and issues around delivery methods.

Levac et al. (2010) suggest the broad review question is combined with a well-defined scope of inquiry. To clarify the application of the review, and design an appropriate search strategy, significant aspects of the review question should be specified such as the intervention, setting and target population. Therefore, parameters were set to ensure no relevant studies were missed using the models of PICO (Population, Intervention, Comparison, Outcome) useful for identifying studies concerned with treatment effectiveness and SPICE (Setting, Perspective, Intervention, Comparison

and Evaluation; useful for identifying qualitative studies (Higgins & Green, 2011:

Booth, 2004) (Tables 2 and 3).

Table 2: PICO table for questions about SHE content and effectiveness.

Population	Intervention	Comparison	Outcome
Children with DD aged 0-18 years with behavioural sleep problems	Any type of sleep assessment, outcome measure or SHE intervention in any setting including at home, sleep clinics, hospital, and short break setting. Delivered by any method including in a group, one-to-one discussion with practitioner. Excluding pharmacological interventions	Any comparison or no comparison	Any outcome for the child, parents and wider family.

Table 3: SPICE table for question about how SHE works and issues around delivery methods.

Setting	Perspective	Intervention or Phenomenon of Interest	Comparison	Evaluation
Any setting in which SHE occurs including home, sleep clinics, hospital, short break setting.	Children aged 0-18 years with DD with experience of SHE. Views of parents or practitioners about SHE.	Any type of SHE delivered by any method including in a group or 1to1 discussion with practitioner. Excluding pharmacological interventions.	Any comparison or no comparison.	To learn about the experiences of parents and practitioners about how SHE works, through evaluating the review evidence.

2.4. Framework Stage 2: Identifying Relevant Studies

2.4.1. Electronic database searches.

A range of electronic databases (Table 4), sleep organisation, national policy websites,

clinical books from the researcher's clinical background and a Bangor University

catalogue search were systematically searched in January 2013 to answer the review

question. The electronic databases were selected as the most likely to produce studies

relevant to the review question, on the advice of a Health Information Scientist at

Bangor University. Relevant studies available in English or Welsh, from 1966 until January 2013 were included. Any important studies identified in languages other than English or Welsh, were recorded and exclusion noted on the grounds that funding was not available for translation in this project.

Table 4: Electronic databases searched.

Database	Date searched	Number of hits
Medline via EBSCO	11.1.13	868
CINAHL (Cummulative index of Nursing and Allied Health Literature)	11.1.13	182
PsycINFO via ProQuest	11.1.13	444
ASSIA (Applied Social Science Index and Abstract) via ProQuest	17.1.13	44
Cochrane Central Register of Controlled Trials	28.2.13	42
Cochrane Database of Systematic Reviews	28.1.13	0
Ovid Nursing Full Text Plus	24.1.13	85
Web of knowledge	4.2.13	328
Google Scholar	4.2.13	124
Total number of hits		**2117**

For electronic database searching, a primary search strategy was developed through Medline via EBSCO (refer Table 5) with the guidance of a Health Information Scientist from Bangor University. The search was divided into three clear search terms- sleep, children and disability. For each of these topics, thesaurus terms (medical subject headings) were explored initially by identifying terms listed in Medline for relevant studies such as Jan et al. (2008) and Vriend et al. (2011). Additional terms were then developed to create the search strategy and expanded where possible. Medline thesaurus terms were then combined with free-text words to produce a comprehensive yield of results; a strategy recommended by Littleton et al. (2004). Lastly, title and abstract searches were performed on these keywords to allow for more specific searching. To produce a final result, the three subject terms: sleep, children and disability were combined.

The results of this search produced 12208 hits in Medline. A Bangor University Health

Information Scientist advised the researcher to increase precision by limiting the

thesaurus terms of "sleep disorders" and "dyssomnias" - earlier classification of sleep

disorders used to describe difficulties with sleep (American Academy of Sleep

Medicine, 2001)- to relevant subject headings (classification (CL), Diagnosis (DI),

Nursing (NU), prevention and control (PC) and psychology (PX)). This yielded a more

manageable result of 868 papers which was tested against six key papers listed in the

bibliography. This strategy was then translated to the format of the other databases

(Appendix 2). In total, database searching generated a total of 2117 hits and citations

were imported to a Refworks bibliographic software database in order to manage the

high volume of references and making inter-library loan requests.

Table 5. Medline via EBSCO search 14.1.13.

1.	exp sleep/(56738)
2.	dyssomnias /cl. di. nu. pc. px (97)
3.	sleep disorders/cl. di. nu. pc. px (4568)
4.	("sleep problem*" or "sleep disturbance*" or "sleep disorder*" or "sleep hygiene"). ti,ab (18035)
5.	**or/1-4 (combines sleep terms) (71085)**
6.	exp child / (1454298)
7.	young adult/(unable to explode) (250550)
8.	(child* or "young adult"). ti, ab (936299)
9.	**or/6-8 (combines child terms) (1949451)**
10.	developmental disabilities/ (unable to explode) (13238)or exp intellectual disability/ (77082)or exp child development disorders pervasive/ (17516)(103035)
11.	autistic disorder (unable to explode)(14545)/ or cerebral palsy (unable to explode) (14474)/ or down syndrome(unable to explode) (19658)(48138)
12.	("learning disab*"or retard* or "intellectual*impair*" or "special needs" or "multiple disab*"). ti,ab (95460)
13.	(autis* or "cerebral palsy" or " down*syndrome") .ti,ab (32204)
14.	**or/10-13 (combines disability terms) (192867)**
15.	**5 and 9 and 14 (868)**

2.4.2. Hand-searches.

To identify further relevant studies, the reference lists of included studies were

scanned for novel studies not identified by electronic searching. The bibliographies of

review papers identified by the search were also checked, (even though review articles

were not included as primary studies in the review). This did highlight some additional studies although a saturation point was achieved where no new studies were being identified.

Journals containing the largest number of papers meeting the inclusion criteria were hand searched: Journal of Intellectual Disability Research (2008-2012) and Developmental Medicine and Child Neurology (2006-2012). Time frames were set to reflect those within which most publications were identified.

An ancestry search (Schlosser, Wendt, Bhavnanis & Nail-Chiwetalu, 2006) was completed on recent studies of high relevance, by studying reference lists to find previously published papers. A forward citation search (Schlosser, Wendt, Bhavnanis & Nail-Chiwetalu, 2006) was also performed using Medline/Web of Knowledge and Google Scholar, whereby subsequent published research was identified from these high relevance studies. From the initial SHE review search on 11.10.12 (Section 1.5) key authors were identified: Beth Malow, Byrony Beresford, Amanda Richdale, Luci Wiggs and Gregory Stores. These authors were contacted to identify ongoing or unpublished work and individual searches were conducted on their names. In addition, relevant clinical books, from the researcher's clinical background and identified through searching Bangor University catalogue were hand searched for bespoke SHE tools.

2.4.3. Existing networks, relevant organisations and conferences.

To generate further information about primary research, websites were searched of relevant sleep organisations such as Cerebra, American Academy of Sleep Medicine (AASM), Southampton Sleep Service, International Paediatric Sleep Association

(IPSA), Scope Sleep Solutions and Sleep Scotland. WIPMD (*Who is Publishing in My Domain?)* which registers current research linked to the researcher's published article on sleep (Sutton, 2011) were also scanned for relevant studies. Where searches did not identify relevant information, organisations were contacted directly to request contributions to the review. Contributions were provided using this method from the Handsel Project, Cerebra, Scope, Contact a Family and the Down's Syndrome Association. The researcher was unable to gather contributions from other sleep organisations that were unable to share their SHE resources for commercial reasons. The various mechanisms for searching in this scoping review yielded a total of 2203 references. In total, 2117 were identified from the electronic database searches and 86 via hand-searching and contacting existing networks (Figure 2).

2.5. Framework Stage 3: Study Selection

Titles and abstracts of the 2203 records identified through the search process were firstly screened by the researcher using the inclusion and exclusion criteria (Table 6). This was developed with the guidance of the supervisory team, to ensure rigor and guide the process of filtering out records which were not relevant to answering the review question. This produced a total of 374 relevant records (Figure 2). A second title and abstract screen was then carried out by the researcher and a fellow research student who independently reviewed these records (again using the inclusion and exclusion criteria). The supervisory team was consulted if there was any doubt as to the eligibility of a paper. This stage identified 273 relevant records for which full texts were obtained through Bangor University library and inter library loans. All 273 full papers were reviewed against the inclusion and exclusion criteria again, by the researcher and members of the supervisory team. Duplicate records were excluded and

non-relevant papers kept with rationale for exclusion. The study selection process

finally identified a total of 52 studies which met the inclusion criteria (Appendix 1).

Taking a systematic approach in the study selection phase and involving a team of

reviewers is recommended by Levac et al. (2010) to ensure rigor in scoping reviews.

Figure 2 Flow diagram of the literature identified by the search process
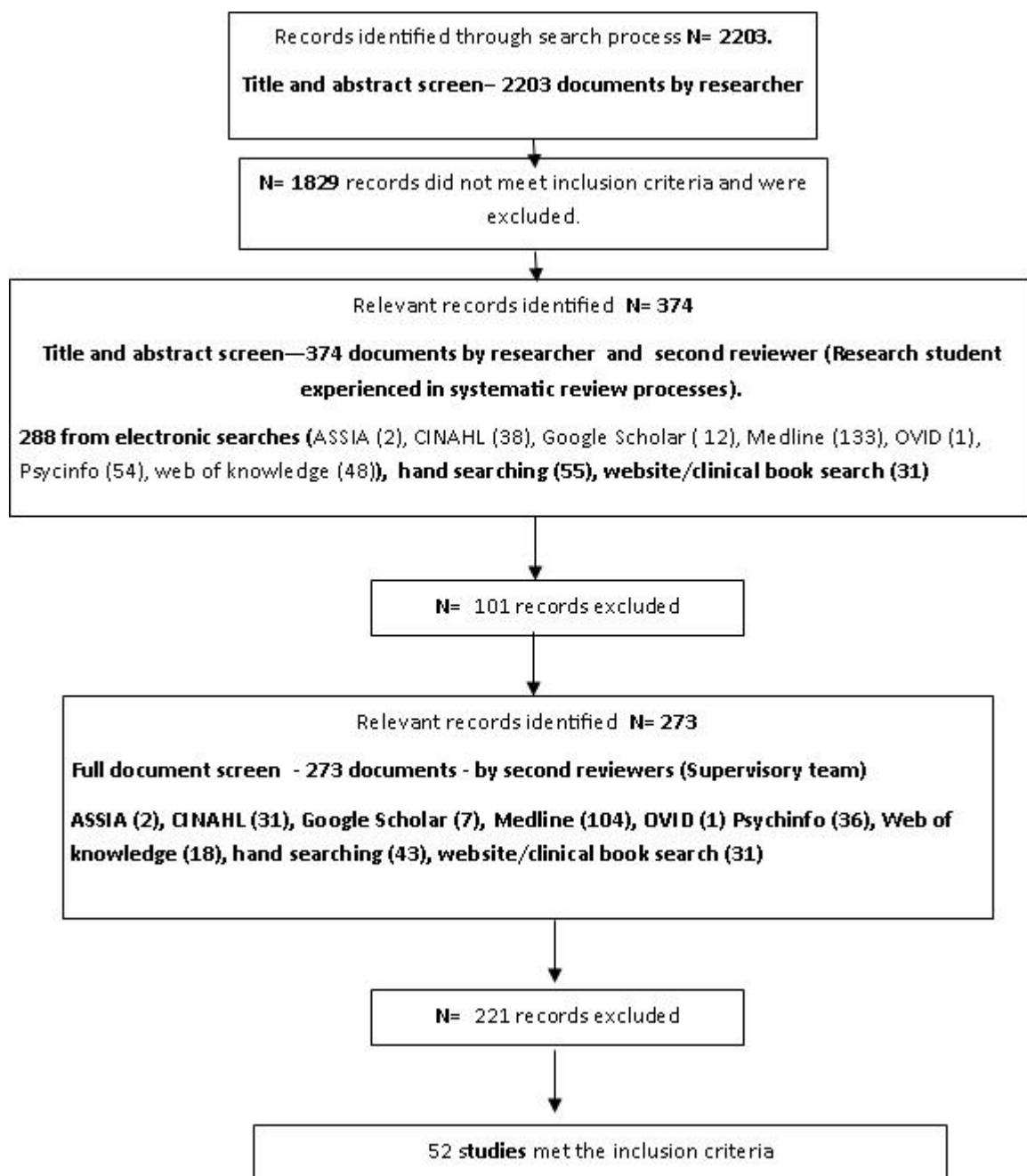
Table 6 Scoping review inclusion and exclusion criteria.

Review aims		Included	Excluded
To determine the content of SHE.	**Types of study**	All types of primary quantitative studies: experimental (including quasi-experimental, surveys, case studies, surveys, descriptive studies and bespoke SHE tools).sleep assessment studies.	Reviews, posters and epidemiological studies.
	Population	Studies focusing on children aged 0-18 years with DD with behavioural sleep problems, their parents and practitioners and SHE.	Studies which focus solely on adults, typically developing children or children with a sole diagnosis of physical disability. Studies focusing solely on pharmacological interventions.
	Types of intervention	Studies that detail any type of SHE for children with DD (and report a minimum of 2 SHE advice areas such as bedtime routines or sleep timing). Studies that focus on the assessment of behavioural sleep problems and outcome measurement.	
To identify evaluative studies	**Types of study**	Primary quantitative studies: experimental (including quasi-experimental, multiple baseline and surveys.)	
	Population	Studies focusing on children aged 0-18 years with DD with behavioural sleep problems, their parents and practitioners and SHE.	Studies which focus solely on adults, typically developing children or children with a sole diagnosis of physical disability. Studies focusing solely on pharmacological interventions
	Types of intervention	Studies that detail any evidence of effectiveness of SHE in treating behavioural sleep problems in children with DD (and report a minimum of 2 SHE advice areas such as bedtime routines or sleep timing).	
To establish how SHE works and explore issues around delivery method.	**Types of study**	All types of primary quantitative studies: experimental (including quasi-experimental), descriptive studies, surveys and case studies. Primary qualitative studies that have elicited views of parents and practitioners on SHE, organisational and policy factors.	
	Population	Studies focusing on children aged 0-18 years with DD with behavioural sleep problems, their parents and practitioners and SHE (and report a minimum of 2 SHE advice areas such as bedtime routines or sleep timing).	Studies which focus solely on adults, typically developing children or children with a sole diagnosis of physical disability. Studies focusing solely on pharmacological interventions
	Phenomenon of interest	Studies that explain how SHE is supposed to work. Experiences of parents and practitioners of SHE. Organisational and policy factors.	

2.6. Framework Stage 4: Charting the Data

A standardised approach was taken to chart extracted data and general and specific information was recorded as recommended by Arksey and O'Malley (2005). This mirrored the 'descriptive-analytical' method, rooted in the narrative tradition which seeks to both apply an analytical framework to charting the data and more broadly includes information about 'process' of interventions so that outcomes can be put in context (Pawson, 2002). The following information was recorded in data charting forms:

- Study details (including author, year of publication and study aims).

- Population and setting.

- Methods.

- Intervention.

- Outcomes and outcome measures.

- Results.

It was not possible to obtain all required data because relevant information was not provided in every study. This was particularly true of bespoke SHE tools because although they counted as evidence within the context of this review, they were not primary research studies which used clearly reported research methods. Extracted data were also second checked by a fellow research student at Bangor University with appropriate reviewing skills and experience, to minimise bias. The data charting form is displayed in Appendix 2 and each study is presented in alphabetical order.

2.7. Framework Stage 5: Collating, Summarising and Reporting the Results

Levac et al. (2010) recommend this stage has three systematic steps to ensure rigor of approach: analysing the evidence, reporting findings and interpreting the meaning of results. Analysis should involve a descriptive numerical account of included studies including elements such as the number of relevant studies and types of study design (Arksey & O'Malley, 2005). Data should then be analysed using qualitative data analytical techniques and represented using tabular or graphical formats (Arksey & O'Malley, 2005; Levac et al., 2012; Pham et al., 2014). In this review data will be analysed and reported using content analysis and descriptive numerical and narrative accounts, in relation to each of three specific review purposes. Finally, the meaning of these combined results and implications for SHE tool and programme theory development will be considered in the discussion of this review.

2.7.1. Review objective one: identifying the content of SHE.

Eighty one percent (n=42) of identified studies reported how sleep problems were assessed prior to SHE advice being given and 87% (n=45) of studies described the components of SHE. These elements which collectively describe the content of SHE interventions will be addressed separately in the following sections.

2.7.2. The sleep assessment process.

Eighty on percent of studies (n=42) reported on sleep assessment and a narrative summary is provided of the main elements underpinning the process: elimination of physical and psychological co-morbidities, sleep history taking, sleep outcome measurement, sleep scoring tools and quality of life outcome measurement.

2.7.3. Elimination of physical and psychological co-morbidities.

Thirty seven percent (n=19) of studies reported how the process should commence with screening for physical and psychological co-morbidities which could potentially explain the causes of sleep problems. Nineteen percent (n=10) of studies which were all bespoke SHE tools recommended that this preliminary stage should be carried out (Autism Society, 2012; Capability Scotland, 2012; Catherall & Williams Jones, 2011; Contact a Family, 2007; Down's Syndrome Association, 2008; Durand, 2011; Durand, 2013a; Durand 2013b; Early Support, 2010; Scope, 2012). Seventeen percent (n=9) of studies which were primary research studies reported engaging in the screening process. This involved a developmental paediatrician carrying out a physical and psychological examination of the child (Adkins, Molly et al., 2012; Beresford et al., 2012c, Christoldulu & Durand, 2004; Malow et al., 2013, Stores & Stores, 2004), screening for co-morbidities using a sleep screening questionnaire (Montgomery et al., 2004) or running neurological examinations such as electroencephalograms (Bartlet & Beaumont, 1998; Cortesi et al., 2012). Reed et al. (2009) who involved a neurology sleep specialist to assist in this process also reported eliminating other primary sleep disorders such as narcolepsy (a hypersomnolence sleep disorder where the child falls to sleep dramatically in the day despite sleeping well at night) at this preliminary stage, although the methods used to identify them were not reported. In all 19 studies which discussed this element of 'best practice', a clear motivation was the need to uncover co-occurring conditions such as epilepsy and depression and ensure they were treated appropriately before behavioural interventions such as SHE were implemented. Indeed, if such conditions were overlooked, behavioural interventions would potentially be less effective (Malow et al., 2013).

2.7.4. Sleep histories.

Twenty nine percent (n=15) of studies reported the use of sleep histories, in the form of semi-structured interviews that asked parents a series of open and closed questions about the history of their child's sleep problem. Three studies (Hewitt, 1985; Johnson et al., 1995; Petta et al., 2012) described sleep histories to assess sleep problems in children with DD, but did not specify specific examples of tools used. Two studies (Weiskop et al., 2001; Weiskop et al., 2005) reported the use of a modified sleep assessment interview taken from a behavioural assessment of children's problems. However, the exact form of sleep history tool used in these two studies was not reported and despite contacting the authors, the researcher was unable to obtain any further detail. Ten studies (Bartlet & Beaumont, 1985; Beresford et al., 2012c; Cerebra, 2011; Christodulu & Durand, 2004; Durand, 2013b; Handsel Project, 2012; Johnson et al., 2012; Montgomery et al., 2004; Stores & Stores, 2004; Wiggs & Stores, 2001) described sleep history tools in detail and a total of seven different tools were identified: Albany Sleep Problems Scale (ASPS), Cerebra sleep service questionnaire (Cerebra SQ), Handsel Project sleep interview schedule (HP), Modified Simmonds and Parraga Sleep questionnaire (MSPSQ) (full version), Modified Simmonds and Parraga Sleep questionnaire (MSPSQ)(shortened version), Sleep history review of 24 hour sleep pattern (SHR) and Southampton Sleep Management Scale (SSMS) (Refer Appendix 4 for characteristics).

2.7.5. Analysis of sleep histories and their functions.

The central purpose of taking a sleep history is to improve understanding of the sleep problem, continue to uncover multiple causal factors and ultimately inform treatment design (Durand 2013b; Stores & Wiggs, 2001). More detailed sleep history functions

were generated by following the principles of content analysis (Bowling, 2014), which guides the researcher in coding sections of the data inductively developed by the researcher. Interesting features across the range of sleep history questions featuring in each sleep history tool reported were coded and collated into categories or functions. For example the questions 'are you affected by your child's sleep problem?' and 'what strategies have already been tried?' both pertained to the function of 'exploring family attitudes towards sleep problems' and judging whether they were ready to embark on a SHE intervention. The coding rule was applied whereby all codes were mutually exclusive and each single unit of data were coded in one category only. Finally, a frequency count of the number of codes in each sleep history function category allowed the data to be summarised. By going back to the data available for each function and organising them into an understandable account, a narrative summary was developed for each of the six specific functions of sleep history taking, further evidencing its integral role in the sleep assessment process. The six functions were defined as : 'Establishing current 24 hour sleep/wake routine', 'Describing the sleep problem', 'Identifying primary sleep disorders', 'Identifying physical and psychological co-morbidities', 'Establishing historical or day to day triggers and 'Exploring family attitudes toward the sleep problem'. Over half of sleep history tools examined (MSPSQ (full version), Cerebra, HP and SSI) included questions designed to elicit information which contributed to all six areas of specific function. These functions will now be presented.

Function 1. Establishing current 24 hour sleep/wake routine.

All seven sleep history tools identified included questions aimed at establishing the activities and behaviour of the child over a period of twenty four hours. Questions

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sought to establish for example how many different settings the child slept in in a typical week, if sleep patterns were different at weekends and what the child's bedtime routine looked like. Usually these were opening questions in sleep history questionnaires and helped to paint a picture of the child's typical day and night. Example questions include:

- *"Who gets the child ready for bed and how? Is it always the same person and done in the same way?" (*SHR, Question 4)

- *"Does your child have a bedtime routine that is followed every night? Yes/No. If yes, please describe"* (MSPSQ (Full and shortened version) Part two, Question 4).

Function 2. Describing the sleep problem.

All seven sleep history tools identified asked parents to describe the presenting sleep problem. Questions intended to establish the nature of the problem i.e. how the child behaved and how the parent responded. Confirming the nuances of the sleep problem and surrounding context, helps to build understanding so that interventions can be tailored to meet the child and family's needs (Durand, 2013b). Example questions include:

- *"Exactly what happens if he will not go to bed or does not sleep readily? Who deals with the problem and how consistently?"* (SHR, Question 14).
- *"What do you do if your child will not go to bed or settle to sleep?"* (MSPSQ, Full and shortened version).

- *"Does this person resist going to bed? 0= never; 1= less than once per week; 2= one to two times per week; 3= three to six times per week; 4= nightly)* (ASPS, Question 7).

Function 3. Identifying underlying sleep disorders.

All sleep history tools (with the exception of the MSPSQ (Shortened version)) included questions which probed for information to assist in the detection of primary sleep disorders such as nightmares or sleep related breathing disorders. Questions were typically closed or asked for details which helped to differentiate the characteristics of specific sleep disorders. For example the following question aimed to uncover evidence of narcolepsy.

"Does this person experience sleep attacks (falling asleep almost immediately and without warning) during the day? 0= never; 1= less than once per week; 2= one to two times per week; 3= three to six times per week; 4= nightly. (ASPS, Question 23).

In addition, the following example question aimed to deduce if there was any family history of restless legs syndrome which could also potentially affect the child being assessed.

- *Does anyone on either side of the family experience restless legs when falling asleep (crawling or aching feelings in legs, unable to keep legs still, having to get out of bed to ease aching legs)?* (MSPSQ, Full version, Question 12).

Function 4. Identifying physical and psychological co-morbidities.

Five of the sleep history tools examined (except MSPSQ (Shortened version) and SHR) included questions which identified associated physical or psychological co-morbidities known to affect sleep such as sensory integration issues or epilepsy. In some intervention studies, this served as an additional check to the initial screening process to eliminate these possible causes (Christodulu & Durand, 2004; Stores & Stores, 2004). The intention was to ensure co-morbidities were identified and treated appropriately prior to embarking on behavioural sleep interventions. Example questions include:

- *"Have you considered food intolerance or allergy as a cause? Yes/No"* SSMS, Section7)
- *"Are there any sensory distractions which may affect the child's ability to sleep e.g. noise from heating system or flickering street lights?"* (HP, Question 9)
- *"Has your child any history of epilepsy/convulsions/seizures?* (MSPSQ (Full version), Part one, Question 4).

Function 5. Establishing historical and day to day triggers.

Five of the sleep history tools examined (except MSPSQ (Shortened version) and SHR) included questions which identified historical causes such as bereavement or abusive situations and daily triggers such as parents' work shift patterns or parental stress. Building up a picture of contextual factors helps to improve sleep problem understanding and may signal the need for referrals to additional support agencies

(Beresford et al., 2012a). In some cases it may be prudent to postpone the implementation of behavioural sleep interventions until the relevant support services have been involved, as overlooking these problems may impact on intervention effectiveness. Example questions include:

- *"Are there any known stressors (such as traumatic events, family difficulties etc) in the family that may affect sleep?"* (Cerebra, Section 3).
- *"When did this person's primary difficulties with sleep begin?"" What was happening in this person's life at that time or a few months before?"*(ASPS, Questions 44 and 45).

Function 6: Exploring family attitudes towards sleep problems.

Four of the sleep history tools (Cerebra SQ, HP, MSPSQ (full version) and SSMS) enquired about sleep help previously received and strategies already tried. This helps to establish the willingness of parents to try subsequent behavioural sleep interventions (Johnson et al., 2012). Qualitative information about the parents' perceptions of their child's sleep problem and impact on family life is also helpful in assessing their readiness to address sleep problems and identifying appropriate methods of support. Example questions include:

- *"What strategies have already been tried? Please detail how long for and level of consistency. In hindsight, was this a good time for the family to undertake sleep work?"* (HP, Question 5).

- *"Is there anything you as a family are not willing to try and is this the right time to start a sleep programme?"* (Cerebra, Section 5)

- *"Are you affected by your child's sleep pattern? Yes/No. If yes, how does this affect you e.g. feelings of irritation, arguments with partner, feeling powerless" (*MSPSQ (Full version), Part 5, Question 9).

Three sleep history tools (SHR, Cerebra and HP) were described by bespoke SHE tools only and had not been tested in any included intervention or assessment studies. The SSMS was utilised in one intervention study (Bartlet & Beaumont., 1998) and the ASPS was described in a bespoke SHE tool (Durand, 2013b) and tested in one intervention study (Christodulu & Durand, 2004). The MSPSQ (Shortened and Full version) was utilised in one assessment study (Johnson et al., 2012) and one intervention study (Stores & Stores, 2004). Therefore there was limited use of sleep history tools reported across assessment and intervention studies.

2.7.6. Sleep outcome measures.

Sixty three percent (n=33) of studies documented the use of sleep outcome measures which are used to establish a baseline recording of the sleep problem and quantify sleep improvements following intervention (Cortesi et al., 2012; Malow et al., 2011; Adkins, Molloy et al., 2012). Measures can also facilitate the identification of underlying sleep disorders such as circadian rhythm sleep disorders or sleep terrors (intense episodes of panic and crying during sleep) (Durand, 2013b; Wiggs & Stores, 2001), prompting further clinical investigation to confirm diagnosis and appropriate treatment. Lastly, as Reed et al. (2009) report outcome measures can help identify specific sleep challenges for individual children to ensure behavioural treatment can be

tailored to their needs. Therefore, some overlap exists between the purposes of sleep histories and sleep outcome measures.

A range of sleep outcome measures were reported in the literature with varying depths of description. In their intervention studies, Allison et al. (1993), Bramble (1997) and Piazza et al. (1997) briefly described sleep outcome measures purposively designed for each study, such as visual analogue scale and momentary time sampling procedures. However, no evidence of reliability or validity was reported in any of these studies. Four studies (Beresford et al., 2012c, 2012d & 2012e; O'Connell & Vanaan 2008; Weiskop et al., 2001; Weiskop et al., 2005) reported the utilisation of a Goal Achievement Scale (GAS) based on parent stated goals. This method was adopted to understand how sleep interventions achieve specific outcomes that were important to parents and establish satisfactory positive change in sleep patterns that were acceptable to parents rather than perfect sleep. However, it was important that goals set were realistic, accurate and measurable to ensure this method of outcome measurement was successful (Beresford et al., 2012c). In contrast, measures with reported evidence of reliability and validity were described by 31% (n=16) of studies and a total of nine outcome measurement tools were specified: Child Sleep Habit Questionnaire (CSHQ) (33 item), CSHQ (39 item), CSHQ (52 item), Family Inventory of Sleep Habits (FISH) (12 item), FISH (22 item), Cerebral Palsy Questionnaire (CPQ), MSPSQ (Part 4), MSPSQ (shortened version) and MSPSQ (full version).The characteristics of these nine outcome measures are detailed in Appendix 4 and it is important to clarify that MSPSQ (shortened version) and MSPSQ (full version) have both sleep history and sleep outcome properties.

The most commonly used validated outcome measure was the CSHQ (17% n=9)

although the exact version used in studies was not always reported. Studies typically

chose this tool based on its reported validity and increasing use in studies with

children with DD (Reed et al., 2009; Malow et al., 2013). However, the CSHQ was

primarily developed for typically developing children (Adkins, Molloy., et al, 2012)

and has not been adapted for DD, therefore has questionable applicability. The FISH

(12 and 22 item version) tool was reported in just 8% (n=4) of studies and has a

narrow focus which measures the frequency of the child's sleep habits in children with

ASD (Malow et al., 2009). The MSPSQ was reported in fewer included studies (6%,

n=3) but had the highest applicability being modified specifically for the needs of

children with a broad range of DD. Finally, the CPQ was developed, tested and

reported in just one study (Khan and Underhill, 2006) and had limited applicability to

children with a diagnosis of cerebral palsy.

Sleep diaries.

Thirty seven percent (n=19) of studies recommended or used a sleep diary (a

subjective and non-validated outcome measure) as part of the sleep assessment

process; making this the most commonly used outcome measure. The sleep diary is

described as a weekly form that requires parents to record daily specific sleep events

(Cortesi et al., 2012). Typically for a period of two weeks, parents chart subjective

information about settling, night time awakenings, daytime naps and sleepiness and

describe how they manage sleep problems (Montgomery et al., 2004). Three studies

(Christodulu & Durand, 2004; Weiskop et al., 2001; Weiskop et al., 2005) used the

sleep diary as a stand-alone outcome measure, reporting outcomes taken directly from

the parent completed charts. (These were also the only studies to acknowledge the

need to train and support parents in the task of sleep diary completion). However, the sleep diary was more commonly used in intervention studies as an adjunct to validated outcome measures such as the CSHQ or actigraphy (Adkins, Molloy et al., 2012; Cortesi et al., 2012; Reed et al., 2009) to corroborate findings or to feed into sleep scoring tools (Bartlet & Beaumont, 1998; Montgomery et al., 2004).

Sleep scoring tools.

Sleep scoring tools were reported in the literature: Composite Sleep Disturbance Score (CSDS) (Montgomery et al., 2004), Composite Sleep Problems Score (CSPS) (Stores & Stores, 2004), Sleep Disturbance Index (SDI) (Bartlet & Beaumont, 1998) and the Sleep Related Breathing Problems Score (SRBPS) (Stores & Stores, 2004). These all worked in a similar way by extracting data from sleep outcome measures and calculating the frequency and severity of the sleep problem. For example, Montgomery et al. (2004) calculated a CSDS score for the recorded frequency of settling duration problems. Settling problems that were reported three or four times a week were given a score of one, whereas a score of two was given to problems occurring five or more times. Scoring tools then produced an overall score which was used as an outcome measurement, before and after a sleep intervention. Just three intervention studies used sleep scoring tools and there was a lack of consistency across these studies in terms of types of tools used.

Actigraphy.

Fifteen percent (n=8) of studies reported using actigraphy as an objective sleep outcome measure. Actigraphy involves the use of a small mechanical device worn by the child on the wrist, shoulder or ankle, which analyses physical activity using linked

computer software and objectively measures sleep-wake patterns (Goodlin-Jones et al., 2008; Cortesi et al., 2012).

Intervention studies that used this method of outcome measurement reported actigraphy to be a well-established and validated tool (Adkins, Molloy et al., 2012; Goodlin-Jones et al., 2008). The need to invest time in training parents in its use was reported by Malow et al. (2013) and Adkins, Molloy et al. (2012). Problems with equipment failure and compliance issues with children with DD were reported in some studies (Stores and Stores, 2004; Malow et al., 2013). Two studies (Adkins, Goldman et al., 2012; Sitnick et al., 2008) specifically aimed to test the psychometric properties of actigraphy. Adkins, Goldman et al. (2012) compared possible placement sites for the actigraph device when used with children with ASD, and found that the shoulder offered a feasible alternative to the more commonly used wrist location. Sitnick et al. (2008) reported some concerns regarding the ability of actigraphy to detect night awakenings in children with DD when compared with the alternative mechanical outcome measure of videosomnography (time-lapse video recording of sleep).

Sleep outcomes.

Sleep outcomes were reported in 42% (n=22) of studies. These are summarised in the data charting forms (Appendix 3). The majority of these studies 29% (n=15) used validated outcome measures and reported sixty different sleep outcomes (Table 7). However, studies did not consistently report all available outcomes when using the same outcome measure. Only Cortesi et al. (2012), Reed et al. (2009) and Malow et al. (2013) reported all nine available CSHQ sleep outcomes. In studies which used actigraphy, Akins, Molloy et al. (2012) reported total sleep time, sleep latency, sleep

efficiency and wake after sleep onset outcomes whereas Cortesi et al. (2012) reported

these and also nap time and bedtime outcomes.

Table 7. Summary of sleep outcomes reported from validated outcome measures.

Validated outcome measure	Sleep outcomes
Actigraphy	Total sleep time, sleep latency, sleep efficiency, wake after sleep onset, naptime, bedtime, mean duration of wake episodes, number of waking episodes, duration of longest waking episode, mean activity score, total minutes scored as sleep. **(n-=11)**
MSPSQ (Part 4)	Total MSPSQ score. **(n=1)**
CSHQ (33, 39 and 52 item)	Total CSHQ score, bedtime resistance, sleep anxiety, night awakenings, sleep onset delay, sleep duration, daytime sleepiness, parasomnias, sleep disorder breathing. **(n= 9)**.
CPQ	Sleep latency, night awakenings with pain, position changes through night. **(n=3)**
FISH (12 item)	Total FISH score, exercise in day, wakes up about same time each day, relaxing activities before bed, caffeine after 5pm, child engages in exciting or stimulating activities in hour before bed, room is darkly lit, room is quiet, child goes to bed same time every night, regular bedtime routine (15-30mins), parent stays with child until asleep, parent checks on child before he sleeps, child watches TV to fall asleep.**(n=13)**
FISH (22 item)	Total FISH score, exercise in day, bedroom used as play area in day, wakes up about same time each day, relaxing activities before bed, caffeine after 5pm, stimulating activities before bed, room is darkly lit, room is quiet, follows regular bedtime, parent stays with child until asleep, parent checks on child before he sleeps, child watches TV to fall asleep, child listens to music to fall asleep, parent keeps interactions brief if child awakens, parent returns child to bed if awakens, napping in day, bedroom used for timeout in day, sleeps better in certain sleepwear, sleeps better in certain sheets, sleeps better in certain room temperature, goes to bed at the same time each night, uses favourite comfort object. **(n=23)**

Quality of life outcome measures.

Twelve percent (n=6) of included studies reported the use of quality of life (QOL)

measures to assess how improved sleep impacted on families' day to day lives.

Numerous QOL measures were reported, however the only measure to be used in

more than one study was the Parenting Sense of Competence Scale (PSOC) (Beresford

et al., 2012c, 2012d & 2012e; Malow et al., 2013). This measures how competent the

parent feels in their caring role and incorporates two QOL outcomes: 'satisfaction'-

which indicates how satisfied parents are with their parenting and 'efficacy' – which

captures how parents consider they are managing the parenting role in terms of

aptitude (Beresford et al., 2012c).

Review findings have provided comprehensive evidence to address the review purpose

of identifying SHE content and how sleep problems are assessed. The need to screen

for physical and mental co-morbidities, administer sleep histories and sleep outcome measures has been established. Sleep history functions and numerous sleep history tools, outcome measures, sleep outcomes and QOL measures have also been identified. The second element which describes SHE content: SHE advice components will now be addressed.

2.7.7. The components of SHE.

Data illustrating the components of SHE were charted from the majority (87%, n=45) of studies. Seven studies focused on assessment and so did not report SHE content. A content analysis method (Bowling, 2014) was used to summarise the evidence, which involved coding sections of the data related to categories inductively developed by the researcher as the data were collected. Categories and potential categories were noted in the margins of the studies where SHE advice components were described. All categories (SHE component areas) and codes (SHE advice points) were stored on a table (refer Appendix 5 for excerpt) to allow for easy retrieval and analysis. For example, advice points such as 'consistent bedtimes and wake times' and 'age appropriate bedtimes' both related to the timing of sleep therefore a 'sleep timing' SHE component area was identified. The coding rule was applied whereby all codes were mutually exclusive and each single unit of data were coded in one category only. Finally, a frequency count of the number of SHE advice points in each component area allowed the data to be summarised. Six component areas were identified: sleep timing, bedtime routines, behavioural management, environment, physiological and communication adaptations and fifty two individual SHE advice points (Figures 3-6).

Figure 3 Graph showing sleep timing and bedtime routines advice areas.
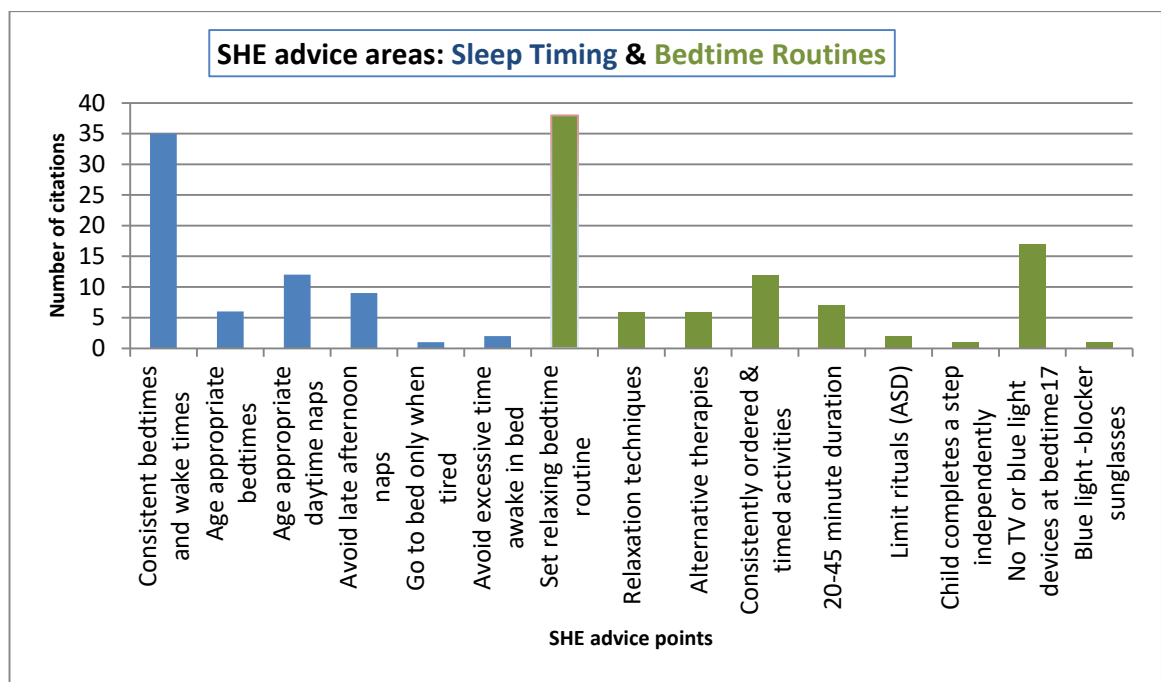
Figure 4 Graph showing behaviour management and communication adaptations advice areas.
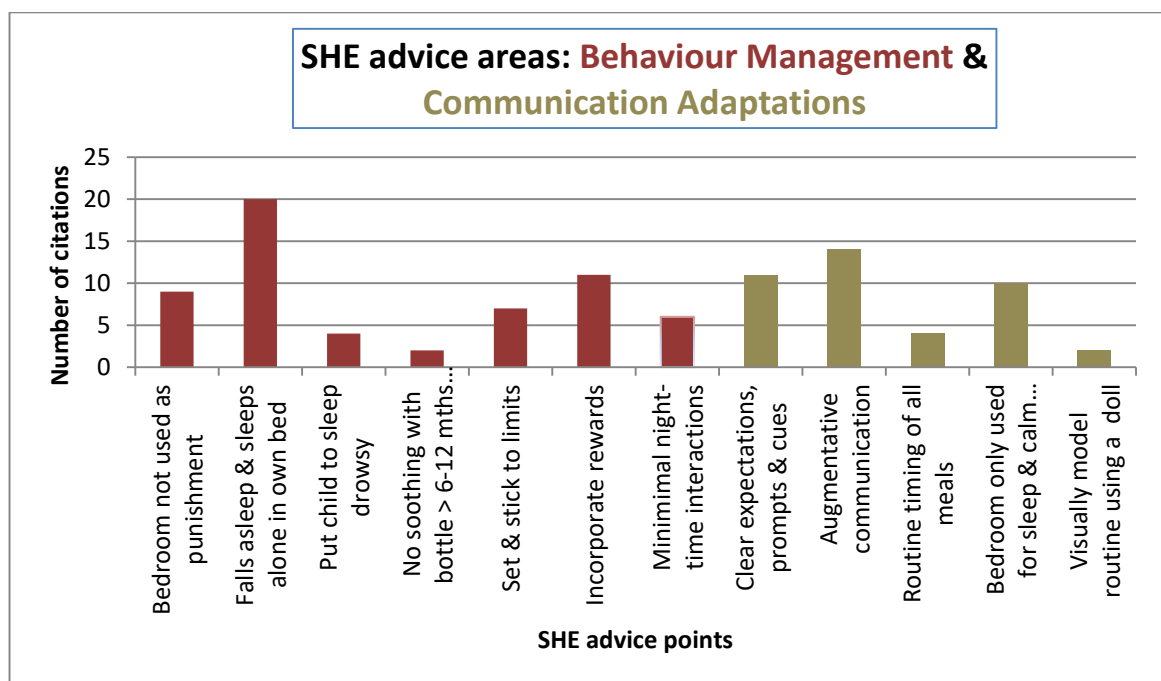
Figure 5 Graph showing physiological advice area.

SHE advice area:Physiological
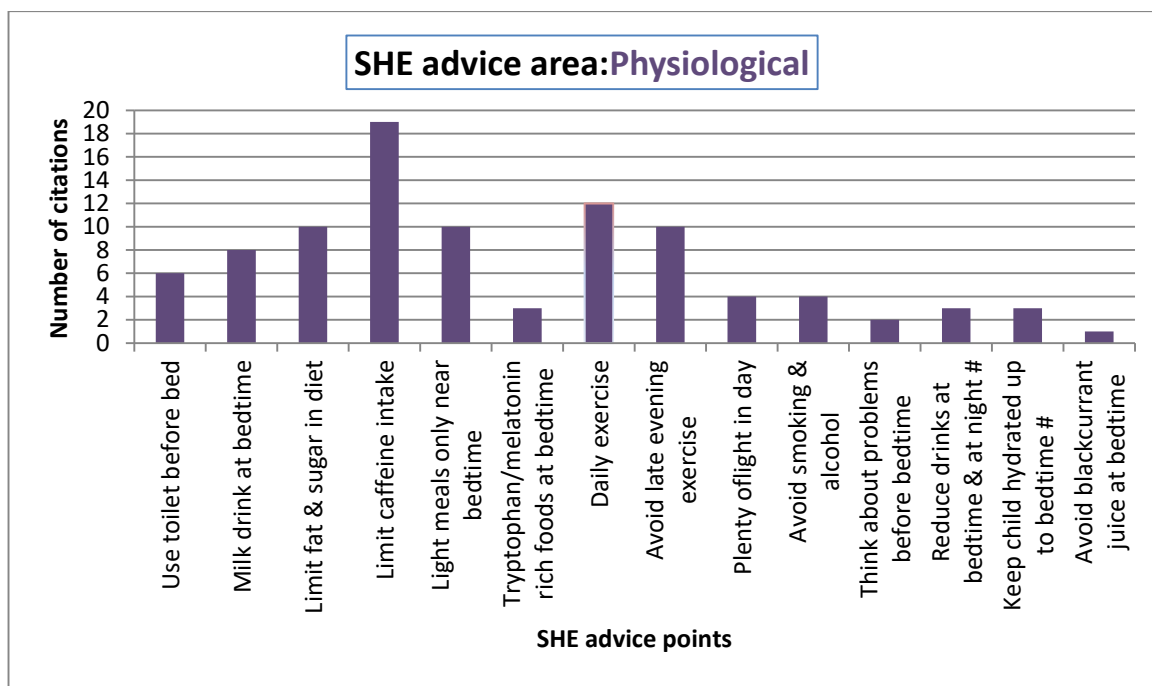
Figure 6 Graph showing environment advice area.

SHE advice area: Environment
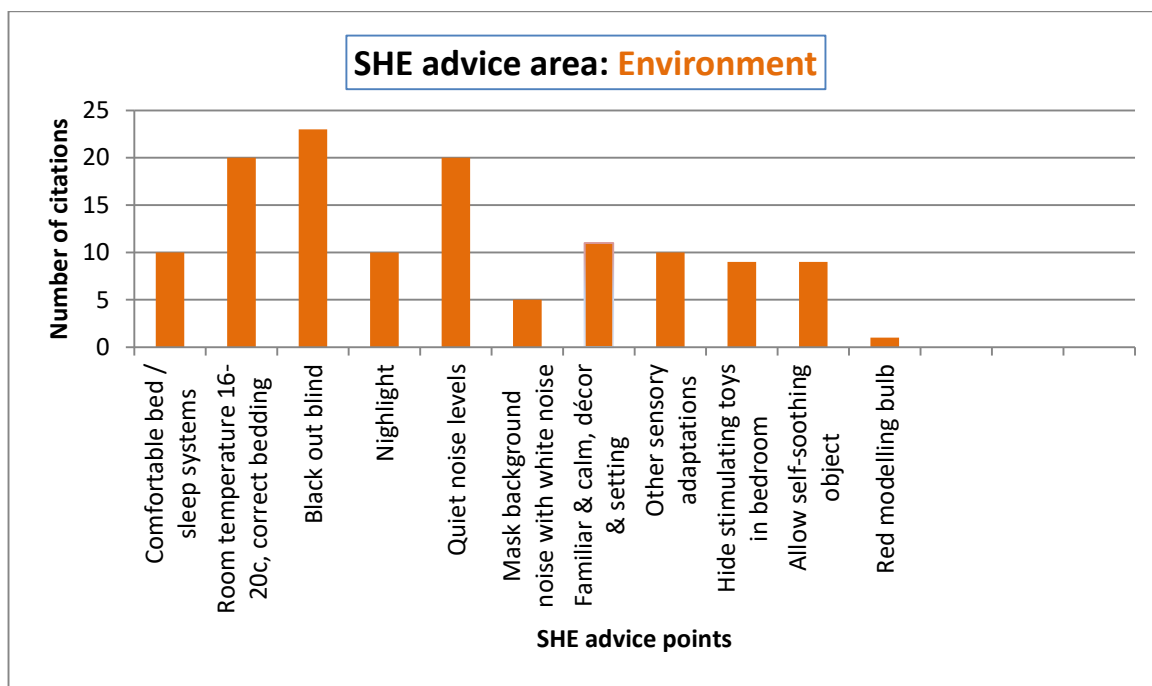
Sleep timing.

This describes six advice points which promote optimal sleep timing and focuses on bed times, wake times and daytime naps (Figure 3). Sixty seven percent (n=35) of studies reported the advice point of 'encouraging consistent bedtimes and wake times, including weekends and holidays'. Some studies also gave the rationale for this and explained how this helped to regulate the child's sleep pattern (Cerebra, 2011; Sense, 2012; Scope, 2012). Twenty three percent (n=12) of studies stated advice around 'age appropriate daytime napping' which included recommendations for phasing out daytime sleeps soon after the child's 3rd birthday (Handsel Project, 2012).

Bedtime routines.

This category describes nine advice points on the importance of setting a bedtime routine for the child (Figure 3). Seventy three percent (n=38) of studies broadly stated that a regular and structured routine should be developed. This was the most frequently cited SHE advice point. Studies often gave sample bedtime routines (Early Support 2010; National Autistic Society, 2008; Scope, 2012) and explained how following a consistent routine was helpful for children with DD as this helped them predict events and reduce anxiety (Cerebra, 2011; National Autistic Society, 2008; Sense, 2012). Thirty three percent (n=17) of studies cited ensuring the child avoids television, stimulating electronic games or blue-light emitting devices at bedtime, in the bedtime routines category. A solution for children with DD who may be resistant to this was put forward in the advice point 'consider blue light blocker sunglasses' however this was only reported in one study (Handsel Project, 2012). Some variance was evident amongst studies about the recommended duration of bedtime routines and this ranged from 20 minutes (Research Autism, 2007) to a maximum of 45 minutes

(Handsel Project, 2012). This range was recorded as one code "20-45 minute duration".

Behaviour management.

This describes seven advice points which deal with a child's resistive or challenging behaviour at sleep times such as refusing to go to bed, screaming or self-injury (Figure 4). Thirty eight percent (n=20) of studies advised that the 'child falls asleep and sleeps alone in own bed'; this was the most commonly reported advice point in the behaviour management category. Research Autism (2007) explained how parents' practice of sleeping with their children (co-sleeping) forms a negative sleep association and can reinforce disrupted night awakenings. Furthermore, Research Autism highlighted how the acceptability of this practice reduces as the child grows older. Twenty one percent (n=11) of studies emphasised the importance of being positive with a child who is learning how to sleep better and suggested 'incorporating rewards' into any SHE strategy. Seventeen percent (n=9) of studies also promoted the positive practice of ensuring 'the child's bedroom was not used as a punishment setting' which continues the theme of making sure the child develops healthy sleep associations and associates their bedroom with calmness and sleep.

Environment.

This relates to advice around creating an environment conducive to sleep (Figure 6). Many studies 44% (n=23) reported the recommendation of 'darkened room/black out blind' and some studies explained how darkness stimulates the natural production of melatonin which promotes sleep (Early Support, 2010; Scope, 2012). Thirty eight percent (n=20) of studies reported the recommendation of 'correct room temperature

16-20c' and some of these studies also advised on appropriate bedding and sleep wear to ensure the child was not too warm or cold for sleep (Scope, 2012; Leeds Community Healthcare, 2010). The advice point of 'quiet noise levels' was also reported by over a quarter of studies 38% (n=20), whereby parents are advised to reduce background noise rather than eliminate it completely. The advice point 'sensory adaptations' reported by 19% (n=10) is particularly significant for children with DD and ASD in particular who may have sensory integration difficulties incompatible with sleep and this point broadly advises parents to consider what these might be for their individual child.

Physiological advice.

This category includes 14 advice points around continence, diet and exercise which ensure the child is physically able to fall and stay asleep (Figure 5). The most commonly cited advice point: 'limit caffeine intake' 36% (n=19), advises parents to avoid food and drink high in caffeine around bedtime such as chocolate and carbonated drinks. Cerebra (2011) explain how caffeine acts as a stimulant and can prevent a child from settling to sleep. Advice points also include 'healthy diet limited in fat and sugar' (19%, n=10 of studies) and 'light meals only near bedtime' (19%, n=10 of studies) which link the child's ability to sleep with what and when they eat. The importance of 'daily exercise' (23%, n=12 of studies) was cited with some frequency and is particularly significant for children with DD who also have limited mobility and may not be physically tired enough for sleep. Linked to this is 'avoid late evening exercise' (23%, n=12 of studies) which dispels the myth that children should exercise vigorously close to bedtime to tire them. As further explained by Durand (2011) this practice raises core body temperature at bedtime, when it should lower to

promote sleep. In relation to continence advice points there was some divergence in the literature as to whether a child should limit fluids close to bedtime (Cerebra, 2011; Johnson et al., 1995) or stay hydrated (Early Support, 2010; National Autistic Society, 2008). This disagreement was dealt with by coding for two separate sleep advice points 'avoid fluids at bedtime and night' (6%, n=3 of studies) and 'encourage child to stay hydrated' (6%, n=3 of studies) and it is also important to note that fluid intake did not feature in the majority of SHE advice tools reported.

Communication adaptations.

This relates to five advice points which promote the understanding of children with DD and helping them learn how to change their sleep habits (Figure 4). Twenty seven percent (n=14) of studies reported the need to incorporate augmentative communication strategies such as social stories (National Autistic Society, 2008), visual timetabling (Early Support, 2010) or objects of reference (Leeds Community Healthcare, 2010). Twenty one percent (n=11) of studies made broader reference to the need to clearly communicate expectations around sleep to the child 'clear expectations, prompts and cues' and included strategies such as not confusing the child by bringing them back into the living area once the bedtime routine was complete (Handsel Project, 2012). Nineteen percent (n=10) of studies also suggested giving clear messages to the child about the use of the bedroom 'bedroom only used for sleep and calm activities'. Studies commonly referred to avoiding highly stimulating play in the bedroom so that the child learned to associate the environment with sleep.

2.7.8. Review objective 1. Conclusions.

Review findings provided a range of evidence to address the review purpose of identifying SHE content. Comprehensive evidence was found relating to sleep assessment and the need for a three step assessment process of physical and mental co-morbidity screening, sleep history taking and use of sleep outcome measures. Further content analysis of the data established six functions of sleep history taking and identified numerous sleep history tools, outcome measures, sleep outcomes and QOL measures. Mixed evidence was identified which related to the components of SHE. Using a content analysis method, SHE advice was organised into six categories incorporating 52 advice points, which represent a preliminary SHE tool. Conclusive evidence was presented for some advice points; however, others were supported by limited evidence and require further verification. Divergence existed in the literature regarding advice around the need to limit fluids near bedtime to promote nocturnal continence, which warrants further inquiry. Lastly, further qualitative evidence is required regarding parents' views around the acceptability of SHE advice to inform the further systematic development of a SHE tool for children with DD.

2.7.9. Review Objective 2. Identifying evaluative evidence of SHE.

This review objective was addressed by extracting data from 18 intervention studies and summarising findings using basic numerical descriptions and narrative summaries (refer Appendix 3).

2.7.10. Effectiveness of SHE.

Only two intervention studies investigated the effectiveness of SHE as a stand-alone intervention (Adkins, Molloy et al., 2012; Piazza et al., 1997) highlighting a limited

evidence base supporting SHE as an intervention in its own right. A small scale randomised controlled trial (n=36) of children with ASD, compared a detailed SHE intervention containing six component areas delivered unsupported by practitioners (in pamphlet form) with no intervention (Adkins, Molloy et al., 2012). Statistically significant improvement (P = 0.04) was reported in just one actigraph outcome- sleep efficiency (percentage of total sleep time or time spent in bed). Authors concluded that SHE delivered unsupported by practitioners was ineffective in improving sleep. These results were relevant to this review purpose and demonstrated the need to explore the efficacy of SHE accompanied with practitioner guidance. However, it is noted this study had limited applicability to children with ASD.

In a smaller scale randomised controlled trial (n=14) Piazza et al. (1997) compared a simple SHE intervention incorporating sleep timing and bedtime routine advice only with the behavioural sleep intervention, faded bedtime with response cost (FBRC) in children with DD. Participating children were in-patients in an assessment and treatment unit and the intervention group received SHE implemented by a professional. Piazza et al. (1997) concluded that based on reduction in mean hours of disturbed sleep, FBRC was more effective than SHE. However, the statistical significance of these results was not reported, sample size was small and methodological quality of the study was poor, therefore study findings should be viewed with caution. Furthermore, the study evaluated an incomprehensive SHE intervention delivered by sleep practitioners in a hospital setting which has limited applicability to current practice which is typically home setting based.

2.7.11. Effectiveness of multi-component SHE (MCSHE).

The majority of intervention studies (n=16) instead evaluated SHE as part of a package combined with behavioural interventions such as graduated extinction or sleep restriction. These studies therefore evaluated SHE with additional intervention content and different delivery methods, making them less relevant in addressing the review purpose of identifying evaluative SHE evidence. To aid clarity these mixed behavioural sleep interventions will be termed multi-component SHE (MCSHE) and incorporate at least two SHE advice areas. MCSHE was found to be effective in 10 studies which used a range of limited methodological designs such as multiple baseline across participants and uncontrolled pre-post designs (Allison et al., 1993; Bartlet & Beaumont, 2012; Beresford et al., 2012c, 2012d & 2012e; Bramble, 1997; Christodulu & Durand, 2004; Hewit, 1985; O'Connell & Vanaan, 2008; Reed et al., 2009; Weiskop et al., 2001; Weiskop et al., 2005).

Four intervention studies which evaluated MCSHE were randomised controlled trials (Cortesi et al., 2012; Malow et al., 2013, Montgomery et al., 2004; Stores & Stores, 2004); three of which had limited applicability to specific disability diagnosis. Montgomery et al. (2004) studied the broadest diagnostic group, children with learning disabilities (n=66) and found MCSHE to be effective when compared with waiting list controls (P<0.001). From a large sample size (n=160) of children with ASD, Cortesi et al. (2012) reported MCSHE was effective when compared with a placebo group (P<0.01). MCSHE was also compared with other interventions: melatonin and melatonin combined with MCSHE and MCSHE was reported to be less effective than comparator interventions. Malow et al. (2013) found MCSHE to be effective (P<0.001) in a sample of 80 children with ASD. Stores and Stores (2004) reported an improvement in composite sleep problem score (P<0.05) in children with

Down's syndrome (n=46), following MCSHE, however, no statistical improvement in actigraphy outcomes (P>0.05). In summary, studies largely reported MCSHE to be effective when compared with no intervention.

2.7.12. Complexity of SHE and MCSHE interventions evaluated.

There was considerable variation across intervention studies, in relation to the complexity of SHE or MCSHE and number of SHE advice component areas reported. No study investigated the effectiveness of interventions featuring all six SHE component areas identified in this review. The majority of intervention studies (n=13) evaluated treatments which incorporated between two and four SHE advice component areas, which most commonly included bedtime routines and sleep timing. This demonstrates how the majority of studies focused on basic and in-exhaustive SHE interventions and are of limited significance to the review purpose of identifying the evaluative evidence of SHE.

2.7.13. Quality of intervention studies.

In keeping with the characteristics of scoping reviews, studies were not quality assessed in a systematic manner (Arksey & O'Malley, 2005). However, the decision was taken to broadly consider the methodological quality of intervention studies to aid analysis and this was guided by the principles of the Critical Appraisal Skills Programme tool (CASP, 2013). Across randomised controlled trials blinding was not usually feasible for SHE and MCSHE interventions, so this affected the quality of results. Just two studies had sample sizes of over 50 (Cortesi et al., 2012; Montgomery et al., 2004) and most studies had a short follow up of less than six months. Precision of treatment effect was not commonly reported with only three studies reporting

confidence intervals (Beresford et al., 2012c, 2012d, 2012e; Bramble, 1997; Cortesi et al., 2012). In most pre-post and before and after studies, researchers did not adequately consider their relationships with participants introducing significant bias. Other pilot and multiple-baseline across participant studies were of poor methodological quality and could not provide any useful statistically significant results.

There was considerable variation in the comprehensiveness of sleep assessment reported in the 16 intervention studies. Only five studies took the most thorough approach to sleep assessment and engaged in a three-step process which involved, screening for physical and psychological co-morbidities, taking a sleep history and using sleep outcome measures. (Bartlet & Beaumont, 1998; Beresford et al., 2012c; Christodulu & Durand, 2004; Montgomery et al.,2004; Stores & Stores, 2004). This draws into question the validity of study results of the majority of intervention studies because it is unclear whether they are based on sleep problems that were clearly behavioural in cause. It is possible that other underlying sleep disorders, social and historical causes may have been overlooked in the largely incomplete sleep assessment stages which would render SHE and MCSHE less effective.

2.7.14. Review objective 2. Conclusions.

Review findings provided limited evidence to address the review purpose of identifying evaluative evidence of SHE. The majority of intervention studies evaluated MCSHE only, evidencing the need for further evaluative inquiry into SHE as a stand-alone intervention. The majority of intervention studies were of poor methodological quality and evaluated simplistic interventions incorporating a maximum of four SHE advice component areas. Furthermore, only five studies reported a thorough

assessment of sleep problems involving co-morbidity screening, sleep history taking and outcome measures, which called into question the validity of their results.

2.7.15 Review objective 3. Identifying what is known about how SHE works and issues around delivery method.

In order to find out what is known about how SHE works to improve sleep, data were firstly extracted from studies which reported proposed theories underpinning the intervention. Secondly, the reported views of stakeholders (parents and practitioners) regarding intervention delivery modes and related barriers and facilitators were narratively summarised. An exploration of such delivery issues helped to build a picture of how programme activities and contextual factors can activate change, providing some preliminary evidence for the development of an explicit SHE programme theory.

2.7.16. Theories underpinning SHE.

The majority of studies did not describe any theories explaining why SHE is supposed to work. Whilst intervention studies which tested hypotheses around intervention effectiveness could be said to be describing theory, only three studies made specific statements around how SHE works to achieve outcomes of improved sleep (Bramble, 1996; Cortesi et al., 2012; Piazza et al., 1997). The theories posited were brief and poorly defined (refer Table 8) and made broad reference to how encouraging good sleep hygiene can positively affect the regulation of sleep patterns or circadian rhythms. However, there was a lack of clear consensus on exactly which SHE advice components could achieve this, therefore it was difficult to draw any conclusions about SHE theory from the scarcity of research evidence available.

Table 8 Reports of SHE theory in included studies.

Study name	SHE theory reported
Bramble (1996)	"*The theory is that complete withdrawal of the primary reinforcing element (i.e. the parents' attention) will lead to rapid extinction of the child's demands while cueing the child into a regular sleep pattern by employing stimulus control techniques and promoting good sleep hygiene*" p. 358
Cortesi et al.(2012)	"*Healthy sleep practices also promote sleep and enhance sleep regulation by reducing environment stimulation and behavioural sleep conditioning, which reinforce the association of certain activities and environments with sleep, limit wake-promoting activities and may play a crucial role in sleep promotion.*"p707
Piazza et al.(1997)	"*By setting a regular sleep and wake time, the bedtime scheduling procedure might be hypothesized to maximise the likelihood that the individual's circadian rhythms would become entrained to the 24 hour cycle*" p418.

2.7.17. Effectiveness of modes of SHE delivery.

There was no evidence reported across studies pertaining to the relative effectiveness of different delivery methods where SHE was applied as a stand-alone intervention. All available evidence exploring the advantages and disadvantages of various modes of delivery were relevant to MCSHE only. Whilst these findings have some relevance to this review purpose, they are not entirely applicable to SHE as a stand-alone intervention.

2.7.18. Effectiveness of MCSHE modes of delivery.

A minority of intervention studies (6% n=3) compared the effectiveness of MCSHE delivery methods in terms of intensity of practitioner support and environment (Beresford et al., 2012c; Malow et al., 2013; Montgomery et al.,2004), but found largely inconclusive results. In a pilot study comparing home visits with telephone support Beresford et al (2012c) concluded that a small sample size (n=15) prevented evaluation of relative effectiveness. Malow et al. (2013) found sleep improvements were achieved regardless of one to one support or group workshop delivery. Lastly, Montgomery et al. (2004) did not detect any statistically significant difference between MCSHE delivered with and without one-to one sleep practitioner support.

Qualitative evidence was reported in three studies (Beresford et al., 2012a, 2012b & 2012g; Bramble, 1996; Weiskop et al., 2005) pertaining to the views of stakeholders about one-to one and group session MCSHE delivery methods (Table 9). The evidence consisted of direct stakeholder participant quotes and author opinion of stakeholder views, and was provided mainly by one qualitative study (Beresford et al., 2012a, 2012b & 2012g). Overall, the bespoke, supportive and flexible nature of one-to-one delivery modes was reported to be favourable for families however, there was some evidence to suggest that they could be time consuming and onerous on families (Beresford et al., 2012b; Weiskop et al., 2005).The group delivery method was reported to offer valuable peer support and could help parents to 'normalise' their child's sleep problem making it easier for them to address (Beresford et al., 2012b & 2012g). Conversely, challenges around venue selection, timings, mix of participants and facilitation style were reported as potential disadvantages of group events (Beresford et al., 2012d, 2012g).

Table 9 Summary of views on MCSHE delivery methods.

Method	Parent/ Practitioner views of delivery method.
One-to- one delivery. (Home visits, clinic sessions and telephone support)	*"Responses to the open ended questions (..) indicated that the best aspects of the programme were the outcome, the support provided, the phone calls and the method of instruction.( ) one mother said the sessions were too long. Three mothers referred to the overall time consuming nature of the programme"* Weiskop et al.(2005) p101-102.(Author opinion of parents views) (Home visits, clinic sessions and telephone support) *"Treatment approach was 'just right' for their children. Only a minority (three families) considered it 'rather tough"* Bramble (1996) p361. (Author opinion of parents views) (Home visits, clinic sessions and telephone support) *"A key theme to emerge from the accounts of parents experiencing these programmes was the value of its bespoke nature, by which they meant both a focus on their individual child/family and flexibility to suit the family in the timing, overall length and format of the support*." Beresford et al. (2012b) p180. (Author opinion of parents' views) (Home visits and telephone support). *"Indeed it was seen as having advantages over home visits because it was less intrusive and time consuming. This was particularly valued in the context that many parents already had multiple appointments regarding their child"* Beresford et al.(2012b) p181. (Author opinion of parents views) (Telephone support) *"Being in the house meant that the sleep practitioner could "pitch"(..) her advice to the environment, having seen it for herself"* Beresford et al. (2012b) p180. (Author opinion of parents' views) (Home visits) *"It's easier to talk to someone in your own home"* Beresford et al. (2012b) p180 (Direct quote from parent participant). (Home visits) *"I think the fact that they know someone is going to be coming back to ask how they're doing..it is such a difficult thing, if you're not getting sleep yourself. Just to have someone who is there going, "I know this is hard, but you are doing so well!" "* Beresford et al. (2012g) p 260 (Direct quote from practitioner participant) (Home visits)
Group delivery. (Group workshops)	*"Ninety four percent rated both the oral presentation and the booklet as "very easy" to understand; the rest considered each component "quite easy". The presentation was said to be "very useful" by 22%. "Quite useful" by 61% and "not very useful" by 17%"* Stores and Stores (2004) p66. (Author opinion of parents views) *"Caution was expressed that other working parents might find daytime sessions difficult.(...)Some parents commented on how the familiarity of*

	venue had helped them feel more confident about attending an intervention" Beresford et al. (2012b)p 179. (Author opinion of parents' views). *"The chief drawback of the workshop mode was its brevity as a programme, given the large subject area of sleep and participants having varying starting points in terms of their existing knowledge"* Beresford et al. (2012b) p183 (Author opinion of parents' views). *"It meant that their child's sleep problem was, in a sense, 'normalised' as accounts of the children's sleep problems were shared. For some parents, this in itself seemed to give them confidence to tackle their child's sleep"* Beresford et al.(2012b) p 186 (Author opinion of parents views) "*It doesn't suit everyone, but those that have experienced [the intervention] have had a very, very positive experience. They use each other's experiences and solutions to help, kind of, find strategies for themselves to work through*" Beresford et al. (2012a) p39. (Direct quote from practitioner research participant). "*Practitioners had observed that meeting other parents experiencing similar behaviour or sleep issues helped parents to realise they were 'not alone' or 'the only one; this in turn, helped reduce parents' sense of inadequacy and social isolation*" Beresford et al. (2012g) p 255 (Author account of practitioner views) "*Practitioners observed that parents appeared to find it less 'shameful' to share details of their child's problem behaviours in a group with other parents experiencing similar difficulties, than to talk about these directly to a professional in a one-to-one situation*" Beresford et al. (2012g) p 254 (Author account of practitioner views) *"Some practitioners believed it was better to match group members as far as possible in terms of the child's age and disability and parents' abilities in order to maximise their learning from each other and to maximise the 'group effect'."* Beresford et al. (2012g) p 254 (Author account of practitioner views) *"You need to actively engage with parents, for them to feel that they've got space to talk and that they're listened to and all those things. So yeah, that definitely has an impact on the effectiveness of the group" "* Beresford et al. (2012.g) p 258 (Direct quote from practitioner participant)

2.7.19. Facilitators and barriers to MCSHE delivery.

Minimal evidence pertaining to facilitators and barriers to MCSHE delivery was included in 10% (n=5) of studies, which reported direct stakeholder participant quotes and author opinion of stakeholder views. Furthermore, the majority of evidence was provided by a solitary primary qualitative study Beresford et al. (2012a, 2012 b, 2012f & 2012g). Themes were generated (Table 10) by following the principles of content analysis (Bowling, 2014), which guides the researcher in coding patterns within data. Interesting features within the data were coded and collated into two categories of enablers and barriers to MCSHE delivery.

Facilitators

'Continued practitioner support' described how parents needed sustained support to implement advice and maintain progress. Whilst this was supported relatively well by three studies, (Bartlet & Beaumont, 1998; Johnson et al., 1995; Beresford et al., 2012g) evidence was drawn from practitioner views only. The enablers: 'building a trusting relationship', 'specialist practitioner knowledge and skills', 'parents as sleep practitioners', 'acquiring new knowledge and understanding' and 'consistency of approach' were supported with more balanced evidence representing parent and practitioner perspectives but were extracted from just one or two studies (Bartlet & Beaumont, 1998; Beresford et al., 2012a, 2012b,2012f & 2012g).

Barriers

The barriers of 'parents' resilience' 'marital conflict' and 'separation anxiety' were reported by Bartlet and Beaumont (1998) and Beresford et al. (2012a, 2012f & 2012g). However, only 'parents' resilience' (which described how a parents'

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physical tiredness and emotional vulnerability could act as a delivery barrier), was

supported by the balanced view of both stakeholders. Evidence from one study

Beresford et al. (2012a, 2012f & 2012g) supported the barriers of 'child's condition',

'busy day to day lives', 'changes and disruptions' and 'home environment' which

were representative of both stakeholder views, and 'lack of readiness' which was

representative of practitioner views only.

Table 10. Summary of enablers and barriers to MCSHE delivery

Enablers to MCSHE delivery	Perceptions of Parents and Practitioners
Continued practitioner support.	"*Adherence to these principles frequently requires support and reinforcement from the PNP (*Paediatric Nurse Practitioner) *or Physician"* Johnson et al. (1995, p163) (Author opinion of practitioner views) "*Support and monitoring from their allied health worker was essential while developing positive sleep practices over several months. While the average time to achieve short-term sleep goals was 8 weeks, support may need to be ongoing and adaptable in order to meet family needs*" O-Connell and Vanaan (2008) p 214 (Author opinion of practitioner views). "*There was broad agreement amongst practitioners about the need for parents to receive ongoing support to help them to maintain positive outcomes in the longer term"* Beresford et al. (2012g) p 260 (Author opinion of practitioner views)

Building a trusting relationship	"*Parents who had numerous short-lived contacts with medical and para-medical professionals needed time to develop trusting relationships with the project workers*" Bartlet and Beaumont (1998) p 597 (Author opinion of practitioner views) "*Parents valued practitioners' non-judgemental approach; it showed respect for a parent's expertise*" Beresford et al. (2012b) p184 (Author opinion of parent views) "*Parents also welcomed not being pushed too hard by practitioners*" Beresford et al. (2012b) p185 (Author opinion of parent views) "*Practitioners believed that parents were more likely to accept support if there was a pre-existing, trusting relationship between themselves and a parent*" Beresford et al. (2012a) p45 (Author opinion of practitioner views) "*It works best where you can gain their trust and confidence over a period of time and you can reassure them that you will be there for the journey with them and are not going to drop out of their lives*" Beresford et al. (2012a) p45 (direct practitioner quote).

Specialist practitioner knowledge and skills	*"Some parents explicitly mentioned that the disability-specific nature of the programme had been an important factor behind their decision to embark on a sleep intervention (..) a mother explained that it had been important to know that the programme would be run by the Learning Disability team"* Beresford et al.(2012b) p.177 (Author opinion of parent views).
	"The practitioner's specialist knowledge of sleep was valued by parents, especially by those who had previously struggled to access the information from 'generalists' (such as paediatricians, or community-based nurses" Beresford et al. (2012b) p185 (Author opinion of parent views).
	"Practitioners believed that their skills and knowledge were essential to the effectiveness of the programme they delivered. A certain level of professional qualification and experience, sometimes alongside specific training, is required to deliver all these programmes" " Beresford et al. (2012g) p 257 (Author opinion of practitioner views).
Parents as sleep practitioners	*"One of the workshop facilitators was also a parent who had experienced sleep problems with her child. This facilitator overtly drew on this experience during the workshop and parents clearly valued this connection" "* Beresford et al. (2012b)p 183 (Author opinion of parents views).
	"*But it was wonderful to have a parent cos they could actually say I know exactly what you mean…and (*were) *happy talking about their own experiences" "* Beresford et al. (2012g) p 259 (direct quote of practitioner participant).

Achieving new knowledge and understanding	*"Parents found it reassuring to have confirmation from the experts running their sleep programme that at least some of what they had already put in place was 'right' and should continue" "* Beresford et al. (2012f) p 236 (Author opinion of parents views). *"The acquisition of this new knowledge and understanding appeared crucial. It was enmeshed within parents' accounts of positive changes in their attitude towards their child's sleep problem. Taken together, these changes in understanding and attitude paved the way for the new or adjusted parent behaviours which proved key to improving the child's sleep"* Beresford et al. (2012f) p 236 (Author opinion of parents views). *"Practitioner identified the educational aspects of the programmes as a key factor in supporting positive outcomes"* Beresford et al. (2012g) p 252 (Author opinion of practitioner views).
Consistency of approach	*"Parents reported it was important that, when implementing new approaches to managing bedtime or night wakenings, others involved with the child were willing to follow a consistent approach, or otherwise act in a way which supported their goals"* Beresford et al. (2012f) p 243 (Author opinion of parents views). *"Some programmes positively encouraged, where appropriate, the direct involvement of both parents and /or other primary carers (such as grandparents). The practitioners who developed these programmes firmly believed it increased the potential effectiveness of the intervention"* Beresford et al. (2012g) p 256 (Author opinion of practitioner views).

Barriers to MCSHE delivery	Perceptions of Parents and Practitioners
Lack of readiness	"*The notion of readiness was a recurrent theme in professionals' accounts. All believed that, unless a parent, or couple, wanted to do something about their child's sleep or behaviour, then it was very unlikely they would accept the offer of support*" Beresford et al. (2012a) p43 (Author opinion of practitioner views). "*Sometimes, level of engagement in a 'pre-intervention task' provided an indicator of a parent's readiness to engage (..)If families are reluctant to do any sort of sleep diary, who absolutely won't put anything down at all, I find they are often the families we struggle to move forward*" Beresford et al. (2012a) p40 (Direct practitioner quote).
Parents' resilience	"*It became apparent early in the project that about one third of parents would not be able to work at a behavioural programme straight away. Physical exhaustion was much in evidence.*" Bartlett and Beaumont (1998) p 597.(Author opinion of practitioner views). "*Parents said that on days when they felt especially tired it was more difficult to maintain optimum behaviours with regard to sleep, such as sticking to a good bedtime routine, responding in the best way to night wakenings, and/or filling in their sleep diary*" Beresford et al. (2012f) p 246 (Author opinion of parents views). "*A number of practitioners observed that individual differences in resilience had a bearing on the potential impact on a programme on parent and child outcomes. Some parents, they observed, were better able to deal with the ordinary and the extraordinary stresses of life, and so had a greater capacity to remain consistent and stick with the sleep or behaviour management strategies*" Beresford et al. (2012g) p 264 (Author opinion of practitioner views).

Marital conflict	*"Marital conflict was common and meant that spouses did not agree about ideas that were put forward for changing the 'status-quo'"* Bartlett and Beaumont (1998) *p 597* (Author opinion of practitioner views). *"Professionals delivering these interventions identified additional issues such as ( ) relationship issues which may ( ) need to be addressed"* Beresford et al. (2012a). p 40 (Author opinion of practitioner views).
Separation anxiety	*"The longing for child free nights was counterbalanced by the fear that the children would suffer as a result of nocturnal separation (..)Experiences pertaining to the perinatal period preoccupied many mothers"* Bartlett and Beaumont (1985) p 597. (Author opinion of practitioner views). *"Professionals delivering these interventions identified additional issues such as ( ) attachment and relationship issues( ) which may also need to be addressed"* Beresford et al. (2012a) p 40 . (Author opinion of practitioner views).
Child's condition	*"Parents identified features of the child's condition which, they believed, limited sleep improvements or could mean progress was more fragile. This was particularly the case for children with ASC or children with on-going health needs"* Beresford et al. (2012f) p 245 (Author opinion of parents views). *"And so you might have that people are very committed or that they are interested and then the child becomes un well and then that's not a good time"* " Beresford et al. (2012g) p 263 (Direct quote practitioner participant).

	"Practitioners reported that parents of children with on-going acute or complex health needs typically found it more difficult to adhere to a sleep management strategy because they had real fears about the health and safety of their child during the night" Beresford et al. (2012g) p 267 (Author opinion of practitioner views).
Busy day to day lives	*"The pace and complexity of everyday 'real life' for parents could make it difficult to sustain their focus on the child's needs regarding sleep"* Beresford et al. (2012f) p 246 (Author opinion of parents views). *"All the practitioners recognised that the everyday pressures of family life made it difficult for parents to find the time to reflect on their learning, to put it into practice and to remain consistent in their parenting approach" "* Beresford et al.(2012g p 262 (Author opinion of practitioner views).
Changes and disruptions	*"Parents found maintaining sleep routines during school holidays were particularly difficult, especially when they were away from their usual surroundings. Weekends, with the lack of school routines, presented this difficulty"* Beresford et al. (2012f) p 246 (author opinion of parents views). "*Christmas, family holidays, all those kinds of things when life's nor running just as it should be…that can be incredibly challenging for parents. So I think sometimes the sleep problems can slip back"* Beresford et al. (2012g) p 263 (Direct quote from practitioner participant).
The home environment	*"Parents judged that aspects of the home environment placed limits on progress with their child's sleep in a variety of ways. This was particularly an issue where the child shared a bedroom with siblings"* Beresford et al. (2012f)

	p 246 (Author opinion of parents views).
	"Families who are in social housing: you're lucky if you've got a three-bedroomed house. So if you've got children with disabilities (..) and you've got other children sharing…I think that's one of the huge things. That is definitely an issue" Beresford et al. (2012g) p 263 (Direct practitioner participant quote).

2.7.20. Review objective 3- conclusions.

Findings provided limited evidence to address the review purpose of identifying what is known about how SHE works and issues around modes of delivery. Only three studies posited brief and poorly constructed theories explaining why SHE is supposed to work. Evidence which compared methods of delivery and identified barriers and enablers was inconclusive and applicable only to MCSHE. Furthermore, no evidence was reported relating to organisational or policy factors and how these affected intervention success. This highlights the need for further inquiry into how SHE works, programme activities and contextual factors which can activate change.

2.7.21. Discussion.

Using Arksey and O'Malley's (2005) scoping framework this review asked "what is known about SHE as a treatment for behavioural sleep problems in children with DD?" and aimed to identify SHE content, evaluative evidence and explore what is known about how SHE works and issues around delivery methods.

The identifying relevant studies stage identified 2203 potential studies demonstrating a search of adequate breadth and depth. The study selection phase then identified 273 studies eligible for inclusion and involved two stages of independent reviewer checks to ensure the chance of selection bias was minimised. However, obtaining this large number of full texts was time consuming and the checking process was challenging to complete within project timescales. A detailed assessment of the methodological quality of the final 52 included studies using quality appraisal tools was not undertaken and no studies were excluded on the basis of poor quality as exemplified by the scoping review method. However, the methodological quality of

intervention studies was broadly considered to aid analysis, guided by the principles of CASP (2013).

The 'charting the data' phase followed a standardised approach for all 52 included studies, which demonstrated transparency in the review process. It was not possible to extrapolate all required data because relevant information was not provided in every study. It was time consuming to identify the different versions of sleep assessment tools used in intervention studies, and only broad descriptions of SHE advice were often reported, limiting the quality of data extracted.

The result reporting stage firstly involved analysing data using descriptive numerical accounts, content analysis and descriptive narrative summaries. This stage identified a plethora of evidence on the sleep assessment process, sleep history tools and outcome measures, demonstrating the fundamental importance of assessment in SHE delivery. Yet the majority of intervention studies failed to utilise this evidence and execute a sufficiently comprehensive assessment, calling into question the validity of their results. This links with personal clinical experience which informs that inadequate sleep assessment is common in practice. This demonstrates the need for more robust evaluative SHE studies and programme theory development which makes explicit how assessment should be an integral part SHE implementation.

The reporting stage also produced a preliminary SHE tool consisting of six advice categories and 52 advice points, which was underpinned by mixed evidence mainly reported in the grey literature. Further research is needed to strengthen the evidence base, reach agreement on specific advice points and explore parental acceptability of the tool. This demonstrates how the first review objective to identify SHE content was addressed.

Review evidence also highlighted paucity in evaluative SHE research. The majority of intervention studies analysed MCSHE only and it was difficult to ascertain the level of impact SHE had on sleep outcomes. These studies were generally of poor methodological quality (although quality was not formally assessed using appraisal tools). Studies evaluated interventions with a maximum of four component areas and did not accurately demonstrate the potential of SHE as a stand-alone intervention. This concurs with personal experience of clinical practice where SHE of limited content is delivered in combination with other behavioural strategies. Furthermore, where there was evidence of SHE intervention studies, sample size was small highlighting the need for larger scale SHE specific trials to better inform practice. This demonstrates how the second review objective to identify evaluative evidence was addressed.

The third review objective aimed to identify what is known about how SHE works and issues around delivery method. This was poorly addressed by review evidence. A few studies posited brief theories which made broad reference to how encouraging good sleep hygiene can positively affect the regulation of sleep patterns or circadian rhythms. Limited evidence was also reported comparing delivery methods and exploring possible barriers and enablers. Furthermore, no evidence was found which addressed organisational or policy factors affecting the delivery of SHE. This identifies a need for stakeholder questioning around what makes SHE work and factors affecting implementation, which will inform the systematic development of a SHE programme theory. Overall, review objectives two and three were poorly addressed by the literature identified and a summary of review results in relation to all three objectives is presented in Table 11.

Table 11: Scoping review findings.

Review objective 1: Identification of SHE content.
Comprehensive evidence relating to sleep assessment   - Evidence to support the need for a three step assessment process including physical and mental co-morbidity screen, sleep history taking and use of sleep outcome measures.   - Six functions of sleep history taking established.   - Numerous sleep history tools, outcome measures, sleep outcomes and QOL measures identified.   **Mixed evidence identifying the components of SHE**.   - Six categories of SHE advice components identified, incorporating 52 advice points.   - Conclusive evidence for some SHE advice points only.   - Disagreement regarding advice to limit fluids near bedtime to promote nocturnal continence   - No qualitative evidence reported on parental acceptability of SHE advice.

Review objective 2: Identification of evaluative evidence
Limited evaluative evidence of SHE.   - The majority of intervention studies evaluated MCSHE only, evidencing the need for further SHE specific evaluative inquiry.   - Most studies were of poor methodological quality and evaluated simplistic interventions incorporating a maximum of four SHE advice component areas.   - Most studies did not report a comprehensive sleep assessment; calling into question the validity of their results.

Review objective 3: Identification of what is known about how SHE works and issues around delivery method.
Limited evidence identifying what is known about how SHE works and issues around delivery method.   - Only three studies posited brief and poorly constructed theories explaining why SHE is supposed to work.   - Evidence relating to delivery method was reported only in studies which evaluated MCSHE   - Limited and inconclusive evidence was reported which compared delivery methods and explored enablers and barriers.   - No evidence was reported on organisational and policy factors affecting the delivery of SHE.

2.7.22. Conclusion.

This review contributes to an emerging evidence base supporting SHE as a first-line intervention for behavioural sleep problems in children with DD. It has identified comprehensive literature underpinning the sleep assessment process and developed a preliminary SHE tool underpinned by mixed evidence. Next steps for research include exploring how this knowledge translates into practice and developing the SHE tool by establishing stakeholder agreement and acceptability of SHE advice.

The review has identified a need for high quality evaluative studies which focus on SHE as a comprehensive, stand-alone intervention for sleep problems which have been comprehensively assessed. Furthermore, review findings have found paucity in research around SHE theory and delivery approaches, which highlights the need for further development of underpinning programme theory. This will make explicit how SHE should be delivered and inform more theory-driven evaluations of this sleep intervention in its' own right. The results of such specific SHE evaluative research will provide professionals with a more credible evidence base, increasing their confidence in this intervention and supporting a more consistent approach in practice. The following chapter will now present the methodological underpinnings of subsequent exploratory and co-design studies.

CHAPTER 3: METHOD

3.1. Introduction

Following on from the scoping review which identified the need to examine key exploratory issues around SHE theory and intervention delivery approaches, this chapter describes the methodological underpinnings of the subsequent exploratory and co-design studies. The rationale for adopting a participatory perspective to guide study designs will be outlined, through exploring alternative approaches such as positivist, post-positivist and constructivist paradigms. Arguments are then presented for selecting an Experience Based Co-Design (EBCD) framework to structure the exploratory and co-design studies, through exploring qualitative methodologies which could have been utilised. An exploration of qualitative design issues, qualitative data collection and analysis methods follows. This chapter concludes with a summary of the design of studies presented in this thesis.

3.2. Philosophical Assumptions

It is good practice for researchers to make explicit how their philosophical assumptions guide the conduct of a research study (Broom & Willis, 2007; Creswell, 2007). This encompasses a researcher's ontological position that reflects how they view the nature of reality, and epistemological position which relates to their understanding of the nature of knowledge. For example, a constructivist researcher will assert that reality is subjective and knowledge is socially constructed, whereas a positivist researcher will argue that reality is fixed and that it is possible to produce objective knowledge through using systematic methods. Once inquirers make a choice regarding these assumptions, their research is designed according to a

paradigm or worldview which is a set of beliefs that guides such activity (Guba & Lincoln, 1989).

3.3. Overview of Paradigms

3.3.1. Positivism.

On a virtual scale of inquiry paradigms, positivism has traditionally been placed at one extreme. The positivist ontology which is sometimes referred to as naive realism, maintains that a real world exists with provable patterns that can be measured (Broom & Willis, 2007; Willig, 2013). The presumption is that a single, objective reality exists which can be determined through scientific inquiry. A positivist epistemology is often termed dualist and objectivist, where the researcher and research participant are independent of each other and the researcher is capable of remaining fully impartial at all times during the research process (Guba & Lincoln, 1994). A positivist researcher seeks to establish the facts of social phenomena, and produce objective, un-biased knowledge. A purely positivist approach to social research has been rejected based on the belief that real knowledge (in contrast to beliefs) is constrained to what can be scientifically proven (Patton, 2015). The positivist approach severely narrows what can pass for knowledge and places unrealistic demands in terms of certainty upon the complex social world (Patton, 2015). The researcher's clinical practice has shown the experience of families of children with DD and SHE to be diverse and individualised; therefore, adopting a positivist approach towards exploring their perceptions would place unhelpful limits on knowledge creation.

3.3.2. Realism.

The realism paradigm emerged in response to arguments against positivism. The realist ontology maintains that reality exists independently of our understanding of it and learning about reality is constricted to the here and now (Stickley, 2006). In other words, this ontological perspective maintains that the world is constructed by many different systems including material, social and psychological and it is important to believe that such theoretical constructs really exist (Westhrop et al., 2011). The epistemological position of realists recognises that new knowledge is historically and socially created and it is important to capture these relationships and causes using rigorous methods (Patton, 2015). Patton explicates how the realist perspective acknowledges that absolute objectivity is impossible to achieve and imperfections or variations in a diverse social world exist, but objectivity in the form of more or less plausible beliefs is worth aspiring to. In the context of this research, a realist approach offered more promise in terms of exploring stakeholders' perspectives of SHE, however, it was considered too restrictive in gaining a comprehensive understanding of their individual experiences.

3.3.3. Social constructionism and constructivism.

Social constructionism lies at the opposite end to positivism on the virtual continuum of research paradigms. It starts with the principle that the human and natural worlds are different and so should be analysed differently (Guba & Lincoln, 1990). Social constructionist ontology is often referred to as relativist and "holds that all tenable statements about existence depend on a worldview and no worldview is uniquely determined by empirical or sense data about the world" (Patton, 2015, p.122). It maintains that realities are socially constructed by individuals making sense of their

experiences and each belief is valid and worthy of equal respect (Willig, 2013). The epistemological position of social constructionists is often referred to as subjectivist, in that researchers are concerned with understanding the social construction of knowledges instead of describing reality from an entirely impartial and unbiased stance (Patton, 2015).

The worldviews of constructivism and social constructionism are commonly referred to interchangeably because they are based on the same ontological and epistemological principles (Patton, 2015). Crotty (1998) offers a distinction to clarify meaning. Crotty suggests constructivism focuses on the ability of individuals to make sense of their experiences and places value and respect on these unique realities. Social constructionism centres on the influence of the social world in terms of how individuals perceive the world. Crotty asserts that social constructionism then describes the influence society and culture has on individuals, and impacts on the way in which we understand and experience phenomena. The example of how differently a western culture views wealth compared to a developing country, further explains the hold society has on the way individuals view things. In the context of this research, social constructionism was a strong contender as a methodological underpinning, because of its ability to acknowledge how society influences the way children's sleep problems are viewed and experienced. However, the participatory approach seemed even more appropriate as it involved an element of action which could improve the delivery of SHE.

3.3.4. Participatory inquiry paradigm.

Proponents of the participatory (or advocacy) paradigm argue that constructionism does not go far enough in advocating on behalf of individuals (Creswell, 2007).

Creswell explains that the participatory approach focuses on research which is collaborative 'with' others as equal research partners, rather than 'on' them as subjects, and also incorporates an action agenda for reform that aims to improve individuals' lives. A participatory ontology is subjective / objective which holds that the mind actively participates with worlds and people (cosmos) and creates a reality as a result of this interaction (Heron & Reason, 1997).

In describing the epistemological position of the participatory paradigm, Heron and Reason (1997) define four interdependent ways of knowing: propositional, practical, experimental and presentational. Propositional knowing refers to the process of people collaborating to define questions to explore, practical knowing describes how they apply this methodology to their world, experiential knowing leads them to new forms of encounter with their world and lastly, presentational knowing refers to how they represent this experience as significant patterns. Heron and Reason continue to expound how participative inquiry is grounded in political participation which deems that people have a right to fully participate in research alongside researchers as equal collaborators. Many versions of participative inquiry have been developed including 4th Generation Evaluation which focuses on the claims, concerns and issues of stakeholders as a basis for gathering information and emphasises negotiation and empowerment in achieving organisational change (Guba & Lincoln, 1989). In addition, Action Research (Reason & Bradbury, 2001) synthesises action and reflection, practice and theory to develop practical solutions which are important to individuals and their communities. However, the overarching principles of a participatory approach seemed the most appropriate to guide the conduct of this research and a discussion on paradigm selection now follows.

3.3.5. Paradigm selection.

A positivist paradigm was considered inappropriate for the series of studies presented in this thesis, based upon common criticisms around its inability to uncover and understand the intricacies of the social world. A realist approach which believes knowledge is relative rather than absolute and objectivity in the form of more or less plausible beliefs is possible, did not seem to go far enough in its attempts to uncover social complexity and therefore this was also rejected as a guiding philosophy. The worldview of social constructionism which purports that multiple valid realities exist and influenced by culture instead, seemed to offer more potential to get to the truth and explore the complexities of stakeholder experiences. However, the participatory paradigm which builds upon the constructionist view and seeks also to involve individuals as equal partners in research process, was selected as the most appropriate philosophy to steer the studies presented in this thesis. This was because the participatory approach leads the researcher to co-construct with stakeholders a product with potential to invoke social change (Reason & Bradbury, 2001). As Reason and Bradbury point out a participatory worldview seeks to do more than just illuminate different perspectives and create new knowledge; it commits the researcher to effecting collaborative change. This then led the researcher to use Experience Based Co-Design (EBCD) (The King's Fund, 2014) as a guiding qualitative method to structure this research as it is rooted in the principles of the participatory paradigm.

3.4. Experience Based Co-Design (EBCD) Method

EBCD (The Kings Fund, 2014) is primarily a health service development tool which draws on qualitative data and melds the perspectives of stakeholders (service users

and staff) in the design of service improvements. The King's Fund developed the approach to put service users at the forefront in improving care delivery and recommends a staged series of activities (Figure 7). Facilitators observe services to learn how they operate then perform video recorded interviews with stakeholders to explore service perceptions. Service user narratives are edited to create a 'trigger film' which summarises emotional 'touch-points' and are used to promote dialogue between stakeholders about service improvement priorities. Stakeholder feedback events and co-design groups are then facilitated which gradually bring stakeholders together, to design quality improvements. The process concludes with celebration events to evaluate outcomes and the impact of EBCD. This process empowers stakeholders to plan for service change as equal co-design partners.

Figure7 Stages in Experience Based Co-design (The King's Fund, 2014).

Observe clinical areas – gain an understanding of what is happening on a daily basis

Interview staff, patients and families – exploring niggles

Edit interviews into 25-30 minute film of themed chapters

Hold staff feedback event – agree areas staff are happy to share with patients

Hold patient feedback event - show the film to patients. Agree improvement areas

Hold joint patient-staff event to share experiences and agree areas for improvement

Run co-design groups to meet over 4-6 month period to work on improvements

Hold a celebration event

Project steering group
meets at critical stages:

1. Before the project starts

2. Before feedback events

4. After first co-design group

5. After celebration event

More than 50 healthcare organisations worldwide have adopted EBCD and have shown it to be an effective health service improvement method (Donetto, Tsianakas & Robert et al., 2014). The benefits of EBCD are well documented and its ability to promote service user engagement and give voice to their experiences is repeatedly cited across studies (Bate & Robert, 2006; Donetto et al., 2014; Locock et al., 2014; Piper et al., 2012; Tsianakas et al., 2012).

However, it was important to be mindful of method limitations, such as problems with engaging staff in the co-design process (Donetto et al., 2014). If staff belonged to an organisational culture that was distrustful of service user participation projects, it was important to gain their confidence to motivate them to take part. Piper et al. (2012) and Tsianakas et al.(2012) also reported a challenge common to other service improvement methods; the tension between service resource constraints and expectations of change. They cautioned against setting unrealistic goals and the need to be aware of organisational capacities and resources. The researcher was therefore mindful of these limitations when designing the studies presented in this thesis.

3.5. Research Methodologies

To ensure EBCD offered the most suitable framework to guide this research, it was important to explore qualitative methodologies which may have also been useful. Within the qualitative tradition, Tesch (1990) identified 27 varieties of research methodologies such as ethnography, case study and narrative research. Phenomenology and grounded theory were selected for consideration. An appraisal of these methodologies and rationale for discarding them now follows.

3.5.1. Phenomenology.

Phenomenology as a methodology aims to establish and illustrate the subjective, lived experience of research participants from their personal view-points (Patton, 2015). This methodology also seeks truth and understanding from the individual experiencing phenomenon and views the world as they live it, and the 'essence' of the experience is captured (Polit & Beck, 2006). Phenomenology could have been appropriate as it provides deep and meaningful data about the individual's experience; however, in focusing on rich description the development of actions for change or solutions to problems are not considered. With this research project came an inherent need to improve SHE content and the way it is delivered; therefore, phenomenology was rejected as a methodology for this research.

3.5.2. Grounded theory.

This methodology focuses on developing theory generated or 'grounded' from participant interviews to provide an explanation about a social process (Strauss & Corbin, 1990). It involves concepts such as the 'constant comparison of data' whereby concepts are coded and analysed concurrently, and 'theoretical sampling' where the researcher makes recruitment decisions inductively to develop a theory as it emerges and until data saturation is achieved (Creswell, 2007). The researcher was drawn to grounded theory because of its emphasis on generating theory which is not pre-formed and ability to draw rich data from individuals. However, the researcher was concerned with how the approach can overlook the inquirer's significant agency in data construction and analysis (Bryant & Charmaz, 2007).This methodology was therefore considered inappropriate for the studies presented in this thesis which were

guided by a participatory approach that empowers stakeholders to act as more equal research partners in theory development.

In conclusion, whilst the strengths of alternative methodologies were acknowledged in getting to the complexities of human experience and generating theory, EBCD was selected as an approach most closely aligned to a participatory paradigm. EBCD focuses on exploring stakeholder experiences and actively involving participants as equal partners. In addition to inquiry, EBCD also focuses on action whereby stakeholders are encouraged to think about possible solutions to challenges.

3.6. Qualitative Design Issues

The strengths of qualitative research include flexibility (which allows the use of creative research methods) and reflexivity (which permits researcher intuition to enter into the participant's world to gain a deep understanding of phenomena) (Parahoo, 2014). Allsop (2013) adds that in achieving a rich description of participants' perspectives rooted in social and cultural contexts, qualitative research has high internal validity (the extent to which findings are a true reflection of reality and not affected by external variables). However, Allsop cautions that qualitative design is criticised for poor external validity (where the objectivity of findings and how representative they are of participants is unclear). Parahoo (2014) contests that this limitation is addressed by credibility through demonstrating how researcher interpretations are checked against respondent's views and qualitative findings relate to existing knowledge in the final analysis. Parahoo highlights other criticisms and counter-arguments relating to qualitative inquiry including limitations around reliability, which is replaced by auditability through making explicit, data collection and analysis methods to allow for study replication. It is important to be aware of the

strengths and limitations in qualitative inquiry and how these can affect rigor. Furthermore, it is prudent to acknowledge that study integrity can be heavily reliant upon researcher competence in exploring and representing people's experiences (Patton, 2015).

3.7. Qualitative Data Collection Methods

The data collection methods of interviews and focus groups, which are commonly used in qualitative research, are now appraised as part of the study design process. In addition a novel approach to data collection is described, which is the creation of an audio podcast to trigger discussion amongst research participants.

3.7.1. Interview method.

An interview is concisely defined by Berg (2007) "as a conversation with a purpose" (p.89) which allows the researcher to enter into the interviewee's perspective to co-create meaning. It starts with the assumption that the experiences of others are meaningful and can be explicated, and that the interview process acts as a vehicle to gather their stories (Patton, 2015). The semi-structured interview method is a widely used technique in qualitative research which guides the researcher using a framework of topic areas whilst allowing freedom to ask additional questions and build a conversational style (Bloom & Crabtree, 2006). This technique, however, requires the inquirer to be confident in their rapport building skills, to build a trusting atmosphere with the interviewee where they feel safe to share their experiences and engage in frank disclosure (Patton, 2015). Based upon the established use of semi-structured interviews within qualitative research and ability to uncover participant experiences, the decision was taken to utilise this method within this research.

In terms of the modality of this data collection method the options of face-to-face or telephone interviews are available to the researcher. Within qualitative research, face-to-face interviewing has traditionally been the preferred option over telephone interviewing as it facilitates rapport building and the collection of rich, contextual data. (Novick, 2008). However, telephone interviews can make respondents feel more relaxed and able to disclose sensitive information due to their relative anonymity in comparison to face-to-face interactions (Novick, 2008). They also allow for greater geographical coverage and are more cost efficient than face-to face interviews (Sturges & Hanrahan, 2004). These potential merits informed the decision to include telephone interviews as a suitable data collection technique alongside face-to face interviews, and indeed it was felt that some participants may prefer to discuss the relatively sensitive parenting topic of sleep over the telephone.

3.7.2. Focus group method.

Focus groups are a type of group interviewing but it is important to differentiate between the two. Group interviewing involves simultaneously interviewing a number of people, the focus being on questions and answers between the participants and researcher. In contrast, focus groups depend on interaction within the group, based on topics presented by the researcher (Morgan, 1997). Hence the fundamental characteristic which characterises focus groups is the understanding and data elicited from the discussion between participants. Focus groups offer a method of listening and learning from others, whereby communication channels are created which connect the separate worlds of participants and the researcher (Morgan, 1998).They ask focused questions, aim for discussion not agreement, operate in a safe environment, consist of up to 12 homogenous participants and are skilfully

facilitated (Krueger & Casey, 2010). Focus groups aim to collect data in a social setting where participants can contemplate their own opinions in relation to others (Robinson, 1999) and clarify ideas in ways that would be difficult to achieve in a one to one interview (Kitzinger, 1995). They can also be used to evaluate programmes of work or ways of working (Krueger & Casey, 2010). As with all techniques, limitations exist such as the challenge of moderating dominant respondents in the group and the risk of minority opinions being overshadowed (Patton, 2015). However, focus groups were selected as an appropriate data collection method in the present research, based on their potential to encourage such stakeholder/researcher collaboration (aligned to a participatory approach) and ability to elicit a range of experiences and views from a sample in short space of time (Plummer D' Amato, 2008; Stewart & Shamdasani, 1990).

3.7.3. Podcasts.

As explained earlier in 3.4, the EBCD method (King's Fund, 2014) incorporates 'trigger films' developed from service user narratives, which are used as a method of qualitative data collection. The theory is participants will personally connect with emotional touch-points presented in the film which then stimulates rich discussion. Indeed, the merits of multi-media techniques in enhancing individual's engagement with subject matter and appeal to visual and auditory learners have been well documented (Boulous, Maramba & Wheeler, 2006). However, the audio channel of communication may be particularly powerful in terms of stimulating the creation of mental images in the minds of listeners and evoking memories of personal experience (Rodero, 2012). Therefore, the use of audio podcast was considered as a suitable and creative method to draw out participant's views and perceptions of SHE.

3.8. Analysing Qualitative Data

Qualitative data can be analysed using a deductive or inductive approach (Burnard, Gill, Stewart, Treasure & Chadwick, 2008). The deductive approach involves using an 'a priori' or predetermined structure to analyse data. In contrast, the inductive approach is not guided by a pre-agreed framework but utilises the data itself to guide the analysis. There was limited specific evidence to draw from to demonstrate the data analysis process used for EBCD studies. In Tsianakas et al's. (2012) EBCD study, thematic analysis was alluded to as an analysis method, whereas Locock et al. (2014) outlined a combination of framework and thematic analysis approaches. Therefore, whilst it is acknowledged that a variety of approaches to data analysis exist rooted in methodological traditions such as discourse analysis and interpretative phenomenological analysis; framework and thematic analysis were selected for appraisal.

3.8.1. Framework method.

This method was developed for use in policy and health research and is distinguished by a matrix of summarised data which the researcher uses to structure and systematically reduce the data in order to analyse it (Spencer, Ritchie, Ormston, O'Connor & Barnard, 2013). It involves using pre-agreed themes to structure the analysis of data, but also allows for themes to be generated from the data through open coding (Gale, Heath, Cameron, Rashid & Redwood, 2013). Gale et al conclude that framework analysis offers a systematic and flexible approach which can be adapted for use with inductive and/or deductive types of qualitative data analysis. This method could have been appropriate for the studies presented in this thesis,

however, the following method was more closely aligned to the principles of a participatory paradigm.

3.8.2. Thematic analysis.

This approach guides the researcher in establishing, analysing and describing patterns or themes within data and involves six distinct stages (Table12) (Braun & Clarke, 2006). Stage one involves the researcher familiarising themselves with the data. Stage two encompasses generating preliminary codes. Stage three prompts the researcher to collate codes into preliminary themes. Stage four involves a review of the themes and checking they are well supported by data extracts. Stage five refers to the final refinement of themes and development of clear thematic definitions. Lastly, stage six concludes with the final report and presentation of themes which relates back to the original research question. Braun and Clarke purport it is a flexible method which is not wed to any specific epistemological position and can be adapted to analyse data inductively or deductively and is a widely used method in qualitative research (Burnard et al., 2008). Thematic analysis is also suitable for early career researchers because it provides the building blocks useful for performing a wide range of other qualitative analysis (Dawson, 2013). Furthermore, it is specifically recommended for studies situated within a participatory paradigm, where research participants are classed as co-researchers (Braun & Clarke, 2006) therefore this was selected as the method of analysis.

Table 12 Phases of thematic analysis (Braun & Clarke, 2006) (p35).

Phase	Description of the process
1.Familiarising yourself with your data:	Transcribing data (if necessary), reading and re-reading the data, noting down initial ideas.
2.Generating initial codes:	Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code.
3. Searching for themes:	Collating codes into potential themes, gathering all data relevant to each potential theme.
4. Reviewing themes:	Checking the themes work in relation to the coded extracts (level1) and the entire data set (Level 2), generating a thematic 'map' of the analysis.
5. Defining and naming themes:	Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells; generating clear definitions and names for each theme.
6. Producing the report:	The final opportunity for analysis. Selection of vivid, compelling extract examples, final analysis of selected extracts, relating back to the analysis of the research question and literature, producing a scholarly report of the analysis.

3.9. Design of Studies Presented in this Thesis

EBCD has been shown to be a versatile method and numerous projects have adapted the standard design (Figure 7) to different settings and budgets (Donetto et al., 2014; Locock et al., 2014). The King's Fund (2014) advises that whilst modifications to EBCD are possible, the fundamental elements of service user interviews and interaction between stakeholders should be maintained. It was therefore felt that EBCD could be adapted to meet project timescales and had the potential to be utilised in a novel way to co-create a SHE tool and supporting programme theory which could ultimately lead to service improvements. First, the researcher's previous Learning Disability Nursing experience of working within sleep teams provided sufficient insight into how sleep services operated which mirrored the introductory observation phase detailed in figure 7. Second, researcher experience and scoping review findings then informed development of the next study to be presented in this thesis- the exploratory study which reflected the interviewing stage of the EBCD

process. This second study aimed to build the qualitative evidence base for SHE through interviewing parents of children with DD and sleep problems and facilitating focus groups with sleep practitioners (professionals who support families with children's sleep problems). A thematic analysis of exploratory study findings then produced thematic findings which represented parent and sleep practitioner views. These stakeholder themes were synthesised with scoping review findings to produce six discussion themes, which were used to guide groups in the succeeding co-design study. The third EBCD step of producing a film of service user themes was then replaced by the development of an audio 'trigger' podcast extracted from the parent audio recorded interviews and parent themes. The decision was taken to audio rather than video record these interviews in the interests of encouraging open and frank parent participation. Development of an audio podcast represented a novel adaptation of the EBCD method.

The final study to be presented in this thesis- the co-design study broadly followed the remaining schedule of feedback and co-design events stipulated in the EBCD framework. The next four EBCD steps (refer Figure 7) were condensed into three co-design events to ensure the project fell within PhD timescales. A parent only, followed by two joint parent and sleep practitioner co-design workshops were facilitated which incorporated the showing of the parent audio podcast to both sets of stakeholders. These events invited stakeholders to challenge, develop or confirm discussion themes with fresh insights to co-create a SHE tool and programme theory. Finally, the intention was to replace the celebration event (which involves communicating results to others and evidencing the value of the research), with the dissemination of research findings to stakeholders at the conclusion of this project. A

detailed description of the methods employed in the exploratory and co-design studies will be presented at start of chapters 4 and 5 respectively.

3.10. Conclusion

This chapter has explained the philosophical and methodological underpinnings of the exploratory and co-design studies. It has also explored qualitative design issues, data collection and analysis methods. The next chapter will report the methods and findings of the exploratory study.

CHAPTER 4: EXPLORATORY STUDY.

4.1. Introduction

This chapter describes the methods and findings of the second study, the exploratory study, which aimed to address the qualitative evidence gaps identified in the scoping review and explore the perceptions of stakeholders about their experiences of SHE. A detailed description of the methods employed in this study will be presented first. The study involved qualitative semi-structured interviews and focus group interviews with stakeholders: parents of children with DD with sleep problems and sleep practitioners (professionals who support families with children's sleep problems). Exploratory study findings were analysed using a thematic analysis approach (Braun & Clarke, 2006) (Table 12): phases one to four involved analysing a first round of parent interview and sleep practitioner focus group data as two separate data sets to produce emergent themes. These were shown at a second round of parent interviews and sleep practitioner focus group to pursue respondent validation and are presented next in this chapter as a thematic map. Phase five-defining and naming themes involved a further refinement of themes, which were developed following the second round of stakeholder interviews. A comprehensive discussion of these final themes follows, supported by data extracts from study participants. This chapter concludes with a summary of the themes presented and synthesis of data sets which then inform the subsequent co-design study.

4.2. Research methods

4.2.1. Research question.

The exploratory study was guided by three broad research questions (Table 13) emulating from review findings and researcher experience. Primarily, it was considered important to affirm whether the evidence on SHE content in relation to sleep assessment and SHE components translated into practice; therefore, the first question asked what current practice was relating to SHE and how this related to scoping review findings. The review also identified evaluative evidence gaps in the literature and paucity in research explaining theory underpinning SHE, therefore, question two explored stakeholder experiences of SHE and what advice worked well, or not so well. Finally, the review highlighted a limited evidence base around methods of delivery, therefore, the study also focused on exploring 'What suggestions are there regarding what SHE should look like and how it should be implemented?' It was envisaged that the answers to these broad questions would address identified evidence gaps and help to inform SHE tool and underpinning programme theory development.

Table 13 Exploratory study research questions.

1.What is the current practice in relation to SHE and how does this relate to scoping review findings?
2.What are the perceptions of stakeholders about their experiences of SHE?
3.What suggestions are there regarding what SHE should look like and how it should be implemented?

4.2.2. Multi-method data collection approach.

A multi-method approach to qualitative data collection which involved one-to one interviews and focus groups was deemed prudent based on the principles of

triangulation. According to Patton (2015), triangulation allows data to be tested for consistency and illuminates real-world inconsistencies which offer richer understandings of the relationship between the phenomenon of interest and chosen inquiry method. Hence, combining these techniques allowed for vulnerabilities associated with each singular method such as leading questions or biased responses.

4.2.3. Ethical standards.

Ethical approval was gained for this study from the Healthcare and Medical Sciences Academic Committee, Bangor University on 5th March 2014 (Appendix 6.1). A research passport (Honorary Researcher) was granted for three years by the local health board on the 31th March 2014. Ethical approval was gained from the Wales Research Ethics Committee (REC) 5 Bangor on the 23rd April 2014 advising minor amendments which were addressed prior to the full approval being given (Appendix 6.2). Of particular note was the amendment made to the parent information sheet (Appendix 7) which dealt with issues around participant confidentiality and the need to safeguard children. The original text was shorter and advised participants that anything which caused concern for a child's safety would be shared outside the research team. The REC approval process ensured more detail was added to this section. This made the issue much clearer to research participants and specified that the local safeguarding team would be informed of any safety concerns for a child or signs of abuse.

Ethical approval was then granted by the local health board Research and Development internal review panel on the 12th May 2014 (Appendix 6). Substantial amendments regarding two separate supervisory team changes were approved by

Wales Research Ethics Committee 5 Bangor on the 10th July 2014 and 8th August 2014.

In March 2013 this study was also registered as a National Institute for Social Care and Health Research NISCHR (now Health and Care Research Wales) portfolio study (Sleep Hygiene Education and children with Developmental Disorders (SHEDD) 13841). Adoption of this study onto the portfolio entitled the researcher to apply for NISCHR CRC Research Network support which was utilised during the exploratory study whereby NISCHR CRC staff co-facilitated sleep practitioner focus groups.

4.2.4. Parent participant sampling procedure.

The purposeful sampling strategy of chain sampling (Patton, 2015) was adopted to recruit the parent sample which firstly leads the researcher to make contact with well-situated people, who are likely to be aware of or in frequent contact with prospective participants. Well-situated people are then asked to identify cases which may match the study's inclusion criteria and typically the names of prospective participants 'snowball'. Support for this research project was acquired at its inception, through building working relationships with the following voluntary organisations which support children with DD: Handsel Project, Down's Syndrome Wales, Cerebra and Children in Wales. Organisational representatives were approached as well-situated people for names of prospective parents.

A purposive sample of up to 10 parents was aspired to in the exploratory study and a sample of nine parents was achieved (Table 14). Each parent was assigned a pseudonym to maintain anonymity. Parent participants Kim and Sarah were recruited

and introduced by Down's Syndrome Wales, Corrine, Maureen and Sally by the Handsel Project and Stella by Children in Wales. Winnie was recruited via an opportunistic sampling approach (Patton, 2015) which takes advantage of opportunities for recruitment as they arise during the data collection process and was introduced by Stella during her home-based interview. As these strategies were only successful in recruiting six participants, an additional more pragmatic version of chain sampling was employed. This involved gaining permission to attend a local voluntary organisation climbing wall group for children with DD where potential parent participants attended and participants: Georgia and Gloria were recruited in this manner.

Table 14 Exploratory study parent participant purposive sample.

	Pseudonym	Peer supporter used	Demographics	Recruited via.	Presenting sleep problems	Participation interview round 1 and/or 2.
1	Kim	Grandmother	Mother to 9 year old boy twins, one with Down's syndrome.	Down 's Syndrome Wales	Unusual episodes interfering with sleep	1
2	Sarah	Husband Friend	Mother to 5 year old girl twins, one with Down's syndrome	Down 's Syndrome Wales	Problems falling, staying asleep.	1 & 2
3	Stella	No	Mother to a 10 year old boy with ASD phenotype*	Children in Wales	Problems staying asleep.	1
4	Corinne	No	Mother to an 8 year old boy with developmental delay	Handsel Project	Problems falling asleep.	1 & 2
5	Maureen	No	Mother to an 18 year old girl with Asperger syndrome, ADHD, Dyslexia and Bipolar disorder	Handsel Project	Problems falling, staying asleep. Early waking and sleeping in the day.	1
6	Winnie	Friend (Stella)	Mother to 15 year old twin boys, one with Asperger syndrome and Developmental Coordination Disorder	Introduced by Stella.	Problems falling asleep and rising in time for school.	1
7	Georgia	No	Mother to two boys aged 4 and 9 both being assessed for ASD.	Local climbing wall group	Problems falling asleep.	2
8	Gloria	None	Mother to a 9 year old boy with learning disabilities and a girl aged 10.	Local climbing wall group	Problems falling asleep.	2
9	Sally	None	Mother to a 6 year old boy with Asperger syndrome and a girl aged 4.	Handsel Project	Problems falling asleep.	2 (telephone)

All prospective parent participants were sent or emailed an appropriate participant information sheet (Appendix 7), contact form (Appendix 8) and consent form

(Appendix 9). All participant documentation was translated into the Welsh language by Bangor University's Translation service and parents were offered the choice of conducting interviews in Welsh with a Welsh speaking research colleague. Making appropriate provision for research participants to give informed consent and participate in their language of preference are best practice principles (WAG, 2009). Before the interview commenced, parents were asked to read the participant information sheet again and any concerns were discussed with the researcher prior to signing the consent form.

4.2.5. Inclusion/ exclusion criteria for parents.

To be eligible to participate in this study, parents needed to be aged over 18 years and have a child with DD aged 3-18 years with experience of sleep problems. Parents required some experience of SHE as an intervention to treat their child's sleep problems.

Parents younger than 18 years old were not eligible to take part in this study. Parents of young people who were 19 years and older or who did not have any experience of SHE were excluded. Parents of typically developing children with sleep problems were also not eligible to take part in this study. No restrictions were placed on the gender of parents

4.2.6. Parent characteristics.

Parents lived in England and Wales, were all first language English and non-Welsh speakers. Participants' children had a range of DD including Down's syndrome, Attention Deficit Hyperactivity Disorder (ADHD) and Autistic Spectrum Disorder (ASD) (Table 14). Eight children were primary school aged and two were teenagers.

All parents were mothers; fathers were not excluded from the study but none volunteered to take part. Seven mothers were married or in a relationship and had support to raise their children; however Stella and Maureen were single parents. Georgia had two children with DD, three parents had twins, one child of which had DD, and the other five parents had a single child with DD or two children one of which had DD. Six parents (Sarah, Stella, Corrine, Georgia, Gloria and Sally) reported that their children had behavioural problems associated with sleep. Maureen and Winnie reported problems with the timing of when their children slept rather than behavioural issues. Lastly, Kim was unique in reporting unusual episodes that interfered with sleep which involved her son walking about the house in a semi-conscious state during the night (Table 15).

4.2.7 Data collection. Parent interviews.

Data were collected from parents using semi-structured, one- to-one interviews that were carried out either in the parent's home or by telephone. A discussion area guide was developed from debate with the supervisory team about scoping review findings and the researcher's clinical experience (Table 15). Discussion areas were also based upon the three broad research questions previously detailed in Table 13.

Table 15 Parent interview discussion area guide.

1. **Description of child's sleep problem and individual circumstances.** When the sleep problem started, its severity and duration. Ways of coping and managing. Support mechanisms in place.
2. **Accessing professional support**. Referral pathways, waiting times, accessing services and attending appointments.
3. **Description of sleep assessment process**. When assessed and by whom. One to one interview with professional, history taking, questionnaire, sleep diaries. Preparation and support to complete sleep diaries.
4. **Description of SHE (general sleep advice) delivered**. Advice given to adapt sleep environment, bedtime routines and managing child's behaviour at sleep times. Other interventions offered-behavioural or pharmacological
5. **SHE (general sleep advice) components family believed were effective.** Which advice worked well /not so well to improve sleep problems?
6. **Description of how SHE (general sleep advice) delivered and how family was supported by professional.** Length and number of home visits, flexibility of appointments, group workshop, clinic appointments. Telephone/text message support. Attitude and approach of professional.
7. **SHE development.** What should good sleep support or SHE look like?

Six parents: Kim, Sarah, Stella, Corrine, Maureen and Winnie took part in a first round of interviews between the 23rd May and the 7th July 2014, which took place in the parent's homes at a convenient time and lasted between 60-90 minutes. Kim, Sarah and Winnie chose to have a friend or relative to support them who was present throughout the interview process.

4.2.8 Digital recording, video recording and transcription.

Parent interviews were digitally recorded to facilitate transcription of interviews, coding of interview transcripts and appropriate selection of anonymised verbatim for quotation within the research write up. Recordings were transcribed verbatim by the researcher and administrative support from Bangor University was also commissioned from the project expenses budget to assist with this process.

4.2.9 Safeguarding children and young people.

The researcher had an enhanced Disclosure and Barring Service (DBS) check, Level 2 Child Protection and Good Clinical Practice training. No safeguarding concerns were flagged up during the course of this study however a commitment to following local safeguarding policies and processes was made if the need had arisen. An identification badge was also worn at all times by the researcher when working outside of the university. Adopting such measures to ensure the safety and well-being of research participants links with the principles of ethical research practice as described by the Research Governance Framework for Health and Social Care in Wales (WAG, 2009).

4.2.10 Data protection.

All personal information was stored in accordance with Bangor University's Data Protection Policy (2015). Transcripts of interviews were anonymised after collection, participants were given pseudonyms and potential identifiers were removed. All paper-based information which potentially identified a participant was stored in a locked filing cabinet at the School of Healthcare Sciences. Research participants were informed that they could withdraw from the study at any point and give the research team permission to use material collected or withdraw and ask for material not to be used. All transcribed data is stored in a locked filing cabinet at the School of Healthcare Sciences for the length of time required by Bangor University policy which is at present 10 years.

4.2.11 Sleep team and sleep practitioner participant sampling procedure.

A chain sampling strategy (Patton, 2015) was employed to recruit a convenience sample of sleep practitioners to take part in the exploratory study. This involved approaching research colleagues at Bangor University, the local health board and partner voluntary organisations: Handsel Project, Down's Syndrome Wales, Cerebra, and Children in Wales for details of sleep teams which supported children with DD. This process identified 11 different sleep teams who were then approached for names of sleep practitioners who may be interested in taking part. A total sample of 16 sleep practitioners was recruited (Table 16).

Table 16 Exploratory study sleep practitioner sample.

No	Participant's professional role.	Participant Code.	Team code	Recruited via:	Focus group 1 and/or 2, Validation phone-call, no participation.
1. 2.	Family Support Officer. Clinical Psychologist.	SFSO. SCP.	} Social care team 1-ST1 (Sleep specific consultation team)	Handsel Project	1&2 1&2
3. 4.	Learning Disability Nurse. Learning Disability Nurse	HLDN (1). HLDN (2).	} Health team 1-HT1 (Consultation team- holistic team including sleep) Local county 2	Local health board	1 & validation phone-call.(HLDN (1))
5. 6.	Specialist Health Visitor. Specialist Health Visitor.	HSHV(1) HSHV(2)	} Health team 2-HT2 (Consultation team- holistic team including sleep) Out of county	Children in Wales	1 Validation phone-call.(HSHV(2))
7.	Sleep Practitioner.	VSP.	Voluntary organisation team 1-VS1(Sleep specific consultation team) Cross county	Cerebra	1&2
8.	Short Break Manager.	VSBM.	Voluntary organisation team 2-VS2 (Signposting team) Local county 2.	Children In Wales	1
9.	Development Officer.	VDO.	Voluntary organisation team 3-VS3 (Signposting team) Cross county	Down's Syndrome Wales	1

10.	Family Officer.	VFO.	Voluntary organisation team 4-VS4 (Signposting team)Cross county	Children in Wales	1&2
11.	Development Officer	VFW	Voluntary organisation team 5-VS5 (Signposting team) Cross county	Bangor University	2
12.	Community Paediatrician	HCP	Health team 3 –HT3 (Consultation team) Local county 3 &4	Local health board	No participation.
13.	Development Officer and Sleep Practitioner	VSS	Voluntary organisation team 6 VS6 (Signposting team) Cross county	Children in Wales	No participation.
14.	Development Officer and Sleep Practitioner	VSS(2)	Voluntary organisation team 6 VS6 (Signposting team) Cross county	Children in Wales	No participation.
15.	(CAMHS) Practitioner	HMP(1)	Health team 4- HT4 (Consultation team) Local county 3 & 4	Local health board	No participation.
16.	CAMHS Practitioner	HMP(2)	Health team 4- HT4 (Consultation team) Local county 3 & 4	Local health board	No participation.

Sleep teams represented in the sample were given a team code to protect confidentiality. Codes prefixed with the letter 'H' referred to local health board teams, 'S' to social care teams and 'V' to voluntary organisation sleep teams. Sleep practitioner participants were also given codes which began with the code assigned to the type of sleep team they represented (health, social care or voluntary organisation), followed by the initials of their professional role and a sequential number if more than one professional represented the same team. Participants: HLDN(1), HLDN(2), HCP, HMP and HMP(2) were recruited through the local health board and HSHV(1), HSHV(2), VSBM, VFO, VSS and VSS(2) were recruited through Children in Wales. SFSO and SCP were recruited through the Handsel Project, VSP through Cerebra, VDO through Down's Syndrome Wales and VFW through Bangor University. All sleep practitioner participants were sent or emailed a participant information sheet (Appendix 7), contact form (Appendix 8) and

consent form (Appendix 9). All participant documentation was translated into the Welsh language by Bangor University's Translation service.

4.2.12 Sleep team characteristics.

The sleep teams represented in the sample (Table 16) covered the six local counties in the area. HT1 represented a local county, HT3 and HT4 both represented two local counties and HT2 covered a county outside of the local area. ST1 represented a local county and VS1, VS2, VS3, VS4, VS5 and VS6 all operated across the six local counties.

Teams represented delivered a diverse range of sleep service provision. Six teams (ST1, HT1, HT2, VS1, HT3 and HT4) had a direct role assessing children's sleep problems and designing SHE interventions whereas voluntary sector teams (VS2, VS3, VS4 VS5 and VS6) gave ad-hoc SHE advice and worked in more awareness raising and signposting capacities. To clarify role diversity, represented sleep services were categorised into 'consultation teams' or 'signposting teams' in the analysis process. Furthermore, consultation teams either specialised solely in sleep (ST1 and VS1) or offered sleep advice as part of a broader range of health services (HT1 and HT2).

4.2.13. Inclusion/exclusion criteria for sleep practitioners.

Sleep practitioners met the inclusion criteria if their role involved supporting families of children aged 3-18 years with DD with sleep problems and use of SHE. No restrictions were set around the need to have professional qualifications; however sleep practitioners did need at least six months experience of supporting families with sleep.

Sleep practitioners who only supported families of typically developing children, older children aged 10-18 years or adults with learning disabilities were excluded. Sleep practitioners with less than six months sleep work experience and/or no experience of SHE were also excluded. No restrictions were placed on age or gender of sleep practitioner participants.

4.2.14. Sleep practitioner participant characteristics.

Sleep practitioners held posts ranging from Community Children's Learning Disability Nurses (n=2), Specialist Health Visitors (n=2) and Development Officers (n=4). All sleep practitioners were female; male participants were not excluded from the study, however, none came forward to take part. Only one participant: SFSO was Welsh speaking however, she chose to participate through the medium of English.

Some participants represented the same team (Table 16). It is important to clarify that participant and team codes were used interchangeably throughout the analysis to reflect how participants often described current practice in terms of how their represented team operated. It is acknowledged however that focus group discussions were a reflection of the views of individual participating sleep practitioners rather than the official opinions of their representative teams.

4.2.15. Data collection. First sleep practitioner focus group.

Data were collected from sleep practitioners utilising a focus group that was carried out at a local research organisation venue on the 15th July 2014. A discussion area guide was developed through discussions with the supervisory team regarding the researcher's clinical experience and scoping review findings (Table 17). Discussion

areas were based upon the three broad research questions previously detailed in

Table 13.

Table 17 Sleep practitioner focus group discussion area guide

1.**Referral.** How children with DD and sleep problems are referred to sleep services. Sleep screening initiatives. Eligibility criteria.
2.**Waiting lists.** Numbers, length, priority given.
3.**Sleep assessment.** When done-prior to or following initial sleep intervention (including pharmacological). Type of assessments (sleep histories, open ended questionnaires, outcome measures), time commitments. Measuring outcomes. Who assesses/training needs of sleep practitioners?
4. **Sleep problem origin.** Identification of behavioural sleep problems. Referral pathways in place for sleep problems with other distinct causes.
5. **Interventions.** First- line sleep interventions offered by sleep services for behavioural sleep problems, time commitments. Who delivers/training needs of sleep practitioners? Description of SHE and components currently delivered to families of children with DD. Adaptations necessary for children with DD/tailoring intervention to meet individual need. SHE delivered in combination with other interventions (behavioural/pharmacological). The intentions and goals of SHE. What SHE aims to achieve for families.
6. **Intervention delivery.** Mode of SHE delivery and models of family support.
7. **SHE development.** What should SHE for children with DD look like?

Nine sleep practitioner participants from the sample were able to take part in the two

hour focus group. It was conducted in English as the researcher was non-Welsh

speaking; however participants were offered the choice of separate interviews

through the medium of Welsh with a Welsh speaking research colleague if they

preferred (none chose to take up this offer). After reading the participant information

leaflets sleep practitioners either returned signed consent forms by post/email or

consent was taken on the day of the focus group. Participants were given the

opportunity to discuss any questions they had regarding the research at the start of

the meeting.

A NISCHR research officer co-facilitated the group and organised the consent forms,

signing in sheets, house-keeping and kept a written record of proceedings. The group

was arranged using a Doodle planner system, whereby prospective dates and times based on researcher and venue availability were emailed to all sleep practitioners, who recorded and shared their availability. The most popular date and time was selected and the booking confirmed with the venue and all sleep practitioners.

4.2.16. Digital recording, video recording and transcription.

The sleep practitioner focus group was digitally recorded to facilitate transcription of the interview, coding of the interview transcript and appropriate selection of anonymised verbatim for quotation within the research write up. The focus group was also filmed using video recording equipment in order to distinguish conversation between participants and to aid data analysis if problems arose with respect to audible clarity of voices on the digital sound recorder. Recordings were transcribed verbatim by the researcher and administrative support at Bangor University.

4.2.17. Safeguarding children and young people.

Safeguarding issues were addressed in order to conduct the focus group as detailed earlier (section 4.2.9).

4.2.18. Data protection.

Data protection issues were addressed in order to conduct the focus group as detailed earlier (section 4.2.10).

4.2.19. Analysis.

Following data collection, an eight week period of transcription and preliminary data analysis ensued. Transcribed data were organised into parent and sleep practitioner

data sets and analysed separately by hand using the six phase thematic analysis process (Braun &Clarke, 2006) (Table 12)

The data within each set was actively considered and connections made based on prevalence and 'keyness' in relation to the research questions. Initial codes were developed to describe data which demonstrated a certain issue or perspective which was particularly poignant or repeated by different participants. Commonalities between codes were then linked to formulate potential themes which aimed to explain the data and were systematically reviewed, refined and defined. This process is also described by Braun and Clarke (2006) as "searching *across* a data set- be that a number of interviews, or focus groups or a range of texts-to find repeated patterns of meaning" (p.15). Themes were developed inductively and data driven rather than fitting them into a predetermined coding frame because the data had been collected specifically for the study and participants shared a wealth of information that did not always link directly to the interview questions.

4.2.20. Participant validation of findings.

Preliminary parent themes were then validated in an additional round of interviews with five parents: Sarah, Corrine, Georgia, Gloria and Sally between the 1st and 30th September 2014. These interviews were organised and conducted in the same way as the first round of interviews but the original discussion area guide (Table 15) was replaced with the preliminary parent themes. Parent participants: Kim, Stella, Maureen and Winnie were unable to commit to a second interview. Sally's interview was performed by telephone because she lived a long distance away and budgetary constraints did not allow travel for a home interview.

Preliminary sleep practitioner themes were also presented for validation at a second focus group on the 5th September 2014 which was attended by five sleep practitioners: SFSO, SCP, VSP, VFO and VFW, which was organised and conducted in the same way and in the same venue as the first group but the original discussion area guide was discarded. The remaining 11 members of the sleep practitioner sample were unable to make the date and time of the second focus group. Some validation questions pertaining to sleep teams HT1 and HT2, remained unanswered following the second focus group because sleep practitioner representatives were unable to attend. Therefore, validation phone-calls with HLDN(1) and HSHV(2) were made afterwards to answer questions. This process of 'member checking' where participants responses are checked against a researcher's conclusions demonstrates rigour in qualitative research according to Burnard et al. (2008). It also shows how participants were actively involved in testing and developing constructions as called for in participative inquiry (Guba &Lincoln, 1989).

Data from the second round of parent interviews and second sleep practitioner focus group were transcribed and a second level thematic analysis was performed which produced a set of refined themes (presented in the following exploratory study findings chapter). This allowed for triangulation between stakeholder perspectives and drove the process of co-construction of a SHE tool and programme theory leading the researcher into the following co-design study. Melding stakeholder perspectives to co-create ideas for positive change also demonstrates a commitment to participative inquiry (Guba & Lincoln, 1989).

4.3. Overview of Preliminary Themes

From the first round of stakeholder interviews (prior to member checking), five parent themes and three sleep practitioner themes were developed which incorporated various sub-themes (Figure 8). The first parent theme: 'Living with sleep problems' represented parents' experiences of managing children's sleep problems and the impact they had on the child and family. 'Asking for help' encapsulated experiences of seeking sleep help and showed how finding sleep services and being taken seriously by generic practitioners (such as Health Visitors or GPs) was a common challenge. 'Understanding the sleep problem' encapsulated parents' comprehension of what caused children's sleep problems and how sleep practitioners had attempted to assess them. 'Getting help' represented parents' experiences of receiving professional support in the form of medication and sleep advice with and without professional guidance. The final parent theme 'Describing good sleep support' represented views on how SHE should be delivered.

The first sleep practitioner theme 'Sleep service accessibility' represented sleep practitioners' perceptions of how accessible sleep teams were and challenges around service awareness, referral systems and service waiting times. 'Service inconsistencies' described a significant variation across sleep teams represented in terms of sleep assessment and treatment offered to parents. The final sleep practitioner theme 'Sleep service improvements' encapsulated views on enhancing services and described the need to standardise services, better promote services and explore the possibility of parents taking a more active role in supporting other parents with sleep problems.

Figure 8 Map of preliminary parent and sleep practitioner themes
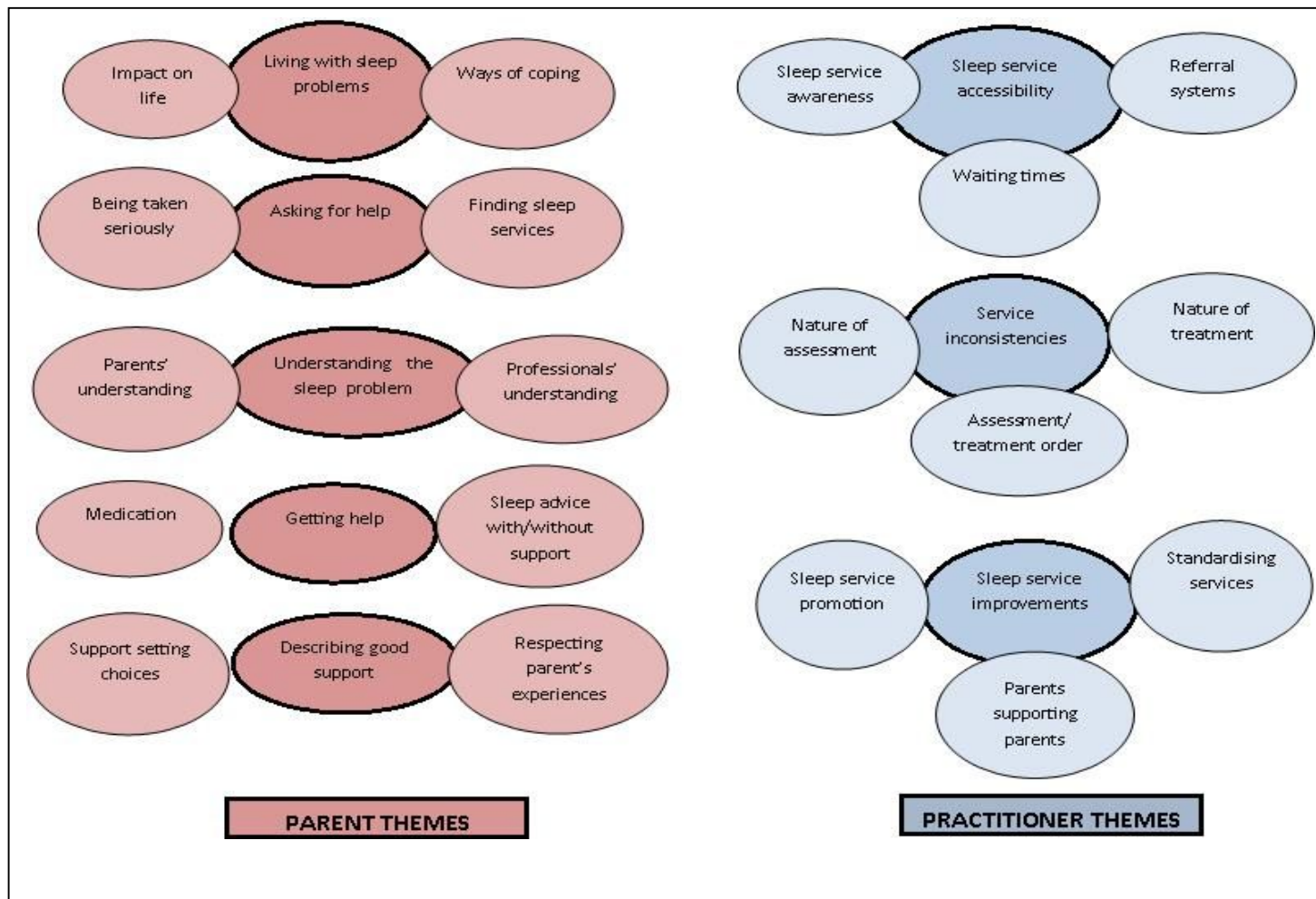
4.4. Overview of Refined Themes

Phase five of Braun and Clarke's (2006) thematic analysis –defining and naming themes resulted in a refined set of themes developed from both rounds of stakeholder interviews (Figures 12 and 13). Four parent themes and two sleep practitioner themes which incorporated various sub-themes were developed following the process of respondent validation in the second round of parent interviews and second focus group. Parent theme '"*Well this is how it is"*; beliefs about sleep problems' describes how parents believe sleep problems affect multiple areas of family life (developed from preliminary sub-theme 'impact on life'). This theme builds a more detailed picture of how parents describe the challenges sleep problems present and how they often resign themselves to the status quo. The theme '"*The biggest problem is knowing where to get the help"*; getting professional help' is derived from the preliminary theme 'Asking for help' but explores in more detail parents' accounts of accessing sleep support, challenges around parent/professional responsibility for sleep problems and convincing generic practitioners (such as Health Visitors and GPs) to take parents' sleep concerns seriously. The theme 'Ways of knowing about sleep' builds on the preliminary sub-theme' Ways of coping'. It describes the various coping mechanisms parents employ and their reported effectiveness in improving children's sleep. 'Ways of knowing about sleep' is also derived from the preliminary theme 'Understanding the sleep problem' and encapsulates parents' understanding of the causes of children's sleep problems and how sleep practitioners have attempted to assess them. Finally, the theme '"*I'd love Supernanny just to come and spend some time with us"*; visions of sleep support' draws on 'Getting help' and 'Describing good support' preliminary themes and

summarises parents' views on the use of sleep medication, the importance of sleep advice that has been tailored to individual need and supportive implementation settings.

The first sleep practitioner theme: 'Sleep service accessibility' builds on the original preliminary 'Sleep service accessibility' theme. It represents sleep practitioners' perceptions of parents' expectations of sleep help and public awareness of the support that sleep consultation teams provide. The 'Sleep service accessibility' theme also combines the preliminary sub-themes of 'Referral systems' and 'Waiting times' into the refined sub-theme of '"*Some families seem to fall through the net*"; negotiating referral systems'. It highlights a myriad of referral criteria stipulated by sleep services which parents have to negotiate to access specialist support and also different periods of waiting times. The second refined theme of 'Inconsistent sleep support' draws on preliminary themes 'Service inconsistencies' and 'Sleep service improvements'. It describes existing (and suggested) approaches to the assessment and treatment of sleep problems across sleep consultation teams.

Figure 9 Parent refined themes.

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Figure 10 Sleep practitioner refined themes.
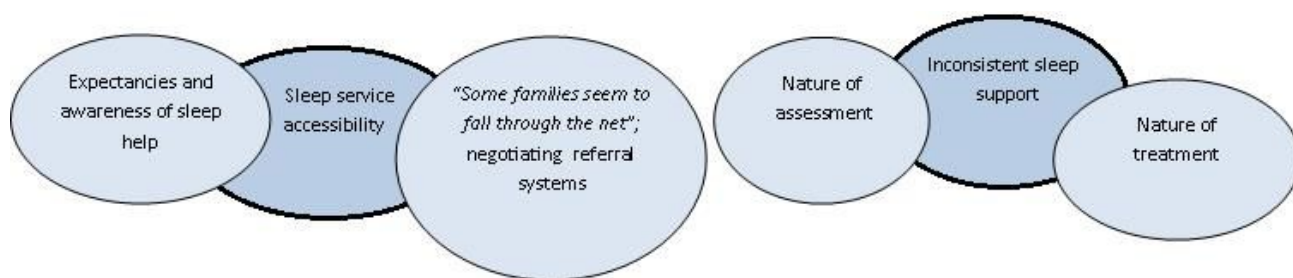
4.5. Presentation of Parent Refined Themes

In the following narratives direct quotes from participants are used to evidence findings and the symbol (…) denotes extraneous dialogue which has been removed from the extract.

Each of the four parent refined themes displayed in Figure 9 will now be presented.

4.6. Theme One- '*Well this is how it is'; beliefs about sleep problems.*'

Theme one '"*Well this is how it is";* beliefs about sleep problems.' (Figure 9) represents parents' experiences of living with chronic and severe sleep problems, and the daily and wider impact of managing them. Sub-theme- '"*It was a battle";* the challenges of sleep problems' focuses on the constant struggle parents face when coping with children's sleep problems, usually on a nightly basis over a period of many years. Sub-theme 'Resignation and acceptance' represents parents' acceptance of children's sleep problems and curtailment of their hopes that sleep will improve. Sub-theme '"*The sleep problems compound everything else";* broader impacts' encapsulates parents' perceptions of the wider impact of severe and persistent sleep problems on children's development. Each sub-theme will now be presented.

4.6.1. Sub-theme one- '"It was a battle"; the challenges of sleep problems.'

All parents articulated detailed accounts of their children's sleep difficulties but the majority of parents (n=7, Winnie, Kim, Stella, Sarah, Sally, Georgia and Maureen) reported particularly severe and enduring problems. When these parents described the nature of children's sleep problems, a sense of relentlessness was evident. Dialogue revealed significant difficulties with going to sleep and staying asleep every night over the majority of their son or daughter's childhood years. Parent perceptions of relentlessness were illustrated by Winnie whose 15 year old son was diagnosed with Asperger syndrome and regularly struggled to fall asleep until the early hours of the morning. Winnie's narrative indicated how she considered this late settling hour to be unacceptable and highlighted how her son seldom slept continuously all night. A sense of concern about her son's inability to sleep was clear from her words:

> *"The rarity is the fact he will go to bed at a reasonable hour and actually sleep the whole night through, which probably only happens twice a year, if that.(..)The rest of the nights it's the same thing."*
>
> *(*Winnie.)

Kim's story of managing her nine year old son's sleep problems on a nightly basis also demonstrated a sense of unremitting struggle and challenge. Kim was mother to nine year old boy twins, one of whom had Down's syndrome and problems with repeated night awakenings. These awakenings were further complicated by unusual behaviours and sleep walking which increased Kim's concerns for her son's safety. Kim's words illustrate how hard it was to manage such sleep problems on a regular basis:

"It has been; it was..it went..it's been really bad, it was every night"

(Kim)

A sense of continuous battle and deep concern about her child's sleep problems was also expressed by Stella. Stella was a single parent with a ten year old son who was diagnosed with Autistic Spectrum Disorder phenotype (mild version of ASD symptoms). Stella described how her son's sleep was disrupted on consecutive nights and how he rarely slept continuously. She described how challenging this was for a single parent to manage and how at one point, the situation became untenable:

"As well as not sleeping through. So it was all getting a bit ... and I was regularly doing sort of seventy two hour stints with no sleep (..) and I'm a lone parent (...) and that's just not ... sustainable."

(Stella)

The specific effect of enduring sleep problems on siblings and the inherent difficulties this can bring were expressed by Winnie, Kim and Sarah in particular who similarly had twins, one of which had DD and sleep problems. These parents expressed how the often close twin relationship had been affected by the sleep problems and how the typically developing twin's sleep was compromised. Sarah communicated how it was challenging to manage these dynamics and described the process as a constant struggle. Sarah's daughter was five years old, had Down's syndrome and problems settling to sleep independently:

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> *"She was quite distraught, you couldn't leave her, she'd wake her twin (..)it was a battle"*

> (Sarah.)

The significant sense of challenge that living with severe and persistent sleep problems had on parents and other family members was also described by Sally. Sally described how her son's sleep problems had affected the physical safety of his sibling and caused her and her partner significant stress. Sally's son was six, diagnosed with Asperger syndrome and presented with challenging behaviour at bedtime. Sally explained how her son would commonly lash out at his younger sister and at one point the situation became unbearable where Sally temporarily moved her daughter out of the family home to ensure her safety. She described this episode as incredibly challenging and that it had impacted upon her and her husband's mental well-being:

> *"When it peaked I just had to leave with [..] and I just went away for a few days because it just wasn't fair for her.(..) and when he didn't have his sister to take it out on then at least it was safer for her (..) and so he could ignore it a bit more(..) it's the only way we could cope.(..) because it was driving us absolutely crazy."*

> *(*Sally)

Sarah, Stella, Georgia and Maureen also described how living with children's sleep problems influenced their own mental health and ability to function on a daily basis. Georgia made very explicit links between her own sleep deprivation and clinical

depression which she disclosed she was taking medication for. This is demonstrated in the following extract which also reveals further stressors in her marital relationship and sense of guilt around devoting excessive time to her son's sleep problems and neglecting her marriage:

> *"I'm tired. Yeah, it's tiring (..)I've got depression (..) as has my husband; my husband's depression is severe.(..)Whereas I'm just ... you know, I'm fine, I'm on ... a level with antidepressants, and yes we cope (..) it's shattering at times(..) You know, with ... with everything. I'm supposed to be giving more time to my husband."*

> (Georgia)

Maureen communicated how all-encompassing the psychological effects were of living with children's sleep problems. Maureen was mother to an 18 year old girl with Asperger syndrome, ADHD, Dyslexia and Bipolar disorder. For many years Maureen had experienced severe difficulties with her daughter's sleep pattern which meant her daughter would sometimes not sleep for 24 to 36 hours consecutively. Maureen explained with a weariness of tone, how this had profoundly affected multiple areas of her life and how distressing her experience had been:

> *"People are starting to understand the impact of sleep deprivation because it almost like a form of torture, (short laugh). Because you will just get off to sleep and then you think, especially when she was younger, think oh she's up again. You know and it's exhausting"*

> (Maureen)

4.6.2. Sub-theme two- 'Resignation and acceptance.'

When expressing beliefs about children's sleep problems a feeling of resignation was described by the majority of parents (n=6, Kim, Gloria, Sarah, Corrine, Stella and Winnie.) Their descriptions of sleep problems indicated how they had come to view them as normal occurrences for their family and had started to accept them as part of everyday life. Inherent in the tone of resignation seemed to be a lowered expectation that sleep could improve for their children. A sense of resignation is firstly demonstrated by Kim who expressed how she felt about improving her son's sleep and tackling his disturbed night awakenings. Kim gave a frank account of how she felt a sense of acquiescence toward this sleep problem and could not foresee how things could change:

> *'To be honest, it was always something I thought 'Well this is how it is' "*

> (Kim)

Both Gloria and Sarah made very similar comments when conveying how they felt about their children's sleep problems. Gloria was mother to a nine year old boy with learning disabilities who refused to sleep in his own bedroom. Gloria recounted nightly battles to persuade her son to sleep in his room and often allowed him to fall asleep downstairs which meant she had to carry him to bed. Both parents indicated a sense of needing to accept the status quo and carry on coping with their situations. Their words also conveyed a feeling of despondency and belief that their children' s sleep problems appeared so entrenched that it was unlikely they would improve:

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"Just think oh, you know ... just get on with it."

(Gloria)

"So I just thought ... 'You've got to put up with it'

(Sarah)

Corrine further reflected how she felt about her son's sleep problems and believed she had begun to 'normalise' them. Corrine's son was eight years old and was diagnosed with developmental delay. Corrine described how she had become accustomed to the everyday nature of her son's refusal and protestations at bedtime. Her narrative conveyed how her son's sleep difficulties had become habitual and part of the fabric of family life. In addition, she described how she believed sleep problems were an inherent part of her son's character which was something she had begun to accept:

> *"Because you accept its normal, you sort of think it's just part of them."* *(*Corrine*)*

In resigning herself to her son's sleep problems, parent participant Stella explained how she had started to adjust and limit her expectations that his sleep could improve. Stella expressed how she would be content with her son just remaining settled in his bedroom at night and she would consider an increase in hours slept to be a bonus. Stella's narrative seemed to indicate how any improvement in sleep would be welcome but she lacked hope that this could be achieved:

> *"If he's not wetting the bed four times and he's not sleep walking, and I don't have to deal with a major meltdown at two am…that's a result (laughs)…I'm not looking for perfection..(..) just liveable"*

> (Stella)

In her acceptance of her son's sleep problems, Winnie also appeared to feel pessimistic about the possibilities that sleep could improve. Winnie explained how she believed her son could not help having sleep problems and they were part of his personality. Winnie's words indicated how she felt it would be difficult to understand why her son struggled to get to sleep and stay asleep and it would be best to just accept the status-quo:

> *"He obviously can't help it……because I.. I don't think he would do it as a choice..: …personally, I don't think anybody would choose to be awake till silly o'clock in the morning doing silly things"* (Winnie)

Gloria also spoke about how feelings of resignation could continue after receiving sleep advice and support from sleep practitioners. Gloria explained how despite the best efforts of sleep practitioners, her son was resistant to change and continued to refuse to sleep in his own bedroom. Over time, her son had developed a preference for sleeping in the master bedroom. Gloria accepted her son's right to choose and decided to build an extension and new master bedroom for her and husband to use. Gloria concluded that there was no possibility of persuading her son to sleep in his own bedroom and it was sensible to resign herself to his preferences:

> *"I have to say I think they were as helpful as they… as they could be. Because at the end of the day I can't change what's in his mind…. So they've got no chance".*

<div align="right">(Gloria).</div>

4.6.3. Sub-theme three – '"The sleep problems compound everything else"; broader impacts.'

When describing their beliefs about children's sleep problems, Maureen, Corrine, Winnie, Sally, Sarah and Stella expressed concern about impacts on child development. Parents explained how they felt enduring sleep problems had impacted specifically on their child's education, sociability and behaviour in the day and how these 'knock-on' effects had made caring for their children significantly more challenging. Maureen described how her daughter's 18 year struggle with irregular sleep patterns had affected other developmental areas. Her daughter would not sleep for up to 36 hours or sleep all day and be awake all night which affected her ability to attend school and maintain friendships. Maureen also reflected that her daughter's sleep problem worsened the other difficulties she had, associated with her multiple diagnoses:

> *"It really impacted on school and education when she was younger. It impacts on everything, the sleep problems compound everything else, the other difficulties she has. She already has difficulties with social integration and social relationships because of the autism. But obviously this makes it worse because she's asleep when other people are awake "*

<div align="right">(Maureen)</div>

Corrine also believed that her son's sleep problems had negatively affected his education. Corrine's narrative links her son's sleep deprivation with waking up feeling un-rested and grumpy which in turn affected his behaviour in the school day and ability to concentrate. Her reflection on the connection between her son's sleep problems, behaviour and education concerns is demonstrated in the following data extract:

> *"His behaviour, when he .. when he wasn't having a good chunk of sleep his behaviour would obviously be ..erm, be very poor and in school as well, his learning was being affected by him (..)not being able to get a really good chunk of sleep"*
>
> (Corrine)

Sally also made links between her son's lack of sleep, school performance and ability to behave and be sociable. She explained how following a poor night's sleep, her son would struggle to focus on his homework and would often be behaviourally challenging with peers. Sally's narrative provides a sense of how children's sleep problems can impact on their overall development:

> *"He was just so tired that he just cannot concentrate on the reading.(..)And then it impacts on his behaviour as well."*
>
> *(*Sally)

Winnie, Sarah and Stella all reported concerns about the impact of sleep problems on children's daytime behaviour. Winnie described how her son experienced behaviour

problems in addition to struggling with sleep and generic practitioners had attributed this behaviour to sleep deprivation:

> *"He had behavioural issues as well, and it was said that his behaviour was caused due to the lack of sleeping"*

<p style="text-align:right">(Winnie)</p>

Sarah also reported experiencing behavioural problems with her daughter on top of her sleep problems. Sarah recounted she believed her daughter became so tired in the day that she became irritable and struck out at others. She described how she felt compassion with her daughter and understood how a lack of sleep must make her feel:

> *"Sarah: She can get so frustrated, and a lot of it's tiredness and sort of like hitting out, sort of not meaning to."*

Stella described how coping with behaviour problems that resulted from sleep difficulties gave her added pressure as a parent. She explained how her son would often be very irritable and challenging when he was sleep deprived which added to his difficulties. In particular, she described how her son's communication difficulties were compounded by behaviour problems which further compromised his development:

> *"Like most children (..) any child that's over-tired ..becomes behaviourally very difficult. (..)Erm so on top of his communication*

issues and stuff like that you also had just 'I'm a too tired little

brat'."

(Stella)

4.6.4. Theme one conclusions. '"*Well this is how it is"*; beliefs about sleep problems'.

This theme paints a poignant picture of parent's perceptions of children's sleep problems. In the sub-theme '"*It was a battle",* the challenges of sleep problems.' a recurring sense of relentlessness is reported amongst the majority of parents when describing the constant challenge involved in managing chronic sleep problems. Within this, parents also describe how their own mental health and family relationships could be negatively affected. The second sub-theme demonstrates how most parents felt a sense of resignation towards children's sleep problems and lowered their expectations that sleep could improve. Lastly, the sub-theme-'"*The sleep problems compound everything else*"; broader impacts' identifies undesirable impacts of sleep problems on children's education, mood and behaviour. The analysis demonstrates parents' need to share experiences of their daily realities of living with sleep problems and has implications for sleep practitioners seeking to develop trusting relationships with parents at the start of any SHE intervention. In establishing an effective parent /sleep practitioner partnership which allows parents to move forward with sleep difficulties, it is therefore posited that sleep practitioners need to appreciate how parents experience and perceive children's sleep problems and believe they impact on children's development.

4.7. Theme two- Ways of Knowing about Sleep

Theme two 'Ways of knowing about sleep' (Figure 9) describes the different forms of knowledge parents have about sleep problems, SHE advice and various sleep management strategies. In the first sub-theme 'Understanding sleep problems' parent perceptions are explored for what they and sleep practitioners think causes children's sleep problems. Sub-theme two -'Forms of coping' depicts parents' views on intuitive coping mechanisms which mirror SHE practices and their effectiveness in addressing sleep problems. It also describes parent's experiences of trying out alternative strategies to SHE, to manage children's sleep problems. Each sub-theme will now be presented.

4.7.1. Sub-theme one- Understanding sleep problems.

Parents were keen to share their thoughts about what caused their children's sleep problems. Sally, Corrine, Sarah and Kim directly attributed their children's sleep problems to their disability diagnosis. For Sally and Corrine this understanding appeared to have a positive effect in terms of their attitude towards the sleep problems. For example, Sally's narrative communicated a clear sense of empowerment when she described her detailed understanding of how her son's Asperger diagnosis meant he found falling asleep distressing. Her words indicated how she felt in tune with her son's needs and able to empathise with his difficulties at bedtimes:

> *"I've read that Asperger children, people on the autistic spectrum*
>
> *have difficulties with sleep, because it's another sort of transition,*
>
> *and he struggles with transitions, with letting go, and he'll be out of*

control. So I do understand, and I've got more sympathy with it now

that I know that"

(Sally)

Corrine also understood her son's sleep problems to be caused by his developmental delay and seemed to find some comfort in this knowledge. Furthermore, she hypothesised that his problems with settling to sleep were triggered by external social factors such as past or future stimulating events. She felt he often struggled to relax on these occasions and in making this connection acknowledged that there were multiple causes to her son's sleep problems (i.e. they could not be attributed to his diagnosis in isolation):

*"He was just really unable to switch off. And this is part of [*Child's name*]'s erm ... sort of disability anyway* (..) *and it all depends on what he's been doing in the day,(..) or if he's going to be doing something exciting his mind can't switch off so...(..)that's when he becomes obviously challenging with ... with going to sleep, or being able to fall asleep."*

(Corrine)

Sarah described how she initially linked her daughter's sleep problem exclusively to her Down's syndrome diagnosis, but how over time had identified daily triggers and a more nuanced understanding. She learnt that her daughter thrived on routine and hypothesised that changes would result in increased settling difficulties. In making

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these conclusions she was acknowledging that her daughter's sleep problem had multiple causes which included her disability diagnosis, personality traits and daily routine practices:

> "Sarah: *"We thought it was literally to do with the (..) Down's syndrome (..) But we've worked out since as well , erm..she's a..very much a sticker for routine....If anything changes it throws her"*.

Kim also described how she rationalised her son's sleep problems by linking them to his disability diagnosis. Her son was diagnosed with Down's syndrome and Kim recounted a moment of realisation when discussing his sleep problems with others. It had been suggested that children with Down's syndrome had a high prevalence of sleep problems, which convinced Kim that her son's diagnosis was the primary cause of his night awakening difficulties. Kim's narrative conveyed how this new understanding brought a sense of acceptance and tolerance to her situation, which also arguably curtailed a need to develop a richer understanding of other possible triggers to her son's disturbed night awakenings:

> "*They said to me "How does he sleep?" and I said ' Oh, it's a bit of a nightmare' (..) and they said 'Oh, it's quite a common thing with children with Down's syndrome.(..) So I thought ' Oh, well that explains it then… I kind of took it as part and parcel.*"

> *(*Kim)

Instead of attributing sleep problems to a disability diagnosis, Maureen linked her daughter's difficulties to an allergy to polyester, the onset of puberty and her mental health condition. Through a process of trial and error, Maureen concluded that her daughter would sleep better if her contact with polyester based cuddly toys was limited at bedtime. Furthermore, she deduced that her daughter's sleep timing problems coincided with the onset of puberty and reflected that Bipolar Disorder may have impacted on her ability to sleep at socially appropriate times. Maureen had appeared to give considerable thought to the nature of her child's sleep problem, possibly owing to the severity and chronicity of its presentation. Her narrative communicated both a feeling of acceptance and sadness that her daughter's sleep problem was very complex and had multiple triggers:

> *"We found out she had an allergy to polyester so first of all I thought some of her sleep difficulties were cause she slept with loads of soft toys"*

> *"It wasn't till "…."she hit puberty that the being awake for 28, 24, 48 hours really started to kick in."*

> *"Because "…"has a , has a social phobia to be asleep during the day when everybody else is out and to be awake at night, can be quite calming for her, quite comforting. So there's a little bit about how much is this self-regulating? (..)I do think there's a, there's an element of a sleep disorder but one that is actually made worse by "…."need for, for not to socialise"*

> (Maureen)

Stella, Gloria, Sarah and Sally reported having their children's sleep problems assessed by sleep practitioners and mixed views as to how this had contributed to their understanding. Stella told how she had developed her own theory to explain her son's sleep problems and reasoned it was because he was stressed at school. At the time of the interview, her son was being home schooled and Stella reported some improvements in his sleep as a result. Being unhappy at school was described as the primary sleep problem cause which Stella had identified rather than any specific links being made to his disability diagnosis of ASD Phenotype:

> *"Well his big stressor was school (..) now keeping him relaxed*
>
> *enough to go to bed at bedtimes is within my control.."*

> (Stella)

Stella also described how a Paediatrician had evaluated her son's sleep using an actigraph, which resulted in a diagnosis of 'sleep maintenance disorder'. Stella understood this caused her son to have night waking difficulties but indicated she had not been fully informed about the nature of this sleep disorder. Furthermore, Stella's dialogue suggested some confusion around an understanding that her son had missed a developmental window of opportunity to learn to sleep, which seemed to unhelpfully reduce her expectations that his sleep could improve:

> *"Maintenance disorder, yes.(..) And that's because he's never learnt*
>
> *to sleep through .(..).. that's how it was described to me by the*
>
> *consultant.(..) There is a stage at which babies learn to sleep through*

...(..)... and he skipped that (..)I'm not pretending that I understand

quite how"

<div align="right">(Stella)</div>

Gloria had also developed an understanding of her son's sleep problem following a sleep practitioner's sleep assessment. Gloria described her involvement with a specialist sleep team which consisted of Learning Disability Nurses trained in sleep assessment and interventions. These sleep practitioners supported her to complete a sleep history questionnaire and gave written guidance on how to complete sleep diaries at home. Sleep practitioners concluded that her son refused to sleep in his own room because he preferred to sleep in her larger, master bedroom instead. This interpretation prompted Gloria to be sensitive to her son's preferred choice of sleeping environment and her narrative conveyed a sense of satisfaction with this new understanding:

"They said that I was looking at it from the wrong perspective in that

if I put myself into his shoes, he's already made…had the..of the

opinion that he didn't like where he was…(..)the little box room..He

didn't want that"

<div align="right">(Gloria)</div>

Both Sarah and Sally also described experiences of having their children's sleep problems formally assessed by sleep practitioners using sleep history questionnaires and sleep diaries. Sarah recounted a very positive and supportive experience of this process which also included an overnight sleep study at home to eliminate sleep

related breathing disorders prevalent in children with Down's syndrome. In contrast, Sally reported an experience of completing the sleep questionnaires and diaries independently of sleep practitioner support. However, at the conclusion of the sleep assessments neither parent reported receiving a clear interpretation of what was causing their child's sleep problem. Sarah explained how sleep practitioners concluded by reassuring her she was not alone in experiencing sleep problems and Sally reported how sleep practitioners appeared to proceed straight to sleep advice:

> *"It was just a ... because (*sleep service) *are involved obviously a lot with the sleep ...(..). I think they were used to it, and erm ... they were very confident, they said "This happens to a lot of children, you're not the only one" "*
>
> (Sarah)

> *"INV: What was their sort of final kind of ... analysis of the situation? Did they say (..) This is what's causing it'(..)?*
> Sally*: No they didn't, they just sort of said a generic idea of how to sort it"*
>
> (Sally)

4.7.2. Sub-theme two- 'Forms of coping'.

This sub-theme represents how parents adopted intuitive coping strategies and their perceptions of effectiveness in addressing sleep problems. Some coping mechanisms described mirrored SHE practices, although parents did not make the link or refer to their strategies as SHE methods. Parents also tried alternative forms of coping such as co-sleeping and some were aware of how such strategies were incompatible with

usual SHE advice. The strategies that were cognisant with SHE principles will firstly be represented. The majority of parents (n=7, Corrine, Maureen, Kim, Georgia, Sally, Stella and Sarah) described learning about some coping mechanisms intuitively. For example, the notion that the broad based SHE principle of creating a calming bedtime routine was common sense and based on usual parenting practice is demonstrated in the following extract from Corrine:

> *"It's just something you ... you're kind of in tune with knowing isn't*
> *it? (..)Mm, it's that ... you try and wind them down at night, have a*
> *story, go to bed."*

(Corrine)

Both Corrine and Maureen seemed to agree that certain types of intuitive coping methods however were ineffective in improving their children's sleep problems. Corrine stated her belief that trying to instigate and maintain a consistent sleep routine which involved bath-time and story-telling did not have the desired effect of improving her son's difficulties with falling asleep:

> *"We tried to train him into a better sleep pattern (...). sort of having a*
> *bath erm, having stories, and that ... and none ... none of it would*
> *actually make a ... make an impact."*

(Corrine)

Maureen made specific reference to instinctively trying out the strategies of no television in the bedroom, introducing black-out blinds and a healthy diet with her daughter which mirrored SHE principles. Maureen described how she had tried all

these measures to improve her daughter's sleep over the years. Her words indicated disappointment that her attempts had been unsuccessful in improving how her daughter slept and demonstrated the view that such forms of coping were inadequate:

> *"She's never had a television in her bedroom (..) she did have black out curtains (..)Strict diet, absolutely no colourings at all and when I mean strict I mean cooking everything from scratch, again very little effect."*

> *(*Maureen)

Kim, Georgia and Sally in comparison reported a view of mixed effectiveness when describing their experiences of trying out intuitive coping strategies that reflected SHE advice. Kim made reference to trying to physically tire her son before bedtime by encouraging plenty of fresh air and exercise (weather permitting). However, she reported that this was not always successful in helping to keep her son asleep throughout the night:

> *"What we would always do is, if the weather was nice, after school I'd take them to the park, let them run themselves ragged(..)in the hope that…he'd be that tired he would sleep through the night. But it doesn't always work"*

> (Kim)

Georgia described how she had always set a consistent bedtime routine for both her sons, which always started at the same time. She portrayed how she was strict and

consistent in applying these principles. However, in Georgia's view this coping strategy was only effective when her sons were younger and now had mixed effectiveness in helping them to fall asleep independently:

> *"But.. the routine, like I say, is fixed, and it's set out…and you know, that…that used to work, it works some days"*
>
> *(*Georgia)

Similarly, Sally expressed how a calming and consistent bedtime routine was helpful in some ways but was not fully effective in solving her child's sleep problems. Sally described how a bath and bedtime story did indeed have a soothing effect on her son's mood, but he always presented with challenging behaviour as soon as it was time to fall asleep. Her narrative indicated that she believed such coping mechanisms were necessary but inadequate in fully solving her son's sleep problems. Sally appeared to be arguing that additional advice and support was necessary to help her son learn how to fall asleep independently and without resistance:

> *"He has a bath…and he's all calm and nice and he's…then he gets his pyjamas on…and we read him a story (..) it's the moment we come to say goodnight that it all kicks off"*
>
> *(*Sally)

In contrast, Stella and Sarah described a view that some intuitive coping practices were effective in helping their children to sleep well. Stella explained how the

coping strategy of being very strict with the timing of bedtimes and putting her son to bed at the same time every night, including weekends and holidays was effective. The consequences of not enforcing this boundary had been experienced by Stella as deterioration in her son's daytime behaviour. Stella's narrative conveyed an understanding that her son responded to the regularity of set bedtimes which enabled him to enjoy some quality sleep:

> *"I was so ruthless about him having to go to bed at half past seven, because that period between then and midnight was when I could expect him to get some decent sleep (..). that was his window for like ... what I'd call 'a proper sleep'(..) If he didn't get that then behaviourally the next day was a nightmare."*
>
> (Stella)

Sarah explained how her daughter self-soothed to sleep with a drink of milk before bedtime. She described how this intuitive parenting practice had continued since her daughter was a baby and believed it continued to be effective in promoting sleep:

> *"She's always, even now she has a ... not a ... baby's bottle ... but she has erm ... a lidded cup ... of milk, and it's a good sized milk, and she doesn't settle without that.*
>
> (Sarah)

Winnie, Georgia, Sarah, Kim, Stella and Gloria also reported experimenting with alternative sleep management strategies to SHE. In the absence of or whilst awaiting professional sleep support, parents reported how they had experimented with various

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techniques to manage or improve their child's sleep. Some parents described how they were aware these strategies were incompatible with usual sleep advice from sleep practitioners, but others did not. Winnie reported being concerned that her 15 year old son was becoming sleep deprived because he did not fall asleep till after midnight and still needed to get up early for school in the week. Winnie explained that she had never received any professional advice and so decided to try letting her son sleep late at the weekends to maximise his sleep. Her dialogue demonstrated how she felt this was her only option and implied a lack of awareness that this practice was incompatible with usual SHE advice:

> *"He has to be gotten up in the mornings obviously (..)Yeah, he's got school. But then on the weekends he's sometimes not getting up till ten, eleven; it can be twelve o-clock (..)..he's got to get his sleep somewhere."*
>
> (Winnie)

Winnie also reported experimenting with electronic media to help manage the effects of her son's problems with settling at night. After going to bed in the evenings Winnie's son would often slip downstairs and steal food from the fridge for example. To handle this behaviour Winnie explained she felt her only option, was to allow him to watch television on his phone whilst in bed. This was largely effective in encouraging Winnie's son to remain in his bedroom and was a practice that appeared acceptable to her but she seemed unaware that this was also inconsistent with usual SHE advice:

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"He has his mobile phone, he watches telly in bed, because it's the

only way I can keep him in bed(..) until he eventually falls asleep."

(Winnie)

Georgia also reported resorting to electronic media to manage her children's sleep

problems and being unaware of alternative strategies. She explained how she felt her

eldest son needed some occupation to help him fall asleep and he preferred using his

IPad over reading a book. Georgia recognised this was contradictory to usual SHE

advice but described a belief that discontinuing its use would be very challenging.

Her narrative portrayed a sense that whilst Georgia felt this strategy was effective,

she would have liked to learn about alternatives:

"He's on the IPad, you know, and that is ... that is difficult because

they say blue light, take the ...... take screens away (..) that's easy

enough for them to say, but if his brain will not switch off what am I

supposed to do, leave him with nothing? You know, he won't read a

book."

(Georgia)

Georgia, Sarah and Kim reported experimenting with another strategy to help

manage their children's sleep problems: co-sleeping, which was also contradictory to

usual SHE advice. Georgia described how her youngest son struggled to fall asleep

alone and demanded her constant presence. She explained how she made a 'make-

shift' bed beside her own, to offer physical reassurance and closer sleeping

proximity to coax him to sleep. Gemma's words revealed how she felt helpless and

constrained by her situation, which contributed to her decision to move her son into her own bedroom to sleep. She portrayed how this strategy was effective in helping her child fall asleep but did not indicate an understanding that this was contradictory to usual professional sleep advice:

> *"He will not sleep in his bed, he cries and cries and cries (..)I'm like 'Mummy needs my own space', so I've put his bed on the floor so he's next to me and I hold his hand or I stroke his back and he will sleep there."*
>
> (Georgia)

Sarah's account of why she had experimented with a co-sleeping strategy with her daughter, also gave a sense of despair in not knowing what else to do. She recounted how prior to receiving some professional sleep support, she had resorted to lying beside her daughter at bedtime until she fell asleep. Sarah explained how she was aware this practice conflicted with usual SHE advice however co-sleeping helped her to cope with her situation in the absence of any other solutions she could think of:

> *"I know all the books tell you never do it, but out of desperation from. (..) Me I had to then, because she has got a normal sized bed, not a toddler bed. (..) had to literally lie next to her .(..) to just keep her settled."*
>
> *(*Sarah)

Kim also reported experimenting with co-sleeping but instead used this technique to help manage her son's disturbed night awakenings. Kim's son was able to settle

himself to sleep however on a regular basis woke in the night and wandered around the house. Despite trying to direct him back into his own bed, he would only return to sleep beside her or his Grandma in their beds. Kim's narrative implied that co-sleeping was not a preferred option and she would rather her son learn to settle independently back to sleep however, it was effective in managing her situation. Furthermore, Kim did not report any awareness that this practice was contrary to usual SHE advice:

> *"Then we guide him and put him back in bed (..)But more often than not.. that would continue till he gets in bed with somebody. Erm, we've found the easiest thing is that if he does that, either he gets in bed with his ..erm Grandma who lives with us..erm or he''ll get in bed with me.. Erm, and then he'll settle down for the night"*

(Kim)

The use of weighted blankets to help children sleep was another strategy Georgia and Stella experimented with. For her youngest son, Georgia had ordered a weighted blanket on-line to help him settle to sleep. Georgia suspected he had some sensory sensitivities linked to his possible ASD diagnosis that he was currently being assessed for and had researched the use of weighted blankets for such children who had sleep problems. (Whilst usual SHE advice recommends that sensory issues are considered, due to safety concerns, trained Occupational Therapists should dispense and monitor the use of weighted blankets.) Georgia hoped the blanket would help to replace her physical presence at bedtime and help her son fall asleep independently. Georgia was awaiting some professional sleep support and her narrative indicates a

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feeling of bitterness around having to take this initiative un-supported by generic practitioners:

> *"Our four year old loves ... cuddles, got sensory issues, but he's not diagnosed.(..)And I've ordered him a ... a weighted blanket (..):. so I'm just hoping that that will help, so he wasn't expecting the cuddles off me and ...... you know, that might make it a little bit easier(..)So I've just ... done that off my own ... you know, off my own steam basically (..) researched it and done it."*

> *(Georgia)*

Stella told a different story of how she had come to the decision to experiment with weighted blankets. Stella explained how health professionals previously involved had suggested the blankets, but she had made her own due to the high purchase cost. Stella described how she felt the blanket was effective in calming her son who moved around in an un-settled manner during sleep and was a technique she found acceptable. Her narrative gave a sense that in common with Georgia, she had taken some initiative independently of professional support in trialling a new technique to help improve her son's difficulties with staying asleep. Both Georgia and Stella seemed unaware of the potential safety risks involved in using weighted blankets unsupervised by trained Occupational Therapists. (Refer to reflexive section 6.6.2. for a description of how this was addressed):

> *"I think part of the reason the weighted blanket helps ...... is because it reduces the movements..(..) A friend err, we were (..) Hospital ... suggested it. Erm, so we thought well we'll try the weighted blanket,*

and actually ...... it has made a massive difference.(..) a friend helped

me make one."

<p align="center">(Stella).</p>

Lastly, Sally reported experimenting with reflexology to calm her son at bedtimes. Whilst usual SHE advice broadly recommends relaxation techniques to help aid sleep, like weighted blankets, reflexology is a specialist intervention outside the realms of typical sleep hygiene principles. However, Sally described how she found reflexology to be an acceptable and very effective method of soothing her son at bedtimes which she used on a regular basis when her son would allow the contact. Trying out this additional coping mechanism seemed to be another example of a parent needing to be very resourceful in managing their children's sleep problems in the absence of adequate professional sleep support:

> *"One thing I've started to do is give him reflexology ...... which is*
>
> *absolutely wonderful, that has a really big effect....But he has to be in*
>
> *the mood to let me do it.*

<p align="center">(Sally)</p>

4.7.3. Theme two conclusions. 'Ways of knowing about sleep'.

This theme describes the different forms of knowledge parents have about sleep problems and the strategies they have adopted to cope with them. Sub-theme 'Understanding sleep problems' shows how some parents attributed their children's sleep problems to their disability diagnosis and how this could have a positive or

negative effect. In some cases this knowledge gave comfort and understanding to parents which helped them cope. However, using the diagnosis as the sole explanation for a child's sleep problems appeared to lower the expectations of some parents that sleep could improve. Some parents also demonstrated a deeper layer of understanding as to why their child struggled with sleep and made links with day-to-day triggers which may contribute to sleep disturbances. Four parents reported having a sleep practitioner sleep assessment and mixed experiences were described around how this improved their understanding of their children's sleep problems.

The sub-theme 'Ways of coping' describes both intuitive coping strategies parents adopted that mirrored SHE advice and alternative strategies which were incompatible with usual advice. Most parents described how they were aware of advice around consistent and calming bedtime routines for example, although they did not explicitly link it to SHE. Parents often referred to such advice as commonly known good parenting practice and gave an overall sense that they were only partly effective and inadequate as a sole intervention for their children's sleep problems. Over half of parents experimented with other sleep management strategies, which were inconsistent with usual SHE advice such as co-sleeping or use of electronic media. These techniques were tried out in the absence of or whilst awaiting professional sleep support. Parents often felt they had to try 'something', were not always comfortable with their chosen strategy or were aware they were not ideal but felt they helped make their lives more manageable in the short term.

The 'Ways of coping' theme firstly demonstrates a need to improve parent and sleep practitioner understanding of children's sleep problems, to establish multiple causes and ensure eventual SHE advice or other interventions are appropriate. It also reveals

parents' views on intuitive SHE advice which they have already implemented. Their views are significant for sleep practitioners who are seeking to offer SHE advice and need to appreciate parents' existing experience of broad based strategies. Finally, this theme highlights the need for sleep service providers to be aware of how parents feel compelled to experiment with strategies which may contradict usual SHE advice, in the absence of professional support. How parents resort to these potentially un-helpful strategies which can reinforce or sometimes worsen sleep problems is a potential area of concern.

4.8. Theme Three- '"The Biggest Problem is Knowing Where to Get the Help"; Getting Professional Help.'

Theme three '"*The biggest problem is knowing where to get the help"; getting professional help' (Figure 9) explores parent's experiences of becoming involved with professionals with regards to children's sleep problems. The first sub-theme '"*Nobody's willing to take responsibility"*; challenges around professional responsibility' considers how parents looked to generic practitioners to take some responsibility in supporting them with children's sleep difficulties and the challenges they experienced in sourcing this help. The second sub-theme '"*They couldn't quite understand the severity of it"; validation of sleep concerns' encapsulates how parents experienced positive and negative experiences of convincing generic practitioners that sleep was a problem for their children. Each sub-theme will now be presented.

4.8.1. Sub-theme one- '"*Nobody's willing to take responsibility"*; challenges around professional responsibility.'

Winnie, Sally, Maureen and Georgia gave a clear sense that they believed generic practitioners needed to take some responsibility for their children's sleep problems and saw this as a professional issue. Winnie spoke about repeatedly approaching generic practitioners such as Child and Adolescent Mental Health Service (CAMHS) workers or Health Visitors for advice on where to go for sleep help or referrals to specialist sleep teams. She expressed her need to be very tenacious in sourcing appropriate sleep support and described a perception that generic practitioners were reluctant to be accountable for her son's sleep problems. Winnie explained how this left her feeling vulnerable and concerned that generic practitioners would apportion blame for her son's sleep problems and resultant effects. The whole experience of getting generic practitioners involved was summarised by Winnie as a continuous battle and one that she was still engaged with:

> "*Whenever I asked, It's the school who has to refer you, and then when you go to school "Oh it's your GP…and you go to your GP but it's down to the school because it's part of mental health it's done under school or…yeah". So you're just going round in a circle (..) and nobody's willing to take responsibility, until something goes wrong and then it's my fault(laugh). So it's just…it's just a constant fight."*

(Winnie)

Winne also described how she felt sourcing specialist sleep support was challenging. In being convinced that generic practitioners should take more responsibility for children's sleep problems, she concluded that there was a lack of awareness and knowledge of where to go for sleep help. For Winnie she felt this was a major problem experienced by families:

> *"I think the biggest problem is knowing where to go for the help. That is the biggest problem(..) Where do I go for the help?"*

> *(*Winnie)

Sally explained how she was convinced that professional sleep help was available for children with Asperger syndrome and understood sleep problems were a common issue for affected families. Her words gave a sense that her son's Asperger diagnosis and resultant sleep problems warranted specialist sleep advice which increased her determination to find appropriate support. She was eventually successful in finding specialist help, although recounted how generic practitioners were commonly poorly informed of sleep teams, which was an area of surprise and concern for Sally:

> *"There must be ways of dealing with it, because lots of people must be in the same position (..) So it made me seek help even more actually (..) that really struck me, nobody knew where to point us, you know, the school, not even a doctor knew."*

> (Sally)

Maureen also described how she felt generic practitioners should have taken more responsibility for her daughter's sleep problem. Her story supported the common thread running through Winnie and Sally's stories around having to continually ask for professional help which they found very challenging. Maureen had been unsuccessful in finding the sleep help she was looking for for her daughter. Her words conveyed a sense of needing to be very tenacious in her search and despondency that generic practitioners were unaware of who could offer sleep help:

> *"I've asked and asked and asked CAMHS and they say there's nothing available."*

> (Maureen*)*

Lastly, Georgia was also very clear how she expected more professional help and responsibility for her son's sleep difficulties. She described how she had had some involvement from a Health Visitor, but felt they were not able to offer the specialist sleep advice she needed for her children. Her words portrayed a sense that she thought her son's sleep problems were a professional issue, but she felt the availability of local services did not match up to her expectations which left her feeling very unsupported:

> *"The Health Visitor, there's only so much that they can do….So you are on your own, you don't know who to go to.: So you do feel like you're on your own."*

> (Georgia)

Kim, Corrine and Sarah conversely were less expectant of professional help and did not assume that help might be available. In describing her son's difficulties with disturbed night awakenings, Kim intimated how she accepted responsibility for this sleep problem. Her narrative suggested that she had never thought to ask for professional help with this type of sleep difficulty. This low expectancy of professional sleep help was coupled with a personal lack of knowledge of local services that might have been available to help her:

> *"I never questioned it(..) with professionals(..) I don't know what support there could be..erm in fact I didn't think there was any."*

> (Kim*)*

Corrine described how at the start of her journey she had not assumed that professional help with her son's sleep problems could be on offer. Her narrative also demonstrated a belief that generic practitioners already involved in her son's care were not aware of specialist sleep teams that could help. This was expressed as an unhelpful state of affairs for parents struggling to cope with sleep problems:

> *"You don't think that there is sort of support available. And if they're not aware then you're never ..(..).:... you're never going to know are you?"*

> (Corrine)

Lastly, Sarah who was ultimately successful in receiving specialist sleep help recounted how she had initial low expectations of professional sleep support and

believed that her daughter's sleep problems remained her responsibility. Her dialogue revealed a sense that generic practitioners originally involved in her care should have been more knowledgeable about available sleep services and taken a more active role in informing parents. Sarah also concluded that specialist sleep services themselves needed to take more responsibility for promoting their services:

> *"I didn't realise that you could have help…and nobody had really mentioned it before(..) even though we had Health Visitors, Paediatricians, every….nobody had said(..) You don't really realise there is something out there….it wasn't publicised."*
>
> *(Sarah)*

4.8.2. Sub-theme two- '"They couldn't quite understand the severity of it"; validation of sleep concerns.'

Sally, Maureen, Winnie and Georgia articulated a strong belief that generic practitioners should take some responsibility for children's sleep problems and also described how they had struggled to convince professionals to take their sleep concerns seriously. These parents expressed a sense of disappointment that they themselves had acknowledged the need for professional sleep help but needed to deal with the additional challenge of achieving professional validation of their sleep concerns. For example, Sally articulated how she felt education staff did not appreciate the challenge of her son's sleep problems. Despite describing her sleep concerns to school personnel, Sally described how they struggled to appreciate her situation and made the assumption that they would subside over time as he grew

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older. She expressed how she found this 'wait and see' approach to be a barrier to her receiving the help she needed:

> "I did talk to his school about this (..) I think they couldn't quite understand the severity of it. You know, 'Oh yes, he'll grow out of it' or whatever, and I'd think no, this is a real issue and ... yes, I thought they didn't maybe take it that seriously"

> (Sally)

In addition to generic practitioners, Sally also encountered difficulties having her sleep concerns validated by specialist sleep services. She described how she made a self-referral to a specialist sleep team, but her referral was only accepted with her Paediatrician's endorsement. Sally's story conveyed a sense of needing to invest significant time in persuading generic practitioners to take the responsibility she expected them to. This process involved presenting a convincing case for sleep help, being tenacious in discussing sleep concerns with professionals and gaining the endorsement of generic practitioners prior to securing appropriate help:

> "In fact actually I tried to contact them before the Paediatrician appointment and nothing was happening. So the Paediatrician said Right, I'll ring them up, we'll ring together' and they did take more notice because it was a Paediatrician ringing up"

> (Sally)

Maureen also recounted a difficult experience of gaining professional validation of her sleep concerns and described how she had been unsuccessful in persuading her

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GP to take her daughter's sleep problems seriously. Maureen felt the GP believed that the sleep problems were caused by poor parenting and she felt blamed and ignored. Her account also illustrated a sense that the GP's attitude was linked to a lack of listening and empathy:

> "I've been to my GP and asked for a sleep assessment and er, no, (..), very, very unsympathetic (..), it's like falling on deaf ears as far as I'm concerned, (..)I think they've always seen it as a parental issue rather than my daughter actually having real difficulties with sleep."

> (Maureen)

Maureen also reported how she took the autonomous step of recording details about her child's sleep in sleep diaries, which she used to evidence sleep problem severity and persuade generic practitioners to take her seriously. This addiitional activity was representative of the exceptional lengths parents sometimes felt they had to go to, to gain some professional validation of sleep concerns. However, Maureen described how she felt this was an unsuccessful strategy in terms of convincing generic practitioners of the gravity of their situation and getting the sleep support she needed:

> "I've done my own sleep diaries, and it's still not been enough for any professional to think oh maybe need to have a sleep study done on this child.(..) I started using it as evidence, to say look this is how bad it is. It wasn't anybody suggested I did it, it was really just to get other people to sit up and take notice"

Winnie's story of trying to secure professional validation of her sleep concerns indicated a belief that generic practitioners often failed to spend time learning about parent's individual situations. Winnie described how in her experience she felt professionals would treat all children with sleep problems in a similar way and did not appreciate individuality. Her account gave a sense that generic practitioners had not fully understood her son's sleep difficulties and more priority needed to be given to appreciating the characteristics of each case to ensure professionals were successful in responding to parent's need for support:

> *"They don't like to look outside the box at that next child that walks through the door; they're all the same, they're all labelled the same, and they're all… as far as I'm concerned treated the same, and it doesn't work"*

(Winnie)

Georgia considered the label of a disability diagnosis to be essential in gaining professional validation of sleep concerns and subsequent sleep support. She believed her concerns about her child's sleep were often not taken seriously, because her children did not currently have a disability diagnosis. Georgia was experiencing bedtime settling problems with both her primary school aged children who were being assessed for ASD. She believed that an ASD diagnosis was a 'passport' to accessing specialist sleep support and her requests for help would be ignored until this point:

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> *"We haven't got a diagnosis so ... I'm used to doors being shut, 'Wait*
>
> *'till you've got a diagnosis'."*

<div align="right">(Georgia)</div>

In comparison, Gloria and Sarah reported positive experiences of persuading generic practitioners and sleep practitioners to professionally validate their sleep concerns. Both parents described an initial assumption that they had sole responsibility for their children's sleep problems and had low expectations of professional sleep support. Therefore, they expressed an element of surprise and good fortune that generic practitioners had agreed with their concerns and set the wheels in motion for help. Gloria and Sarah's children both had a disability diagnosis (learning disabilities and Down's syndrome respectively) and were able to access specialist and sleep teams in a timely and straightforward manner. The following extract from Gloria describes her experience of having her sleep concerns professionally validated:

> *"I think we were possibly fortunate in that (..).once I'd explained*
>
> *what my problem was, and I must admit I didn't ... I didn't think I'd*
>
> *get any help, I didn't think anyone would actually ... take me*
>
> *seriously, just think oh, you know ... just get on with it. But Dr '..'*
>
> *actually put me through to the (..) team"*

<div align="right">*(Gloria)*</div>

Gloria and Sarah both reflected how their positive experiences had been helped by an existing relationship they had with generic practitioners and sleep practitioners

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who knew their families and understood their needs. They believed being known to and in regular contact with services were pivotal to their success in being taken seriously and accessing the support they needed. This understanding is shown in the following extract from Sarah:

> *"They knew about us, and their response was great, it was 'We know it's a problem, if you ringing us..and, bless her, she came out that night ..."*

> (Sarah)

4.8.3. Theme three conclusions. '"*The biggest problem is knowing where to get the help";* getting professional help'.

This theme describes the many challenges experienced by parents at the start of their journey towards improving their children's sleep problems. In the first sub-theme '"*Nobody's willing to take responsibility";* challenges around professional responsibility' parents considered what duty generic practitioners had to support them with sleep. Four parents conveyed how they viewed their children's sleep problems as a professional issue, and they expected more help from them and experienced significant challenges in sourcing help. Three parents conversely did not expect generic practitioners to take responsibility for their children's sleep problems and had a low expectation that help could be available. However, all seven parents who discussed the issue of professional responsibility also raised concerns that parents and generic practitioners had a poor awareness of specialist sleep services and that they were inadequately promoted.

The sub-theme '"*They couldn't quite understand the severity of it";* validation of sleep concerns' describes the experiences of parents in persuading generic practitioners to take their sleep concerns seriously. Four parents who did believe that generic practitioners needed to take more responsibility, reported difficulties convincing them and sleep practitioners that their sleep concerns were legitimate. These parents expressed how their worries had been dismissed and trivialised as poor parenting by generic practitioners with whom they first approached for help. Needing to gain the endorsement of generic practitioners before accessing specialist sleep services and collating sleep diary evidence were addiitional strategies employed by parents to try and achieve the professional validation they needed to access appropriate support. Parents spoke about needing a disability diagnosis to get generic practitioners to take their sleep concerns seriously. Furthermore, the call was made for greater understanding and empathy from professionals of families' individual situations to facilitate the validation process.

Positive experiences of two parents were reported in achieving professional validation of their sleep concerns. Both Gloria and Sarah described how an existing supportive relationship with generic practitioners who understood their individual situations had enabled them to take sleep problems seriously and instigate an opportunity of sleep help. These parents recounted how they had embarked on their journey with initial low expectations of professional help and so felt a sense of providence that they had had their concerns taken seriously by generic practitioners.

This theme identifies the challenges experienced by parents in accepting that professional help may be needed, persuading generic practitioners to support them and sourcing specialist sleep teams. These findings illuminate parents' early

experiences in their journey towards improving their child's sleep and are significant in enhancing professional's understanding of how initial contacts with services can be improved.

4.9. Theme four '"I'd love Supernanny just to come and spend some time with us"; visions of sleep support.'

Theme four '"*I'd love Supernany just to come and spend some time with us*";visions of sleep support' (Figure 9) explores parents' expectations and dreams of professional sleep support. The first sub-theme '"*Their answer was sleeping tablets and I refused;* disappointment with medication' deals with parent's experiences of sleep medication and not having their expectations of sleep help met. Sub-theme two '"*You just tick off the boxes";* the need for customised support' looks at parent's views around receiving SHE advice tailored to individual need. The third sub-theme '"*At home I think you can just relax more";* the importance of place' focuses on parents' preferences of where and how sleep support should be delivered. Each sub-theme will now be presented.

4.9.1. Sub-theme one –'"Their answer was sleeping tablets and I refused"; disappointment with medication'.

Sarah, Maureen, Corrine, Winnie and Kim reflected on being offered the sleep medication, melatonin, and described a sense of disappointment that their child needed medication to improve their sleep. Sarah was the only participant who reported being offered melatonin following a sleep assessment which involved an overnight sleep study, sleep history questionnaire and completion of sleep diaries. She expressed how she felt an initial sense of failure in needing to manage her

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daughter's sleep with medication. However, Sarah confirmed that the prescribing Paediatrician reassured her that many families required sleep medication which she took comfort in and influenced her decision to agree to melatonin. Whilst expressing these preliminary feelings of disappointment, she described how her overall experience of being offered melatonin was a supportive and positive one:

> *"I can remember breaking down in tears(..)..but the thought that she had to have medication ... I felt "What am I doing that's so wrong she's got to have medication?" and they said "Hey, we've got a book full of people who have this"...(..).. and I thought "Oh thank goodness for that, it's not me": So it was just that reassurance."*

> (Sarah)

The testimonies of Maureen, Kim, Corrine, and Winnie confirmed that they were offered melatonin without having their children's sleep problems formally assessed. A strong a sense of disappointment ran through these parents' stories that sleep practitioners had not invested time understanding the causes of children's sleep problems prior to deciding on treatment. Maureen had agreed to give her daughter melatonin and she had been taking it for some years. However, she described how she had expected generic practitioners to perform a sleep assessment and reassure her that possible underlying causes had not been overlooked prior to prescription. Her words conveyed how she felt giving medication 'glossed over' the sleep problem and merely treated the consequences rather than root causes. Furthermore, Maureen felt melatonin was only useful as a short-term measure as she felt it would be unacceptable to continue increasing the dose to maintain effectiveness:

"I would have liked to have seen a professional and somebody interview her and say yes we think she needs a sleep assessment or no this is just about you know, maybe they could have ruled out something else I don't know(..) It's like well how much more medication are they going to give her? Because all they're doing is reacting to something, they are not actually getting to the cause of it."

*(*Maureen)

Corrine's account of being offered melatonin for her son also conveyed a sense of disappointment that the prescription had been offered without due consideration. Corrine described how a Paediatrician had recommended melatonin following a brief conversation about her children's sleep problems. Her words indicate a perception that the sleep difficulty was over simplified and the decision to introduce melatonin was rushed:

"it was just purely discussion, and then they just wrote the prescription."

(Corrine)

Corrine additionally expressed a sense of regret that she had chosen to use melatonin to treat her son's sleep problem. Although she reported melatonin had been effective in improving his sleep, Corrine reflected on whether this was the most appropriate form of treatment. In the following extract she questioned if the same results could have been achieved for her son without having to take medication:

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> *"It's like having a headache tablet isn't it ...(..)... and not trying to get rid of that sort of headache tablet without, or that headache without any other means isn't it? So I suppose, looking back, it did the job ... but could there have been an equally better job without having to take, you know, medication."*

<div align="right">(Corrine)</div>

Kim and Winnie (who were also offered medication without prior sleep assessment) took the decision to decline this form of help. These parents communicated how this was down to personal preferences and how the offer of medication did not match their expectations of what sleep help was required. Winnie described an opposition to medication and how she was disappointed with this suggested intervention. She expressed how she felt sleep practitioners had failed to identify the fundamental causes of her son's sleep problem and their offer of medication was inappropriate:

> *"But their answer was sleeping tablets and I refused (..) because I don't… to me because of his problems, there's underlying issues that I think giving a sleeping tablet is just masking over the bigger problem"*

<div align="right">*(*Winnie)</div>

4.9.2. Sub-theme two- '"You just tick off boxes"; the need for customised support.'

Over the years, all parents described being offered some form of broad-based SHE advice from generic practitioners such as Health Visitors, GPs or Child and

Adolescent Mental Health workers, as part of general consultation sessions or for previous sleep issues when their children were younger. Parents reported mixed levels of satisfaction with this standardised type of sleep help, but all parents expressed a feeling of needing something extra to help address their children's current sleep problems (even though they had different views on whether it was the responsibility of generic practitioners to provide this). They described wanting more evidence based strategies that were tailored to their children's individual characteristics and situation. As the account of Kim demonstrates, she had accessed a general parenting course when her twins were younger and had found bedtime routine advice useful to help with moving them into their own, individual beds. However, Kim's son who had Down's syndrome later developed problems with disturbed night awakenings which she felt were not helped by this original advice. Kim acknowledged that his night disturbances needed tackling but was unsure of what was causing them or how they could be helped:

> "*It worked in the beginning with the routine with the stories…(..) I don't know now what you could do ..because it's like in the middle of the night and if his mind's not switching off…if his legs and arms are going, I don't know what support there could be.*"

(Kim)

Georgia also reported receiving standardised SHE advice through the conduit of a parenting workshop and described how she was dissatisfied with its generalist nature and limited relevance to her sons' needs. She explained how she had already experimented with positive bedtime routines with her two sons and found this to be ineffective in improving their sleep. Her words conveyed a sense that such standard

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advice alone was inadequate and she expected some additional, customised advice. Georgia expressed how she was frustrated that her children's need for an enhanced level of sleep help was not being recognised or provided for by generic practitioners:

> *"'Incredible Years' course, I've been on that. And they say that the routine, and I say but the routine's the same every time and ...(..)... Which I do every ... every time, and it doesn't work (..)But its, you know, if it doesn't work ... there's nothing to follow on from ... from that (..) you know, no extra support."*
>
> *(Georgia)*

Further probing revealed how Georgia had explored other avenues for sleep advice such as parenting support groups and social media forums. Georgia expressed how she had found this disability specific advice very helpful and valued how it had been evidenced through the trial and error of other parents in similar situations. Georgia did reflect on the reliability and accuracy of this information but explained it was better than no advice or standardised advice. Her words depicted a sense of resourcefulness but also feelings of desperation and disenchantment with the limited amount of professional support available:

> *"I get more advice going to (*parent support group) *(..)and Facebook..than what I get from professionals. Am I getting the right information? (..) I don't know. But it does help to have some information"*
>
> (Georgia)

Winnie, Maureen, Stella and Corrine reported receiving generic SHE advice by generic practitioners as part of general consultation sessions. Parents described how they were commonly given this advice based on short conversations about sleep and placed little value on its applicability to their children. Winnie explained how a voluntary children's disability organisation had offered some advice around creating a bedtime routine, but she had already found this strategy ineffective:

> "*They said routine, but that was when he was ... 'He needs to be in a routine', I said 'He's always been in a routine, his routine's never changed."*

<div align="right">(Winnie)</div>

Maureen's account of receiving generic SHE advice from generic practitioners also demonstrated how she had already tried suggested strategies and viewed them as intuitive and obvious to her as a parent. Maureen placed little value on such broad based advice and found it unsuccessful in improving her daughter's sleep problems. Her dialogue indicated a belief that giving standardised SHE advice was ineffective and more considered advice was needed to address her daughter's difficulties with sleep:

> "*Just the normal stuff you know, don't let her eat too late at night, no caffeine, really all the things you would do naturally as a mother anyway. (..), but I can't say that any of those have really had the desired impact no. It's just that you just tick off boxes.*"

<div align="right">(Maureen)</div>

Stella recollected her experiences of receiving fragmented standardised SHE advice from generic practitioners around routines and de-stressing techniques over a period of many years. She also described a feeling of frustration with this 'common sense' advice which she had already tried with little positive effect. Furthermore, Stella's words conveyed a perception of being patronised by generic practitioners who repetitively gave generic SHE advice. It was clear that she wanted some extra advice that was evidence based and specific to her son's individual needs:

> *"So there followed several years when I spent time being told to use like lavender oil, did I have a bedroom routine, as if it hadn't occurred to me."*

> (Stella)

Corrine gave a similar account of feeling patronised by standardised SHE advice given by generic practitioners and described her experience as negative and condescending. Corrine confirmed that she had already experimented with such mainstream advice around bedtime routines and this was intuitive to her as a mother. She expressed how she would have liked generic practitioners to assess her son's unique circumstances and customise sleep advice according to their findings, which she believed could have improved his sleep. Her words demonstrated a real tension between wanting professionals to help, being disappointed with what they had to offer and wanting extra, more detailed advice that was tailored to her son's sleep problem and individual needs:

"The community Paediatrician was almost patronising in terms of telling me.. we need to shower sort of.. no television, read him a story, and I said yeah, I'm doing that(..)But I suppose what I would be looking at is there anything in addition I could be doing? There was no extra support there, nobody sort of said 'Oh can we come out and monitor how he goes to sleep at night, or go through with you how you how you put a sleep routine together' ...(..).. And maybe that could have helped."

(Corrinne)

In comparison, Sally, Gloria and Sarah reported receiving SHE advice which had been customised to their child's particular needs following a professional sleep assessment. However, their stories revealed mixed levels of satisfaction with the quality of this advice. Sally described how her son's sleep problems had been assessed remotely by a specialist sleep team, who had interpreted completed sleep questionnaires and diaries and given particularised SHE advice over the telephone. She reflected how their advice was moderately helpful but sensed if sleep practitioners had observed her son at home, they would have achieved a greater understanding of his needs and given more specific advice. In comparison to parents who had received only generic SHE advice, Sally appeared to be more satisfied with the assessment informed, customised advice she was given but needed further specific advice that was evidenced by a more comprehensive sleep assessment process:

> *"They were very helpful but they just sort of kept saying the same thing, bring the bedtime ... closer by five minutes and things, and I thought well I would like them to see, actually see what happens to his brain and how it's suddenly keeping ... you know, wanting to be alert..and actually to give him some skills of how to ... calm himself basically."*

(Sally)

Gloria also described how a specialist sleep team had assessed her son's sleep problems and based their tailored SHE advice upon their findings. She explained how she had been advised to put together a specific bedtime routine for her son and to put him to bed at a consistent time each night. Gloria recounted following recommendations around sleep timing but felt unable to incorporate a consistent bedtime routine for her son due to her daughter's busy after school activity schedule. Gloria's discourse suggested a sense of being unconvinced as to the necessity of the advice around routines and reluctance to follow sleep practitioners recommendations systematically. There seemed to be a tension between wanting some well-considered, customised advice and needing to understand the rationale and research evidence behind it. Gloria's account may alternatively have demonstrated how some families will always find it challenging to follow sleep advice and to fit it in around the dynamics of their day to day lives:

> *"Routine –wise they, I think they just listened to me and said well you know, that's not going to happen…because we're not in (..) so..but I*

> *did always stick with the…still do with the right, eight o'clock, thats*
>
> *as near as damn it, that's where it all starts."*

<div align="right">(Gloria)</div>

Lastly, Sarah described being the most satisfied with the customised SHE advice she had received following a sleep practitioner's comprehensive sleep assessment. Sarah recounted how a specialist sleep team consisting of Portage workers trained in sleep interventions, had given her some very detailed, step-by-step advice around creating a relaxing bedtime routine and calming sleep environment. She reflected how whilst the advice took some time to work, it was effective in improving her daughter's sleep. Sarah's story demonstrated a positive example of a parent having their need met for extra, assessment informed, particularised SHE advice and seeing pleasing improvements in their child's sleep:

> *"From actually entering the room that he told us didn't he? So*
>
> *prepare the room before she goes up..so the bedroom light is on..the*
>
> *temperature's right..the bedcovers slightly open…so we're indicating*
>
> *to her 'Look it's now time to rest'(..)and it took a while didn't it ..but*
>
> *you could see her calm and that was their advice."*

<div align="right">(Sarah).</div>

4.9.3. Sub-theme three- '"At home I think you can just relax more"; the importance of place.'

The geographical location of where sleep support should be delivered to families seemed important to parents. The physical place where parents engaged with sleep

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practitioners and received sleep help received significant discussion in the interviews. Corrine, Sally, Winnie, Sarah, Gloria and Maureen expressed strong views about how it was important to them that sleep help was given in their familiar home environment. Corrine, Sally and Winnie described how they believed home visits could help sleep practitioners learn about their individual child and sleep problem. Corrine explained a perception that sleep practitioners needed to directly observe the child and home setting to accurately understand the nuances of each child's sleep problem and unique family situation. Within her description, Corrine alluded to the need for sleep practitioners to invest time in visiting parents at home, to build a rapport and ensure the ultimate advice was relevant to the child's individual needs. Corrine likened this vision of sleep help to the television personality 'Super Nanny'[2] which for Corrine encompassed the nature of support that she would like to receive at home:

> *"I would prefer the home visit because..(..)…you know, every child is different isn't it, and every set-up's different,(..) I mean there's been times over the year where you just think 'God, I'd love 'Super Nanny' just to come and spend some time with us and …give some advice'*
>
> (Corrine)

Sally also made reference to 'Super Nanny' as an exemplar model of sleep support that she would like to receive within the home setting. Sally described how she felt this type of direct contact with her family where the professional could observe how her son behaved at bedtime would be very helpful. She perceived this would assist

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[2] 'Super Nanny' is a reality television programme where parents receive parenting advice from an experienced nanny.

sleep practitioners in comprehensively assessing her son's sleep problems and result in relevant, effective sleep advice:

> *"I think somebody coming to the house, like Super Nanny, to see what's going on would be brilliant. Because there might be things you don't notice that you're doing(..) that could be better"*
>
> (Sally)

Winnie's thoughts on the merits of being visited at home by a sleep practitioner also focused on the possibilities it could open up for customised sleep advice. Winnie contemplated how if sleep practitioners came to her house and developed a true picture of her son's sleep problem, this would stimulate the production of different strategies to try that could improve his sleep. Her narrative implied a need for sleep practitioners to engage with her in the actual environment where the sleep problem took place, so that tailored advice could be given which she could have confidence in:

> *"For somebody to come out and I don't know, discuss what's going on I suppose, and say "Well have you tried this?", or....yah, you know and give ideas"*
>
> (Winnie)

In contrast, Gloria and Sarah expressed their preferences for home as the place where support should be given, based on their need to feel emotionally safe when discussing their children's sleep difficulties. Gloria firstly described how this was based on her own negative experiences of attending a sleep clinic where she felt that

the physical environment was unwelcoming and made her feel anxious and reluctant to share her concerns with sleep practitioners. She described bringing her mother to appointments to redress a perceived imbalance of power between her and sleep practitioners that the clinic setting magnified. Gloria concluded that she would have felt more comfortable receiving sleep advice in her own home:

> "*I also hated that, those are the worst type of appointments because…you know your heart's in your stomach by the time you've walked through the door, (..) the third time I went I took my mum with me(..) because I thought I can't do this on my own even though it…you know my mum never opened her mouth, it wasn't so much that, It's just I needed someone in my corner.(..) I think some places are too, just clinical. And they don't put you at your ease(..) whereas if you're in your home environment, you could have your support there.*"

> (Gloria)

Sarah's account of preferring home as a location to receive sleep support was also compared to the challenges she encountered with attending hospital appointments. Sarah raised the logistical challenges parents often faced around transportation and time pressures and how this may discourage them from engaging with sleep practitioners. Furthermore, she cited a tendency to feel tense in the physical environment of a clinic setting especially if she was required to bring her child and siblings along. The combination of feeling stressed making the appointment on time and emotionally uncomfortable in the sleep clinic environment discouraged Sarah

from working effectively with sleep practitioners. She resolved that supporting

parents at home made them feel emotionally safe and more open to sleep advice:

> *"For me personally, you have so many hospital appointments and it is the*
> *fact of getting them there and even though you have toys in the room, it's*
> *different when you're at home. Because I found I could really talk to the lady*
> *who came to us and (name) could play with her toys, she was content, she*
> *was calm…(..) at home I think you can just relax more."*

(Sarah)

Maureen similarly described challenges with the practicalities of receiving sleep help

in clinic settings. For Maureen whose teenage daughter had a dual disability and

mental health diagnosis, physically visiting unfamiliar environments was particularly

stress inducing. Maureen reasoned that if her daughter was expected to attend sleep

clinic appointments to receive sleep help, this would create an additional and

unnecessary layer of anxiety. She concluded that her daughter could be most

effectively helped in her own familiar home setting:

> *"With my particular child, home visits because if (child) was asked to*
> *go somewhere else she struggles enough going to medical*
> *appointments and seeing the Psychiatrist, because it takes her out of*
> *her comfort zone."*

(Maureen)

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Winnie, Stella, Sarah and Sally also discussed the merits of receiving sleep support in a group alongside other parents and reported mixed views on the appropriateness and effectiveness of this setting. Winnie expressed a belief that working with other parents who had similar experiences could be very empowering and stimulate the creation of useful ideas. Her dialogue indicated that she placed value on the real experiences of other parents and the place of a workshop setting could create a hotbed for effective sleep support:

> *"I think a group where…Because you can get titbits off other people "Have you tried this?" "No, I've not heard of that?" "Well try it, it might work for yours, it worked for mine."*

> (Winnie)

Stella was more explicit in her views that the sleep advice from other parents was more valuable than that which generic practitioners delivered. She described a clear preference for working in a group setting with other parents and creating solutions to sleep problems together in a supportive way. Stella's experiences of receiving sleep support from generic practitioners had been largely negative and she based her preferences for group settings upon this:

> *"I think with other parents, because I got a lot more out of parents than I have from professionals."*

> (Stella)

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In comparison, Sarah and Sally raised potential concerns with the group setting. Sarah cautioned that it would be difficult to fully appreciate the particular sleep problems of all children involved in a group workshop. She explained how she would worry about feeling ostracised from the group if her daughter's sleep problems were so distinct. Her words conveyed a perception that it would be challenging to achieve a customised level of sleep support within a group workshop environment:

> *"The only thing that worry me is that because each child is so individual, even if somebody's didn't seem as severe you might not feel… you fitted in with that group..I don't know, I just feel that I'd be a bit wary of that… because it could be such a variety of different conditions."*

> *(Sarah)*

Sally expressed similar concerns around the ability of the sleep workshop setting to meet individual need and acknowledged sleep problem variance within disability conditions and individual presentations of each child. Furthermore, she highlighted the potential pitfalls of parents sharing sleep advice that may only apply to their own child. In summary, Sally's views appeared to replicate Sarah's concerns that delivering sleep help in a group setting may be less effective in achieving a customised level of support:

> *"They're such…different levels that it's quite hard to compare in a way(..) so even people with same condition, it affects them in a*

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different way.(..) And it's very hard if you were talking to a parent

they would only know their experience

<div align="right">(Sally)</div>

4.9.4. Theme four- conclusions. '"*I'd love Supernany just to come and spend some time with us"*; visions of sleep support.'

This theme explores parents' hopes for appropriate and effective professional sleep support.' "*Their answer was sleeping tablets and I refused*"; disappointment with medication' considers how parents were commonly disenchanted with the offer of the sleep medication-melatonin. Despite some positive reports of melatonin being effective in treating sleep problems (Sarah and Corrine) all five parents who had been offered it expressed a sense of apprehension or un-ease with this type of sleep help. Additional concerns were raised about how in most cases, melatonin was offered unsupported by a sleep assessment, which appeared to add to parent's reticence about using it. It was clear from parents' accounts that medication was not their first choice of sleep help and they were commonly disappointed that this was the primary intervention offered to treat their children's existing sleep problems.

The sub-theme '"*You just tick off the boxes*"; the need for customised support' portrays parents' experiences of being offered generic SHE advice. Parents described how they felt the advice was intuitive and was therefore patronising, unsophisticated and inadequate in addressing their children's individual sleep problems. Parents illustrated how they expected more customised SHE advice that had been tailored to their child's particular needs, which a minority of parents had experienced some success in receiving.

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The final sub-theme '"*At home I think you can just relax more*"; the importance of place' focuses on the importance parents put on the location where sleep help is delivered by sleep practitioners. Five parents favoured the home setting and preferred sleep practitioners to visit them at home. Some parents believed this would enable sleep practitioners to 'get closer' to the sleep problem and gain a better understanding which may improve the quality of sleep help offered. Others argued that discussing children's sleep problems was easier to do in their own home environment and that this helped them feel emotionally safe which was important.

In summary, this theme illustrates parents' aspirations for effective sleep support. It uncovers parents' views about the acceptability of sleep medication, the effectiveness of generic SHE advice and need for particularised SHE advice delivered within the home setting. These findings are significant in evaluating how current sleep service provision matches up to parents' expectations and identifying opportunities for improvement.

4.10. Presentation of Sleep Practitioner Refined Themes

Two sleep practitioner refined themes 'sleep service accessibility' and 'inconsistent sleep support' (Figure 10) were developed during phase five of Braun and Clarke's (2006) thematic analysis process (Table 12). Following both rounds of sleep practitioner focus groups, the preliminary themes (Figure 8) were subject to another layer of analysis to refine and clearly define the details of each theme. Each theme will now be presented.

4.11. Theme one- Sleep service accessibility'

Theme one 'sleep service accessibility'represents sleep practitioner perceptions of how specialist sleep support services are accessed by parents and generic practitioners. The findings are divided into two sub-themes which will be presented sequentially: 'Expectancies and awareness of sleep help' and '"*Some families seem to fall through the net"; negotiating referral systems*'.

4.11.1. Sub-theme one- 'Expectancies and awareness of sleep help'.

Sleep practitioners discussed their perceptions of parents' expectancies of support with children's sleep problems. VFO from a voluntary organisation signposting team and VSP from a voluntary organisation consultation team described how they believed parents had low expectations that sleep support was available. They put forward the view that parents often believed sleep problems were an inevitable part of disability therefore they needed to carry on coping regardless. Therefore, it would seem that how parents viewed their child's sleep problem, was significant in their drive to find sleep services:

> "VFO: *People might assume the sleep problems are due to any*
>
> *disability the child has and don't realise and just say "Oh that's part*
>
> *of having whatever"*
>
> VSP: *Mmm I'd say that was a big problem.*
>
> VFO: *And just think 'Oh we need to get on with this' and don't*
>
> *realise that there could be underlying issues "*

Furthermore, SFSO from a social care consultation team expressed a perception that parents often believed disability was the main cause of children's sleep problems. She reflected how this was un-helpful in motivating and convincing parents that sleep could improve. This is significant in highlighting the importance of sleep practitioners raising parents' expectations that effective sleep interventions do exist and sleep help is available to aaddress sleep problems in children with DD:

> *"I think a lot of parents tend to think that their child has a sleep problem because they've got a disability and the way we work hopefully it allows parents to see that they can change."*

(SFSO)

Sleep practitioners also reflected how parents and generic practitioners (whom they often have first contact with) did not always agree on how sleep problems were defined. The concern was raised that generic practitioners sometimes overlooked the need to discuss attitudes towards children's sleep practices with parents, especially in relation to co-sleeping. This sometimes resulted in generic practitioners making inappropriate referrals to sleep consultation teams on a parent's behalf, where parents had no wish to change co-sleeping practices and did not view this as a sleep problem:

> *"VSP: We have a lot of families that are referred in by professionals because of a co-sleeping issue that the professional perceives as a problem so they then expect us to go in with this lovely little*

intervention with a parent who is not remotely invested in carrying it

out in the first place

VDO: *There's no point*

VSP: *No and you know, sooner or later 9 times out of 10 it becomes a problem and we can be there then but you can't get families you know to change something because you want them to change it.*

When parents clearly wanted help with children's sleep problems, concerns were raised in the focus groups about generic practitioners being poorly informed of sleep consultation teams. Sleep practitioners from social care, health and voluntary consultation and signposting teams (VFW, HSHV (1), SCP, VFO and VSP) all indicated they believed this was a problem. VSP explained it was uncommon to find generic practitioners knowledgeable about sleep consultation teams which meant there was often an element of chance in parents' quest of finding sleep support:

> *"It's about who you happen to ask about who you happen to phone or email"*
>
> (VSP)

The narrative of HSHV(1) from health consultation team HT2 also revealed the provision of specialist sleep support to be hidden from public view and poorly promoted. When asked if GPs knew about the specialist sleep services her team provided, HSHV(1) admitted they had a poor awareness. She described how GPs

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were aware of her team, but not necessarily of the sleep support which they provided as part of a holistic range of health services. She reported it was unusual for GPs to refer families to her team specifically for sleep advice but sleep problems tended to be identified in a holistic assessment as a secondary issue:

> *"They know about the team, the disability team but they wouldn't know there's people specifically within that team that would help with sleep and they wouldn't make a referral for that, (..), you know it might be one of the things they come with."*

<div align="right">HSHV(1)</div>

One sleep practitioner representing a health organisation consultation team disagreed with the view that generic practitioners were poorly informed about their sleep services. As demonstrated in the following extract, HLDN (1) described how her sleep team was reaching most families who needed it, because education and local authority services knew about them. HLDN (1)'s views indicated a belief that generic practitioners had a responsibility to be aware of specialist sleep services and pass this knowledge on to parents:

> *"Usually the families know about us from school…. Social workers, they're on their caseloads so"*

<div align="right">(HLDN (1))</div>

The majority concern around poor public awareness of sleep services prompted further discussion about how teams were actively publicised. VFO who represented a

signposting team expressed the need for consultation teams to be proactive in promoting their services. VFO conveyed an element of surprise in being unaware of the existence of ST1 for example, prior to their participation in the focus group. This raised concerns for VFO because if they as a signposting practitioner were unaware of this sleep service, the likelihood parents would be was questionable:

> *"I think the first thing is for parents to know that you're out there, cause I didn't know you were out there"*
>
> (VFO)

The on-line presence of sleep consultation teams was also explored by VSP, who reported being unable to identify local sleep teams by running an internet search. Further enquires at the Family Information Service (a family support signposting organisation) also failed to identify available sleep services. This illustrated both how sleep teams fell short of actively publicising their services and generic services had limited knowledge of sleep support:

> *"INV: Has anyone tried to google "Where do I go about my sleep problems?"*
> *VSP: I did (..) and I was hitting a brick wall and I ended up going to the family information service and it was very hard to find out what's out there (..)it just seemed to be you know some stuff was out of date and not great."*

VSP also commented on the success of VS1's website in promoting its' sleep services to parents and generic practitioners. She reported a belief that the service

title and advertising slogan of VS1 were confusing and did not clearly communicate the sleep support on offer. This had implications for the clarity of VS1's on-line presence and was an area in which participant VSP felt there was room for improvement. SCP also responded to questions around on-line presence and confirmed the social care consultation team she represented (ST1) was currently working on their website. This highlighted the importance of effectively branding sleep teams and clearly communicating the services on offer on-line:

> *"That's where (*VS1*' name) fall down because the name (*VS1*' name)*
> *will pop up and it doesn't give any understanding of the word. You*
> *just think brain and you might have a child who has Down's*
> *syndrome and think oh well it doesn't pertain to me."*

> *(*VSP)

A failure to actively promote sleep support also resulted in significant levels of un-met sleep need in the region according to VFW, VFO and VSP. They felt families were commonly unaware that they could receive sleep help and would try to manage without attempting to search for teams. This highlights the belief that poor sleep service awareness contributed to the masking of the true magnitude of sleep service need and more needed to be done to inform families that help was available:

> *"VFW: You've got parents at one end of the scale who have a high*
> *level of need who would get a lot of support. And then you've got the*
> *other parents who are very capable who will go to support groups*
> *and get support that way. And there's a lot of parents in between who*

have children with less need and they've got lower confidence and

they don't seek the help (....)But it's those hard to reach families

which are just managing"

VSP Mmm Firefighting."

SFSO, SCP, VFO and VFW suggested the creation of an on-line sleep team directory that could be quickly accessed by parents and generic practitioners, to improve the awareness and accessibility of sleep services. It was also suggested that linking the directory with well-known health and social care websites, may increase the likelihood of parents locating the site:

> "SFSO: *If someone in Wales types in "sleep, help sleep my child*
>
> *won't sleep" that it comes up top with a directory of what's available*
>
> *(..)*
>
> VFO*: That would solve the issue of having to trawl through lots of*
>
> *other information and you just want information on sleep (..)*
>
> SFSO: *It could be on the NHS website. Where do parents go? Which*
>
> *website do parents go to when they want help with anything? I don't*
>
> *know. Is it early support website?"*

4.11.2. Sub-theme two- '"Some families seem to fall through the net"; negotiating referral systems'.

Once parents had overcome the challenge of acknowledging their child's sleep problems and finding out about sleep servcies, sleep practitioners acknowledged the

additional challenge of negotiating referral systems. Sleep practitioners described how the sleep consultation teams they represented stipulated a myriad of referral criteria parents had to meet in order to enter their service. It can be seen from Figure 11 that VS1 provided the most accessible service for parents and accepted children between 0-18 years with a suspected or diagnosed disability condition of any type and covered a large geographical area spanning multiple regions. Sleep consultation teams HT1, HT2 and ST1 offered a service to single counties and set more restrictions on age range of the child and type of disability diagnosis. The following extract evidences the complexity of referral criteria specified by represented consultation teams and how this may represent an access barrier for parents:

> *"VSP: We take anybody from birth to 18 that either has diagnosis of a neurological condition of any sort or is under investigation for one (..) I've never experienced having to say no to anybody. (...) But we seem to have the widest criteria in my experience I think.*
> *HLDN (1) We only take children that have been diagnosed with a learning disability. There is an autism team, but they are only assessing and diagnosing autism (..) not giving any intervention work or any…………..and then there's CAMHS for children with Asperger, but I don't think they give any sleep advice(..)*
> *SCP: Our referral criteria for assessment and intervention is children with query ASD, Asperger,(...) so with or without er a diagnosis, just with general sleep problems and fitting HT1's criteria ,really of children with developmental difficulties or with chronic health problems."*

The setting of narrow referral criteria had the impact of excluding more families of children with DD and sleep problems. As shown in the previous extract and Figure 11, families of children with an ASD diagnosis did not qualify for any sleep help within the region covered by HT1 and would consequently be left to cope alone. Furthermore, in the teams which worked with younger children only (ST1) or older children only (HT1), children outside the set age range would be excluded from receiving a service. This meant that families either had to wait until their child was older, by which time the sleep problem may have worsened or become entrenched, or may have left it too late to ask for help. It is postulated that this demonstrates a level of inequity of sleep service provision amongst sleep consultation teams represented and a clear barrier for parents to overcome before receiving the sleep help they require.

Figure 11: Map of sleep consultation team accessibility

Referral criteria

Social care team 1 (ST1)

Sleep specific consultation team

0-9 years.

All disability conditions diagnosed or awaiting assessment. One county catchment area.

Internal referrals only from other members of the integrated team currently involved for an additional health need.

3 month waiting list

Health team 1 (HT1)

Holistic (incorporating sleep) consultation team

5-18 years.

Learning disability diagnosis excludes ASD. One county catchment area.

Referral routes

Open referral system

Waiting times

No waiting list

Health team 1 (HT2)

Holistic (incorporating sleep) consultation team

0-18 years.

Moderate to severe disabilities diagnosis. One county catchment area.

Internal referrals mainly from members of the integrated team but self referral possible.

No waiting list

Voluntary organisation team 1 (VS1)

Sleep specific consultation team

0-18 years.

All disability conditions diagnosed or suspected. Multiple county catchment area.

Open referral system

Unconfirmed waiting list

VSP expressed how she felt the restrictive referral criteria of other sleep consultation teams impacted on the team she represented. She confirmed how VS1's broad referral criteria often meant she picked up referrals for families who had been turned down by other teams. This was particularly common for children with ASD diagnoses and the following extract illustrates how their need for sleep support continues to exist even though some services exclude them:

> *"Some families seem to fall through the net as well they don't fit this criteria, CAMHS, they're too young for CAMHS we get a lot of them, they don't seem to fit anywhere else"*

> (VSP)

Discussions also revealed different referral routes through which parents could access sleep consultation teams. The majority of teams represented (HT1, HT2 and VS1) allowed parents to self-refer their child if they met the referral criteria (Figure 11). This represented a support to implementation for parents if they could speak with sleep practitioners and access teams directly and is demonstrated in the following extract from VSP:

> *"A parent could just pick up a phone today and say" help me" and I could go and do that, as long as the child has some sort of developmental, neurological problem (…)so yes in that sense it is very easy to get into our service."*

> (VSP)

In contrast, sleep practitioners representing ST1 reported a complicated referral pathway which parents had to negotiate. SFSO and SCP described how families firstly needed to be 'open' to ST1 or known to other practitioners within the wider integrated team and receiving a service for another health need. Furthermore, this requirement was in addition to earlier stipulations regarding disability diagnosis and age. This referral system was significant for families for two reasons. Firstly, access was controlled by 'gatekeeping' practitioners, introducing a possible period of delay or denial of sleep support. Secondly, sleep problems were classified as a secondary health need and families had to be receiving advice for other health issues such as continence before sleep could be addressed. This may have reflected an attempt to ration the service to those with multiple health needs. However, it is posited that this illustrates a disempowering system for parents that encourages a level of health service dependency. SCP and SFSO accepted the referral pathway was complicated and defended its complexity on the grounds ST1 was a new service as the following extract shows:

> *"They would have to be open , accepted for mainly work to do with ASD maybe some behavioural work, some toileting whatever or on the waiting list for nursing or psychology or social work, then they could be seen. (..)It is complex, (..) and this is our second year and so currently we are just accepting internal referrals"*

> (SCP)

SCP did however reflect that the referral system of ST1 was exclusive in nature and hampered the accessibility of the sleep service. As the following extract shows, SCP

accepted that placing such restrictions on service accessibility may result in significant numbers of families being denied sleep support and revealed a degree of inequitable service provision:

> *"We're only working with children who have been referred and they are open to somebody else so it's the internal referrals and I'm sure there are lots of other children out there who are not open to our service."*

<div align="right">(SCP)</div>

Sleep practitioners also described how the geographical availability of sleep services was another potential access barrier. VSP acknowledged that there was an element of chance involved in parents receiving sleep support according to where they lived and highlighted how some regions provided more services than others. The following extract demonstrates how this introduced another layer of inequitable service provision, parents may experience dependent upon the postcode in which they lived:

> *"It's very hit and miss in terms of where about in the country you live. Erm what kind of services are already in place."*

<div align="right">(VSP)</div>

Finally, sleep practitioners described the need for timely access to sleep consultation teams, and reported that HT1 and HT2 did not currently operate a waiting list (Figure11). HLDN (1) elucidated how HT1 managed parents' requests for help, by

adopting a triage approach to assess whether involvement was appropriate. HLDN (1) explained how referring generic practitioners often failed to adequately discuss sleep problems with parents and it sometimes became clear from triage conversations that parents did not want sleep help. HLDN (1) believed this approach had a positive impact on waiting list size, which was previously 'clogged up' with inappropriate referrals. This demonstrates the need for sleep practitioners and parents to discuss and agree on sleep problems at the beginning of parents' SHE journey:

> "*We use a choice and partnership approach so we ask parents to opt in, because we get a lot of parents that have been referred by social workers and they don't want the input, but they say they want the input cause they are too frightened to say that they don't. So we invite them in and we've cut down our waiting list by doing that.*"

> (HLDN (1))

VSP was less clear on the existence of any waiting list in operation for the sleep services of VS1. VSP described how she was unsure of how long parents typically had to wait to receive a service and this was often dependent on geographical location. In common with HT1, VSP reported that VS1 also adopted a triage approach to manage referrals into the team which was largely useful for prioritising cases. VSP described how this system helped her to see families with the most urgent level of need first but also blurred the boundaries in terms of usual waiting times:

> "*It very much depends on where they are in the country and indeed just how desperate they are, we have somebody who works for us as a*

sleep administrator and they tend to make first contact.(…) So it's

hard to, kind of pinpoint what the real waiting list is"

<div align="right">(VSP)</div>

SFSO confirmed ST1 currently operated a three month waiting list which had built up due to resource constraints. SFSO explained how she was the only sleep practitioner delivering sleep support, and how interventions which were lengthy for some families could impact on waiting times. However, a three month waiting list would still be considered acceptable by NHS standards which stipulate a maximum twenty six week wait for therapy services (WAG, 2015):

> *"It's only me really supervised by SCP(..),every case is different, so*
> *I've got about 6 on the waiting list at any time really(..) So kind of say*
> *3 months."*

<div align="right">*(SFSO)*</div>

4.11.3 Theme one conclusions. 'Sleep service accessibility'.

This theme highlights sleep practitioner perspectives of pre-implementation challenges faced by parents embarking on a journey towards improving their children's sleep. In the sub-theme 'expectancies and awareness of sleep help' how parents viewed their child's sleep problem in relation to the DD diagnosis seemed significant in their drive to find sleep support. Expectations that sleep could be improved with professional help, seemed to be lowered when parents believed that disability caused children's sleep problems. Linked to this was a common disconnect

between how parents and sleep practitioners viewed sleep problems, and the need to discuss them and agree on when and if parents wanted help. Also raised was the poor promotion of sleep consultation teams and limited public awareness of their existence. It was posited that all these factors helped to mask the true magnitude of sleep service need amongst children with DD.

The sub-theme "*Some families seem to fall through the net";* negotiating referral systems' illustrates a myriad of referral criteria that parents have to navigate to access sleep teams, revealing another pre-implementation barrier. Some families in comparison with others may have been able to access sleep consultations teams more easily if their child 'matched' referral criteria specified. It is posited that this implied a level of inequitable service provision. Furthermore, disparate geographical availability and waiting times were described by the four sleep consultation teams represented which introduced another layer of inequity for parents trying to access help. However, one team (HT1) described how they had been successful in eliminating their waiting list, by adopting a triage approach to referrals and openly discussing and agreeing on the existence of sleep problems with parents which links to sub-theme one. Overall the diverse range of referral criteria stipulated by local sleep services implied a level of inequitable service provision and highlighted a need for greater inclusivity.

4.12. Theme Two- Inconsistencies in sleep support

Theme two 'inconsistencies in sleep support' (Figure 10) represents sleep practitioners' reports of how they support parents with children's sleep problems. Two sub-themes will now be presented: 'nature of assessment' and 'nature of treatment implementation'.

4.12.1. Sub-theme one- Nature of assessment.

Discussions revealed wide variations in how sleep practitioners assessed children's sleep problems (Figure 12). Disparity could firstly be seen amongst members of the same sleep consultation team: HT2. First focus group participant HSHV (1) reported not using sleep diaries, but respondent validation interview participant:HSHV(2) described regularly supporting parents to complete them. A lack of clarity around the sleep assessment process was also evident from the dialogue of HSHV (1) who reported combining this with the holistic assessment of the child and family. This practice was unique to sleep consultation team HT2 and revealed a poorly defined sleep assessment process which, it is posited, may have impacted on sleep practitioner's ability to fully understand the sleep problems presented.

Figure 12: Map of sleep assessment processes undertaken by sleep consultation teams

Social care team 1 (ST1)	Health team 1 (HT1)	Health team 1 (HT2)	Voluntary organisation (VS1)
Sleep specific consultation team	Holistic (incorporating sleep) consultation team	Holistic (incorporating sleep) consultation team	Sleep specific consultation team
2-3 week assessment process where parents are supported through home visits to complete sleep diaries, in-house sleep history questionnaire, CSHQ, QOL measures: PSI, PCQ and GHQ. Psychological formulation to interpret findings and identify underlying sleep disorders.	2-3 week assessment process. Triage assessment completed at a clinic consultation. National sleep organisation sleep history questionnaire, CSHQ, SDI, PSI, PCQ and GHQ completed. Parents then advised how to complete sleep diaries. Underlying sleep disorders identified.	Via home visits, sleep assessment completed as part of holistic assessment (no sleep history questionnaires). Sleep diaries used by some practitioners with selected families. Duration of assessment process unspecified. No interpretation of assessment findings reported.	Triage assessment completed by administrator. Sleep diaries and in-house sleep history questionnaire sent to parents in advance of first visit. Support given only in identified cases of additional need. Assessment completed in 1 home visit. Underlying sleep disorders identified.

Abbreviation key:
CSHQ– Child sleep habits questionnaire, QOL– Quality of life, PSI–
Parenting stress index, PCQ– Parenting sense of competence index,
GHQ– General health questionnaire, SDI– Sleep disturbance index.

"The assessment would be a holistic assessment of everything that's going on and you talk about the background, the history (..). We don't have specific sleep diaries"

(HSHV (1))

"I would go through it first with them, just to tell them why we are doing it and what information we need that sort of thing, then obviously I'd give them the diary then to complete over time"

(HSHV (2))

In contrast, sleep practitioners representing all other sleep consultation teams reported using sleep diaries as part of the sleep assessment process. Whilst most agreed completing them could be onerous for parents, sleep practitioners gave varying accounts of how they supported parents with this task. VSP described how VS1 sent parents sleep diaries and a sleep history questionnaire in advance of sleep practitioner contact and guidance was only offered to parents with identified additional support needs. The policy was then to only engage with families once assessments had been returned:

"We don't tend to take on a referral unless they have completed the diaries and the questionnaires and returned them back (..)we have a sleep assistant who would check in and find we have parents with learning disabilities themselves, if there's anything like that going, then all the other rules go out of the window, and we can help on whatever level is necessary"

In contrast, SFSO, SCP, HLDN (1), HSHV (2) and VFO) expressed the view that all parents needed a lot of support to complete sleep diaries effectively. It can be seen in the following extract how SFSO and VFO agreed parents needed reassurance from sleep practitioners and suggestions of ways in which to record sleep information that parents found acceptable and achievable:

> *"Make sure they know they are being supported to fill in those diaries as well and like you said it doesn't matter if there's scribbles .... A lot of parents are afraid of misspelling things or that their English or Welsh isn't up to standard. But you just have to reinforce that it's not a problem. And think of different ways of recording as well. Ask parents would you prefer something where you can just tick the times or having to write out"*

(SFSO)

Other sleep assessment tools used by sleep consultation teams included sleep history questionnaires that were 'home-grown' (VS1 and ST1) or produced by a national sleep training organisation (HT1). ST1 and HT1 also used the validated sleep outcome measure - the Child Sleep Habit Questionnaire (CSHQ) and quality of life (QOL) outcome measures, the Parenting Stress Index (PSI), Parental Concerns Questionnaire (PCQ) and General Health Questionnaire (GHQ) as part of the sleep assessment process. This is significant in demonstrating emerging inconsistencies in type and comprehensiveness of sleep assessment processes across represented teams:

> "*SFSO: We use the Children's Sleep Habits Questionnaire, that's part of our questionnaire pack ( )*
>
> *SCP: We look at parents with mental health issues and stress, we use the PSI Parenting stress index (..) General health questionnaire (..) HLDN (1) We've just done (National sleep training organisation), so we are just completing that really.*"

Sleep practitioners also discussed how assessment findings were interpreted so as to improve sleep problem understanding and inform correct intervention design. ST1, HT1 and VS1 reported how the assessment process helped to identify underlying sleep disorders and referrals could be made to other services where necessary. The interpretation sleep assessment data were not discussed by representatives of HT2, which may have indicated a knowledge gap or stem from their service not using specific sleep assessments as reported earlier:

> "*VSP: If it's a behavioural disorder we can pick it up during the sleep questionnaire the diaries you know not far down the line, but if it's anything medical, we'll refer straight back and very often we can help things like sleep studies happening you know (..)*
>
> *HLDN (1):(*National sleep training organisation)*, the paperwork can diagnose the sleep terrors, night terrors the sleep walking(..)*"

Interpretation of sleep assessment findings was described in more depth by SFSO and SCP, who described using a psychological formulation technique. This involved identifying vulnerability factors such as any medical conditions predisposing a child

to sleep problems or behavioural causes which reinforced the sleep problem. Also highlighted were family strengths that could be drawn upon to help implement intervention strategies. SFSO and SCP emphasised how they used this process to stimulate parental empowerment and partnership working. However, the fundamental purpose of this process was to clearly, visually represent the complex nature of a child's sleep problem and improve parent and sleep practitioner understanding:

> "*We look back at possible vulnerability factors (..) and then we look at the possible triggers, something may have happened recently, some change maybe (..) after the assessment we have a specific session, we have it all out on a piece of paper yer, draw it all out (..) summarising all the information but we want them to kind of voice, we know what's in the file, but we want them to try and put it together like a jigsaw.*"

> (SCP)

SCP, HLDN (1), VSBM and VSP all discussed the importance of starting to build a trusting relationship with parents during the assessment process. At this stage this supported parents to feel comfortable in revealing information pertinent to the understanding of the sleep problem which may not have been shared in the absence of a rapport with sleep practitioners:

> "*SCP: You find that the assessment process is building a relationship with parents it's time consuming but it's really valuable, crucial really*

HLDN (1): Yes you find out things that perhaps you wouldn't have

found out without doing that"

Closely linked to the need to develop a good working relationship with parents was the notion of agreeing with parents if they were ready to embark on sleep work. Determining readiness was explored by VDO, VSP and HLDN (1), who acknowledged how challenging sleep advice could be to implement and argued parents needed to choose a good time to start. This was significant because if the timing of implementation was poor, intervention success could be affected as the next extract shows:

"It's worded in our questionnaire "Is this a good time to start a sleep

programme?"(..) we can perhaps give them more of a coping tool you

know, let's get through this period and perhaps try a few of these

things."

*(*VSP)

4.12.2. Sub-theme two- Nature of treatment.

Wide variation was evident in the nature of treatment offered by sleep consultation teams (Figure 13). VSP, SFSO, HSHV (1), VFO, VDO and VFW discussed how VS1 (and various signposting teams) commonly gave out standardised SHE advice to families un-supported by sleep practitioners and prior to assessment. VSP described how VS1 gave ad-hoc SHE advice in a regular newsletter for parents to try out independently with their child. The following extract demonstrates the intention

of empowering parents to make a positive start by focusing on the fundamentals of
SHE:

> *"We offer little titbits for parents in our quarterly bulletin (..) a*
> *checklist of kind of things to look at before you go any further you*
> *know ruling out the more simple kind of things."*

> *(*VSP)

VFW also noted how SHE advice booklets such as the one signposting team VS4
produced, were abundantly circulated to parents by generic practitioners.
Furthermore, parents were at liberty to freely download similar booklets from other
organisations and implement them without formal sleep assessment. VFW did
however voice some concern that this practice could be ineffective for families of
children with DD.

Figure 13: Map of SHE interventions undertaken by sleep consultation teams

Figure 16 Map of SHE interventions undertaken by sleep consultation teams.

Social care team 1 (ST1)

Sleep specific consultation team

SHE offered tailored to family need, supported by rationale and 'psycho-education'. Total home visits – 15 plus telephone support over 12 month period. Interventions evaluated by repeating sleep diaries, PSI, PCQ and GHQ and completing in-house feedback form.

Health team 1 (HT1)

Holistic (incorporating sleep) consultation team

SHE offered tailored to family need supported by rationale and 'psycho-education'. Total contacts- 4 clinic appointments over 6 week period. Home visits available on request. Interventions evaluated by repeating sleep diaries and SDI.

Health team 1 (HT2)

Holistic (incorporating sleep) consultation team

Standardised SHE offered, supported by unspecified number of home visits over a period of 8 months. No intervention evaluation process reported.

Voluntary organisation (VS1)

Sleep specific consultation team

SHE offered tailored to family need supported by rationale and 'psycho-education'. Total contact 2 visits over 12 month period. Interventions evaluated via in-house feedback form. # Ad-hoc SHE advice also offered prior to sleep assessment in monthly newsletters.

Abbreviation key:
CSHQ– Child sleep habits questionnaire, QOL– Quality of life, PSI–Parenting stress index, PCQ– Parenting sense of competence index, GHQ– General health questionnaire, SDI– Sleep disturbance index.

"But our publications you know lots of other professionals hand out our

sleep booklets etcetera. But any family they are going to be able to access

some kind of sleep publication (..) and that doesn't quite meet the needs of

our families"

(VFW)

VSP, SCP and SFSO further reflected how it was not best practice to offer SHE advice without first establishing the causes of the sleep problem. They confirmed SHE was less effective for some underlying sleep disorders, for example, and it was therefore essential these were identified and ruled out first from assessment information. These sleep practitioners concluded it was preferable to offer SHE advice following a full sleep assessment and interpretation of data:

"VSP: If a child has a circadian rhythm disorder and we're saying

"right get your kid to bed for 8 o'clock" but they are not falling

asleep till four, well nothing that's in that sleep hygiene is going to

work.(..)

SFSO: Yes it's after the assessment, it's after the formulation."

VSP continued to suggest that giving SHE advice on such an ad-hoc basis without formal assessment and support could have a negative impact on families depending upon how they interpreted the advice. For example, there was a risk of parents over estimating the ease and speed of improved sleep outcomes when they followed the advice. Alternatively, families may view the advice as too simplistic and feel sleep

practitioners were overlooking their child's sleep problem. VSP concluded that in some cases, giving impromptu SHE advice to families was not best practice:

> "*I think it can be quite damaging to just deliver sleep hygiene advice as a first port of call (..) parents, sometimes think by painting my child's bedroom neutral colours and taking out all the toys, that this is going to solve the problem, it can be taken as "you're not taking me serious" (...) So think that sometimes it can be the wrong thing to do to just hand out sleep hygiene"*

> (VSP)

The narratives of HSHV (1) and HSHV (2) indicated how they were unique in giving out standardised SHE advice that was not tailored according to individual family need. This chimes with their reported practice of performing holistic rather than sleep specific assessments and subsequent limited comprehension of sleep problem causes. As the following extract from HSHV (2) shows the advice usually given was imprudent and optimistic. It is posited that this implies sleep practitioners sometimes adopt a 'scatter-gun' approach to delivering SHE, which evidences the need for developing supporting programme theory and making interventions explicit:

> "*It would be an on-going process with most of them, trying different things seeing what worked.*"

> (HSHV(2))

In contrast, VSP and SCP showed agreement for reserving SHE advice until after a sleep assessment and customising advice to individual need. Sleep practitioners debated the merit of having a SHE advice checklist to select from based on assessment findings. Sleep practitioners concurred that this practice was helpful but non-directive language should be used when giving advice to ensure parents did not feel patronised:

> "SFSO*: A checklist but not kind of "do this, do not do this" it's a "Try to". Families do not want to be dictated to they want to be indicated to why (..)*
>
> *SCP: Having a little bit of routine for some families would do the trick, whereas having you know a brief routine for some families wouldn't do it, It's very individualised."*

VSP and SFSO agreed that in addition to giving out tailored SHE advice in a supportive manner, parents also needed to know the underpinning rationale. VSP explained how she perceived parents often found SHE too simplistic and were disappointed that novel techniques were not suggested. To counteract this, VSP detailed how when parents understood why they were being asked to make changes they were more likely to follow the advice. It is posited that explaining the reasons behind advice is a hidden mechanism which can make SHE interventions successful:

> "If they are not explained to parents as to why they work like turning the TV off (..) , the science behind the blue light and melatonin (..) you're just being told to do something but you are not being told why

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or actually just how serious it is. And sometimes that knowledge to

parents can be that tiny little piece of a puzzle that's missing"

*(*VSP).

Addtional information on sleep theory termed 'psycho-education' by sleep practitioners was also seen as useful in preparing parents for successful implementation of SHE advice. SCP, SFSO, HLDN (2) and VSP all agreed that it was beneficial to spend time informing parents of the different stages of sleep and the role of circadian rhythms for example and how these relate to the child's sleep problem. This process helped to improve parents' understanding of the sleep problem, put it in perspective and gave them increased confidence to proceed with SHE advice:

> "SCP: *We incorporate psycho-education as well don't we? We think*
> *about the benefits of sleep and how sleep impacts you know on all*
> *other aspects of our life and also sleep rhythms and stuff like that (..)*
>
> VSP: *Sometimes just normalising what a child's doing can be enough*
> *for a parent to see some light at the end of the tunnel."*

Whilst sleep practitioners agreed on the customised nature of SHE, disparity was reported around the intensity and duration of support offered to parents to implement advice (Figure 13). VSP confirmed offering two home visits to parents over a 12 month period incorporating assessment and intervention, whereas SFSO reported offering multiple visits in the same time frame. VSP argued this 'support-light'

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model worked well for motivated parents, but acknowledged other parents may need more support. However, the main implication of this disproportionate provision of support across sleep consultation teams was that parents would receive markedly different levels of assistance to apply SHE advice depending upon which team they engaged with:

> "SFSO: *Well over the first month it can be every week, it can be twice a week, then it drops off a bit ..I've got contact sheets galore for some families(..)*
>
> VSP*: Our service does rely on the parent largely to understand that this is hard work and you have to do it which means when we do get results, we get great results but there is the other side of the spectrum.*"

In discussing how parents could be better supported to implement SHE advice, it was suggested that parents of children with DD who had been successful in overcoming sleep problems in the past could offer other parents moral support. VFO, VSP and SCP discussed the possibilities of a parent buddy system whereby experienced parents could be trained to help parents to try out SHE recommendations. It was proposed that parents could feel more motivated and able to engage with sleep practitioners if their advice was endorsed by other parents with a deeper understanding of their situation:

> "*It's very powerful to have er a parent as a buddy and as er I agree with you really it's more kind of moral support, cause I was there and I've been through it and you will get through it.*" (SCP)

The ways in which sleep improvements were monitored and evaluated by sleep consultation teams were discussed. This revealed another element of disparity between represented teams (Figure 13). For example, the success of sleep interventions were quantitatively measured by only two sleep consultation teams- ST1 and HT1- whose representing sleep practitioners confirmed re-running sleep diaries at the conclusion of a SHE intervention. Furthermore, only ST1 was reported to repeat QOL outcome measures and actively use them to measure how families' lives improved following intervention. However, VSP and SCP were more vocal on the importance of qualitative feedback from parents gathered via bespoke evaluation forms developed by VS1 and ST1. It is posited that this picture represents how sleep consultation teams arguably under value the importance of measuring SHE intervention effectiveness:

> *"VSP: We tend to go down the route of you know anecdotal (..)we*
> *have feedback forms that we send out (..)*
>
> SCP*: We have qualitative feedback forms that we have created as*
> *well not just in terms of is sleep any better but how are parents*
> *feeling in terms of competence(..) but most important is the*
> *qualitative rather than the numbers."*

Sleep practitioners also discussed how parents might be supported with children's sleep problems through the use of melatonin prescribed by Paediatricians. There was broad agreement amongst VFW, SFSO, VSP, SFSO and HSHV (1) that melatonin was often prescribed inappropriately and without the consideration of a sleep assessment. Sleep practitioners expressed how it was common for children to be

prescribed melatonin fundamentally because of their DD diagnosis, rather than taking the time to understand the sleep problem first and tailoring interventions according to actual need:

> "VFO: *They are prescribed melatonin without actually asking about the bedtime routines and things, "What have you got in place?" It was just "This is a medical issue"(..)*
>
> VSP: *It's just compounding the theory that because they have this condition this is just part and parcel, here's another medicine to take."*

Discussions also highlighted tensions between melatonin prescription and motivating parents to try out SHE advice. SFSO, VSP, VDO and VFO discussed a belief that more parents were becoming aware of melatonin and requesting it as a 'quick-fix' solution. Sleep practitioners expressed how the ease of taking melatonin coupled with expected benefits, made it more appealing to parents than following SHE advice and could represent a barrier to implementation:

> *"We always have a massive battle cause when you're giving behavioural strategies (..) what you're giving them is just a big bowl of hard work, because the Paediatrician can hand over the prescription for melatonin like this, and this is something tangible that we can take."*

(VSP)

It was also acknowledged by SFSO, VSP, VDO and VFO that when parents did agree to follow SHE advice and their child was already taking melatonin, it could be difficult to ascertain which intervention was effective. This is significant because implementing SHE and melatonin concurrently, makes it harder to develop an explicit understanding of how SHE works and when it has been successful or not. Furthermore, it may be unnecessary to 'double up' treatments and this represents a disconnect with the prudent healthcare agenda which recommends use of the minimum appropriate interventions only:

> "*I think there's co-existing issues of melatonin and people seeking behavioural advice at the same time, it's hard to know what's doing what?*(..) *it could be the routines that they are putting in place and melatonin isn't really having that effect.*"

> (VSP)

4.12.3. Theme two conclusions. 'Inconsistent sleep support'.

This theme captures the wide-ranging nature of sleep support provided largely by sleep consultation teams. Sub-theme one shows how teams vary in their sleep assessment approaches in terms of tools used, support given to parents to complete them and sleep practitioner competence to interpret assessment results. This reveals further inequities in provision across sleep consultation teams and draws into question the appropriateness of subsequent interventions based on poorly interpreted assessment findings. However, broad agreement was reached on how the sleep assessment process was important in improving parent and sleep practitioner understanding of the sleep problem. Furthermore, it was highlighted how the sleep

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assessment process can determine readiness of parents to embark on a SHE intervention and facilitate supportive working relationships which were considered pivotal to intervention success in the long term.

The 'Nature of treatment' sub-theme also describes significant inconsistencies across teams in terms of SHE treatment offered to families. Although most sleep practitioners conceded that customised advice should be given following a sleep assessment, a lack of agreement existed around the level of support parents needed to implement it. Furthermore, some teams incorporated 'psycho-education' into interventions and gave parents the rationale underpinning advice to support implementation. This demonstrated a need for a more consistent approach to SHE delivery to ensure parents receive an equitable service. This sub-theme also highlights how SHE is often applied concurrently with melatonin which was commonly prescribed inappropriately without a sleep assessment. Sleep practitioners raised concerns that prescription de-motivated parents to implement SHE and concurrent implementation was unhelpful in understanding which intervention worked to improve sleep. It is important to recognise that SHE is commonly implemented as part of a broader package of care which includes medication, when developing an explicit understanding of all the factors which impact on the success of SHE as a complex intervention.

4.13. Summary of data sets

This study aimed to answer three broad questions around SHE practice and how this relates to scoping review findings, stakeholder perceptions of SHE experiences and ideas about what SHE should look like and how it should be implemented. Four themes underpinned by sub-themes which reflected parents' perceptions of SHE

were developed. "'*Well this is how it is*"; beliefs about sleep problems' represented parents' experiences of living with chronic and severe sleep problems and the daily challenge and wider impact of managing them. 'Ways of knowing about sleep' described the different forms of knowledge parents have about sleep problems, SHE advice and various sleep management strategies. "'*The biggest problem is knowing where to get the help*"; getting professional help' explored parents' experiences of becoming involved with sleep practitioners with regards to children's sleep problems. The fourth parent theme: "'*I'd love supernany just to come and spend some time with us*"; visions of sleep support' explored parents' expectations of professional sleep support. Two themes underpinned by sub-themes which reflected sleep practitioners' perecptions of SHE were also developed. 'Sleep service accessibility' represented sleep practitioner views of how specialist sleep services are accessed by parents and generic practitioners. The second theme: 'Inconsistencies in sleep support' which captured the wide-ranging nature of sleep support provided largely by sleep consultation teams. These two data sets (parent and practitioner themes) were then synthesised and combined with scoping review findings to produce six overarching themes which are presented next.

4.14. Synthesis of data sets

To tell the overall story of the evidence base and describe stakeholder's perceptions of SHE and how it should be implemented; a synthesis of parent and practitioner data sets and scoping review findings was developed. Six themes are presented (Table 18).

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Table 18 Six overaching themes

1.	Parents and practitioners have a shared understanding of what a sleep problem is.
2.	Sleep services are well publicised and accessible for parents.
3.	Parents and sleep practitioners develop a safe and supportive relationship.
4.	Parents and sleep practitioners improve their understanding of the sleep problem.
5.	Regularity and quality of child's sleep improves
6.	Quality of life improves for the family

4.14.1. Theme one: Parents and practitioners have a shared understanding of what a sleep problem is.

Exploratory study findings revealed significant pre-implementation challenges around stakeholders agreeing on the existence of children's sleep problems. This was largely un-explored in the scoping review and represented a new strand of evidence to inform understandings of SHE. The parent sub-theme '"*They couldn't quite understand the severity of it";* validation of sleep concerns' illustrated parents' experiences of struggling to convince generic practitioners that their children had sleep problems. The parent sub-theme 'Ways of coping' also emphasised how it was important to discuss with parents at the start of an intervention, any potentially un-helpful coping strategies such as co-sleeping and reflect on their views about them. In addition, the sleep practitioner sub-theme 'Expectancies and awareness of sleep help' highlighted the need for sleep practitioners to explore existing coping strategies such as co-sleeping at the outset and agree with parents if any sleep help was needed. The evidence presented from parent and sleep practitioner themes thus indicates the need for stakeholder agreement around the existence of children's sleep problems as a preliminary requisite of SHE which warrants further discussion in the co-design study.

4.14.2. Theme two- Sleep services are well publicised and accessible for parents.

Exploratory study findings revealed important pre-implementation issues around parents' awareness of and ability to access sleep consultation teams, which were not identified in primary studies included in the scoping review. In the parent sub-theme '"*Nobody's willing to take responsibility";* challenges around professional responsibility', parents reflected how they had low expectations of support and resigned themselves to coping alone which affected their determination to seek out services. Furthermore, parents reported how generic practitioners were commonly unaware of where to refer parents for specialist sleep help, indicating that sleep consultation teams were poorly promoted. The parent sub-theme 'Understanding sleep problems' also showed how parents' beliefs around the causes of their child's sleep problems could affect their drive to seek help. In the majority of cases, parents understood the sleep problem to be part of the disability, making it inevitable and beyond help. The sleep practitioner sub-theme 'Expectancies and awareness of sleep help' similarly evidenced the significance of parents' understanding of sleep problems and how this impacted on their motivation to request support. This sub-theme also highlighted how sleep consultation teams had a low public profile which meant parents, generic practitioners and signposting practitioners had a limited knowledge of their existence.

In terms of accessing sleep teams, the parent sub-theme '"*They couldn't quite understand the severity of it";* validation of sleep concerns' evidenced how parents struggle to navigate their way around the strict referral rules of sleep consultation teams. The sleep practitioner sub-theme '"*Some families seem to fall through the net";* negotiating referral systems' illustrated a myriad of referral criteria and routes stipulated by sleep consultation teams, which confirmed the level of challenge often

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facing parents. Furthermore, such restrictions around accessibility implied a level of inequitable service provision and need for greater inclusivity.The evidence presented from stakeholder themes thus suggests the need for parents to have greater awareness of and accessibility to sleep help, which is worthy of further debate in the co-design phase.

4.14.3. Theme three- Parents and sleep practitioners develop a safe and supportive relationship.

Exploratory study findings substantiated the limited evidence reported in the scoping review around the need to build supportive parent/sleep practitioner relationships. The '"*Well this is how it is";* beliefs about sleep problems' parent theme called for sleep practitioners to develop a deep empathy with parents' situation before parents would consider listening and following SHE advice. The parent sub-theme '"*At home I think you can just relax more";* the importance of place' demonstrated parents' need to feel safe in the environment where sleep support was delivered. In addition, the '"*They couldn't quite understand the severity of it";* validation of sleep concerns' parent sub-theme showed how sleep practitioners needed to empathise with parents' initial sleep concerns. Sleep practitioners also demonstrated how they were aware of the need to foster understanding relationships with parents in the 'Nature of assessment' and 'Nature of treatment' sub-themes. Stakeholder evidence thus shows how a supportive relationship is important across different time points in a parents' journey towards improving their child's sleep, from recognising the sleep problem to assessing and treating it. This is therefore proposed as another issue for stakeholder discussion in the co-design study.

4.14.4. Theme four-Parents and sleep practitioners improve their understanding of the sleep problem.

Exploratory study findings demonstrated a limited application of current sleep assessment evidence identified in the scoping review. The parent sub-theme 'Understanding sleep problems' revealed how most parents did not receive a formal sleep assessment, and those who did felt sleep practitioners failed to achieve a comprehensive sleep problem understanding. Furthermore, these parents reported varying levels of support to complete sleep assesment tools, revealing another layer of inequitable provision across sleep consultation teams. A wide spectrum of sleep assessment approaches were also reported in the sleep practitioner sub-theme 'Nature of assessment' which revealed inconsistencies in tools used, support given to parents and sleep practitioner competence to interpret assessment findings. Furthermore, assessment tools used by teams often included in-house sleep history questionnaires rather than examples cited in the review and only two teams utilised a validated sleep outcome measure (CSHQ). This is significant in demonstrating inequitable SHE provision in terms of sleep assessment, which could be better informed by current evidence. Also, improving how sleep practitioners interpret assessment findings and drawing on best practice examples around psychological formulation techniques is worthy of further debate. In summary, issues around improving stakeholder understanding of children's sleep problems is proposed as another theme for co-design study discussion.

4.14.5. Theme five- Regularity and quality of child's sleep improves.

Exploratory study findings revealed mixed evidence of how parents and sleep practitioners achieved improvements in children's sleep problems. The parent sub-

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theme '"*You just tick off the boxes*"; the need for customised support' reflected how at some point, all parents had been offered broad based SHE with no prior sleep assessment and a minority (n=3) had also received SHE tailored to their child's individual needs. Parents reported how they largely under-valued standardised SHE advice and considered it inadequate in meeting the specific needs of children with DD. Furthermore, the parent sub-theme 'Ways of coping' demonstrated how parents had intiutively already tried some commonly known SHE techniques with limited success, which contributed to their views around the simplicity of this advice. Some satisfaction and effectiveness was reported by the three parents who had received tailored SHE advice following a sleep assessment; an approach which sleep practitioners agreed was best practice in the sleep practitioner sub-theme 'Nature of treatment'. This notion of broad based versus customised SHE advice was largely un-explored in the scoping review and represented a new strand of evidence to inform understandings of SHE.

Exploratory study findings built on the limited evidence reported in the scoping review around methods and approaches to SHE delivery. In the ' *"At home I think you can just relax more"*; the importance of place' parent sub-theme, the role of a supportive setting in the successful implementation of SHE advice was described and parents reported a clear preference for home visits. Rich insights were also reported by one parent who implied the importance of actively offering the opportunity to bring peer supporters to all appointments. Parents' need for moral support, to alleviate any perceived imbalance of power, was also evidenced in the way three parents chose to use peer supporters during exploratory study interviews. Further helpful processes which could enhance the delivery of SHE and improve children's sleep problems, were identified in the sleep practitioner sub-theme

'Nature of treatment' such as supporting parents to implement SHE advice, 'psycho-education' and parent buddy systems. It is proposed to further explore in the co-design study, how all these actions work together with the actual SHE advice components identified in the review to improve the regularity and quality of children's sleep.

In the parent sub-theme '"*Their answer was medication and I refused*"; disappointment with medication' described how most families were dissatisfied with being offered melatonin (often without a prior sleep assessment). Although two parents believed melatonin had been ultimately effective, they described mixed reflections on the acceptability of this approach. Sleep practitioners also reported in the sub-theme 'Nature of treatment' that melatonin was commonly applied concurrently with SHE, sometimes inappropriately and could often interfere with the effectiveness of SHE. It is therefore important to also acknowledge how SHE is commonly implemented as part of a wider package of care which includes medication when considering all the factors which affect how children's sleep can be helped to improve.

4.14.6. Theme six - Quality of life improves for the family.

Scoping review findings identified various QOL outcome measures used in evaluative studies, implying the potential of improved children's sleep to postively affect other areas of family life. Limited evidence was provided in the exploratory study regarding the broader effects of enhanced children's sleep. In the 'Nature of assessment' sleep practitioner sub-theme, sleep practitioners from two sleep consultation team described using QOL outcome measures identified in the review (PSI, PCQ and GHQ) but only one team reported re-running them at the conclusion

of an intervention. In-house evaluation questionaires were used by some sleep practitioners when their involvement with families ceased, to guage how improved sleep had affected family life. In the parent sub-themes '"*The sleep problems compound everything else";* broader impacts' and '"*It was a battle";* the challenges of sleep problems', parents described the negative effects of persistent children's sleep problems on the child and family. The implication is that if sleep difficulties were to improve, the negative impacts of sleep deprivation would also subside. In summary, improving the QOL of the family is a possible bi-product of SHE intervention success, which measures tested in scientific studies aim to quantify. Further discussion is therefore needed in the co-design study to explore how stakeholders perceive the broader impacts of improved children's sleep.

All six themes were then utilised to inform the development of the subsequent co-design study presented in the next chapter.

CHAPTER 5 – CO-DESIGN STUDY.

5.1. Introduction

This chapter describes the methods and findings from the co-design study which aimed to co-create with parents and sleep practitioners, a SHE tool underpinned by programme theory. A detailed description of the methods employed in this study will firstly be presented. In summary, the study consisted of three separate events involving the discussion of six overarching areas (developed from scoping review findings and exploratory study themes) and presenting a parent podcast to co-designers. Stakeholders were asked to reflect on the emotional impact of this information, voice challenge or confirmation, and identify any new priorities for the way SHE should be delivered and parents supported with children's sleep problems. These stakeholder constructions are presented in this chapter and early analytical links made with mid-range (research-based) and novel theories. Co-design events also involved debating the SHE advice components identified in the review, and this chapter concludes with a presentation of a co-designed SHE tool for children with DD.

5.2. Research methods

5.2.1. Study aims.

The overall aim of the co-design study was to synthesise the findings of the scoping review, exploratory and co-design studies, and develop a SHE tool underpinned by programme theory explaining why SHE is supposed to work to improve sleep; this was couched in lay-man's terms for stakeholder participants (Table 19). These were:

- To gather views on how sleep practitioners and parents should work together to ensure families receive effective sleep advice and support.

- To focus on what a parent's journey towards receiving professionally supported sleep advice should look like.

- To understand what makes effective sleep advice and support work.

- To think systematically about what sleep practitioners need to do when they advise and support families with sleep problems.

- To explore participants' views on the acceptability of sleep advice content.

Table 19 Aims of the co-design events.

1. To gather views on how sleep practitioners and parents should work together to ensure families of children with DD receive effective sleep advice# and support with sleep problems.
2. To focus on what a parent's journey towards receiving professionally supported sleep advice should look like.
3. To really understand what makes effective sleep advice and support work.
4. To think systematically about what sleep practitioners need to do when they advise and support families with sleep problems.
5. To explore parent/sleep practitioners' views on the acceptability of sleep advice content.

The term SHE was not used with study participants. Instead 'sleep advice and support' was used to ensure lay understanding.

5.2.2. The co-design study.

The co-design study involved a parent only co-design event followed by two joint parent and sleep practitioner events.These events began with the six overarching themes developed at the conclusion of the Exploratory study (Table 18). These required deeper deliberation by stakeholders and were presented at events as discussion areas. They were used as a flexible rather than constraining guide for debate, in line with a participative inquiry approach. 'Parents and practitioners have

a shared understanding of what a sleep problem is focused on the significant pre-implementation challenges around stakeholders agreeing on the existence of sleep problems. 'Sleep services are well publicised and accessible for parents' illuminated pre-implementation issues around parents' awareness of and ability to access sleep consultation teams. 'Parents and sleep practitioners develop a safe and supportive relationship' highlighted the need to build supportive parent/sleep practitioner working relationships. 'Parents and sleep practitioners impove their understanding of the sleep problem' focused on issues around improving stakeholder understanding of children's sleep problems. 'Regularity and quality of child's sleep improves' highlighted how parents and sleep practitioners achieved improvements in the child's sleep. The final discussion theme: 'Quality of life improves for the family' focused on the potential for improved sleep to positively affect other areas of family life.

5.2.3. Ethical standards.

Discussions with the supervisory team concluded that the co-design study, which used the principles of patient and public stakeholder involvement to develop SHE programme theory, should be framed as a service evaluation for the purposes of ethics. Ethical approval was gained for study from the Healthcare and Medical Sciences Academic Committee, Bangor University on 4[h] March 2015 (Appendix 6).

On the basis that the co-design work was not classed as research by the supervisory team, confirmation was sought and received by the Health Research Authority on the 10th April 2015 that Research Ethics Committee (REC) approval was not required. Following this, permission was granted by the Sponsor to proceed without Research and Development (R&D) approvals (Appendix 6).

School ethical approval for the major amendment to the study in terms of a change in duration of co-design events from three to two hours was received on the 24th April 2015 and appropriate amendments made to participant information forms.

On the basis the co-design work was classed as a service evaluation, advice was taken to gain permission from the clinical effectiveness department at the local health board to involve local NHS staff in co-design events. A written response was received from the local health board on the 22nd June 2015 confirming they had decided not to participate in the co-design study (Appendix 6). Following receipt of this letter, the researcher met with a member of the health board to discuss their decision and it was felt they were mistrustful of and reluctant to engage with the co-design method. Whilst it was regrettable that the local health board had taken this decision and health staff was prevented from taking part, it was felt that the views of recruited social care and voluntary organisation sleep practitioners gave an adequate representation of how SHE was currently delivered in practice.

5.2.4. Parent participant sampling procedure.

All parents who had participated in the exploratory study and resided in North Wales were invited to continue their involvement in the project and take part in the co-design events, however only one parent, Gloria (pseudonym) was able to make the commitment. Therefore, the chain sampling method (Patton, 2015) was employed again whereby well situated people were contacted for names of potential parents. Original contacts with the Handsel Project, Down's Syndrome Wales, Cerebra and Children in Wales were re-contacted to assist with parent recruitment; however, they were unable to provide details of any potential participants.

During this recruitment process, feedback was received from some parents that the proposed three hour length of co-design events was prohibitive. Therefore, to encourage parent participation, the proposed duration of events was shortened to two hours which parents who were eventually recruited found acceptable. This was also considered a substantial amendment to the study design which required the appropriate ethics committees to be notified and participant information sheets to be changed accordingly.

The researcher then embarked on a six week recruitment process where contacts were made with 39 disability specific voluntary organisations. As Coyne (1997) advises, in order to collate a purposive sample in a strategic way, the researcher needs to know where to start looking for well-situated contacts who can provide names of prospective participants. One contact from Conwy and Denbighshire National Autistic Society (CDNAS) was able to provide names of two appropriate parents: Fadheela and Caroline (pseudonyms). However, none of the other key professionals identified were able to provide names directly. Therefore, a more pragmatic approach was taken and permission was sought from key professionals to attend forthcoming parent events such as parent support groups. Eight parent events were attended by the researcher in the six week recruitment period and a further 18 prospective participants were recruited as shown in Table 20.

Table 20 Co-design study: Parent purposive sample.

No	Parent code	Peer supporter used	Child Demographics	Recruited via.	Participation: co-design event 1 ,2, 3. None due to Family commitments FC Work commitments WC
1	Gloria	None	9 year old boy with learning disabilities	Participation in exploratory study	1, 3.
2	Jenny	None	14 year old boy with ASD	Contact a Family event	1,2,3
3	Sharon	Sister (event 1)	7 year old boy with ASD	Create a Smile parent support group	1, 2
4	Emily	None	12 year old boy with ASD	Riding for the Disabled event	1,3
5	Sophia	None	7 year old boy with ASD	Contact a Family event	1
6	Fadheela	None	8 year old boy with ASD	Introduced by Conwy and Denbighshire National Autistic Society (CDNAS)	1
7	Amy	None	16 year old boy with ASD	Introduced by parent participant :Jenny	1,2,3
8	Clarissa	None	14 year old boy with ASD	Afasic Cymru event	3
9	Faith	N/A	Learning disabilities	Llanwrst Family Centre	None. FC.
10	Caroline	N/A	ASD	Introduced by CDNAS	None. WC
11	Chloe	N/A	ASD	Gwynedd and Anglesey National Autistic Society (GANAS) parent support group	None. WC
12	Rhona	N/A	ASD	GANAS parent support group	None. WC
13	Helen	N/A	ASD	GANAS parent support group	None. WC
14	Shelley	N/A	ASD	GANAS parent support group	None. WC and FC
15	Candice	N/A	ASD	CDNAS internet café	None. WC
16	Angela	N/A	Learning disabilities	Mencap event	None. WC
18	Sonia	N/A	Learning disabilities	Mencap event	None. WC.

19	Anna	N/A	Learning disabilities	Mencap event	None. WC
20	Kala	N/A	Learning disabilities	NISCHR Involving People	None. FC
21	Cilla	N/A	ASD	Introduced by parent participant :Fadheela	None. WC

All prospective parents were sent or emailed an appropriate participant information sheet and invitation letter (Appendix 7), contact form (Appendix 8) and consent form (Appendix 9). All participant documentation was translated into the Welsh language by Bangor University's Translation service. Completed consent forms were either emailed to the researcher in advance or completed on the day of co-design events, after participants had had time to read the participant information forms. Participants were offered the opportunity to discuss any concerns or questions with the researcher before signing the consent form. This demonstrated how best practice standards set by the Research Governance framework (WAG, 2009) were adhered to.

5.2.5. Inclusion and exclusion criteria for parents.

The inclusion and exclusion criteria for parent participants in the co-design study mirrored the criteria stipulated earlier in the exploratory study (section 4.2.5).

5.2.6. Parent characteristics.

Each parent was assigned a participant code name to maintain confidentiality that was used during the data analysis process (Table 20). All parents lived in Wales, were first language English or non-Welsh speakers. Participants' children had either a Learning Disability or ASD diagnosis. All parents who participated were mothers; fathers were not excluded from the study but none came forward to take part.

5.2.7. Sleep teams and sleep practitioner participants sampling procedure.

Sleep practitioners were recruited by inviting all 16 sleep practitioners from the exploratory study; five of whom (SFO, SCP, VDO, VDW and VSS) agreed to continue their involvement in the project. The remaining 11 sleep practitioners were unable to take part due to work commitments or not having permission in the case of local NHS sleep practitioners (5.2.3). Three new sleep practitioners (VSP (2), VFW (2) and VSS (3)) were recruited via introduction from existing sleep practitioner participants which brought the purposive sample to a total of eight (Table 21).

Table 21 Co-design study-sleep practitioner purposive sample.

No	Title.	Code.	Team code	Recruited via:	Co-design event 2/3. None
1. 2.	Family Support Officer. Clinical Psychologist.	SFSO. SCP.	} Social care team 1-ST1 (Local county)	Exploratory study purposive sample	2 & 3 None
3.	Sleep Practitioner.	VSP(2)	Voluntary organisation team 1-VS1 (Cross county)	Introduced by VSP	2 & 3
4.	Development Officer.	VDO.	Voluntary organisation team 3-VS3 (Cross county)	Exploratory study purposive sample	2
5.	Development Officer	VFW	Voluntary organisation team 5- VS5 (Cross county)	Exploratory study purposive sample	3

6.	Development Officer	VFW(2)	Voluntary organisation team 5- VS5 (Out of county)	Introduced by VFW	2
7.	Development Officer and Sleep Practitioner	VSS(3)	Voluntary organisation team 6 –VS6 (Cross county)	Introduced by VSS	3
8.	Development Officer and Sleep Practitioner	VSS	Voluntary organisation team 6 –VS6 (Cross county)	Exploratory study purposive sample	None

All prospective participants were sent or emailed a Welsh or English participant information sheet and invitation letter (Appendix 7), contact form (Appendix 8) and consent form (Appendix 9). Completed consent forms were either emailed to the researcher in advance or completed on the day of co-design events after participants had had time to read the participant information forms. Participants were offered the opportunity to discuss any concerns or questions with the researcher before signing the consent form. This demonstrated how best practice standards set by the Research Governance framework (WAG, 2009) were adhered to.

5.2.8. Sleep team characteristics.

The sleep practitioner sample was recruited from five sleep teams which represented one social care organisation and four voluntary organisations in Wales (Table 21). The sleep teams represented were given a team code to protect confidentiality .The social care team (ST1) represented a local county, and voluntary organisation teams VS1, VS3, VS5 and VS6 operated on a cross county level. Teams represented delivered a range of sleep service provision. Two teams: ST1 and VS1, had a direct role assessing children's sleep problems and designing SHE interventions, and

specialised solely in sleep. Whereas teams: VS3, VS5, VS6 were sign-posting teams, who gave ad-hoc SHE advice as part of a broader range of services.

5.2.9. Inclusion/exclusion criteria for sleep practitioners.

The inclusion and exclusion criteria for sleep practitioner participants in the co-design study mirrored the criteria stipulated earlier in the exploratory study (section 4.2.13).

5.2.10. Sleep practitioner participant characteristics.

Sleep practitioners held a range of job titles including Family Support Officer (n=1) and Development Officers (n=6). Each sleep practitioner participant was assigned a participant code name to maintain anonymity that was utilised during the data analysis process (Table 21). The majority of sleep practitioners were female, however, participant VFW (2) was male. Only one participant: SFSO was Welsh speaking however chose to participate through the medium of English.

5.2.11 Data collection- co-design events.

Three co-design groups were organised: one parent only event and two joint parent/sleep practitioner events with the intention of melding stakeholder perspectives on sleep problems and SHE. Events were conducted in English as the researcher was a non-Welsh speaker and participants were offered separate interviews through the medium of Welsh if they preferred.

Preparation involved development of an audio 'trigger' podcast to represent the exploratory study parent themes and stimulate participant discussion which would be presented at the first two events. Illuminating voice extracts representing parent

themes were selected and synthesised into a 15 minute podcast (Refer Appendix 10

for 5 minute podcast clip). Upon the advice of a Sound Technician from Bangor

University, an Audacity software package was utilised, which entailed converting

audio-recorded files to Waveform Audio Format (WAV), then selecting and editing

appropriate audio extracts. First supervisor Dr Jaci Huws checked the final podcast

for sound quality and appropriate removal of participant identifiers prior to use in the

co-design groups. Consent from parents to produce the podcast had already been

given via their agreement to information being used for future educational purposes

(see exploratory study consent form- Appendix 9).The podcast was introduced to

stakeholders in the co-design events with a preparatory script informed by a recent

EBCD study by Locock et al. (2014) (refer Table 22).

Table 22 'Trigger' podcast script.

This short podcast has been put together from analysis of interviews with 11 parents in Wales who have children with developmental disabilities and sleep problems and experiences of professional sleep support.
You will hear some negative views because these can be helpful for learning about when things go wrong and what could have been done to make it a better experience. There are also examples of positive experiences which we feel are important to share and build upon in other sleep teams. We hope the podcast resonates with some of your individual experiences and will help trigger some thoughts for the next session.
N.B: Silence has been inserted in this podcast to conceal any identifiers: names, organisations in the interests of confidentiality.

5.2.12. Digital recording, video recording and transcription.

All co-design events were digitally recorded to facilitate transcription, coding of

transcripts and appropriate selection of anonymised verbatim for quotation within the

research write up. Events were also filmed using video recording equipment in order

to distinguish conversation between participants and to aid data analysis. Recordings

of whole group discussions were transcribed verbatim by the researcher, but small group exercise work was summarised due to difficulties with audible clarity of voices where multiple participants spoke concurrently.

5.2.13. Safeguarding children and young people.

The arrangements made in respect of safeguarding for the co-design study replicated those made in the exploratory study (section 4.2.9.).

5.2.14. Data protection.

The arrangements made in respect of data protection for the co-design study replicated those made in the exploratory study (section 4.2.10).

5.2.15. First co-design event (parent only).

Seven parents attended the first event which was held on a weekday at a voluntary organisation meeting room on the 22 June 2015 between 11am and 1pm. The room was welcoming for parents, in a central location and was free of charge to the researcher. A fellow Bangor University research student co-facilitated the event, took consent from participants and managed any housekeeping issues.The date and time for the event was arranged by logging parents' availability on an Excel spreadsheet, alongside researcher, co-facilitator and venue availability.

The group began with a statement of workshop aims (Table 18). Then following a warm up activity, participants were shown the discussion themes (Table 19) and invited to confirm, challenge or add to them. They were asked to discuss what actions needed to happen on a parent, professional, organisational and policy level to achieve positive change in the way families were supported with sleep problems.

Discussion was facilitated by splitting the parents into two groups and providing additional ideas around process (identified from data analysis to date) where necessary to stimulate dialogue.

Parents were then invited to listen to the 'trigger' podcast, asked to reflect on what they had heard and if there were any new insights, which were recorded visually on a flipchart. The event concluded by preparing parents for the next co-design event alongside sleep practitioners, advising it would follow a similar format and include listening to the podcast. Any concerns were discussed so parents felt comfortable to actively participate alongside sleep practitioners as equal partners.

5.2.16. Analysis of first co-design event.

Parent only co-design event video and audio recordings were transcribed verbatim and data were thematically analysed (Braun & Clarke, 2006) in relation to the six discussion themes. Data were coded to identify repeated debates of topics such as sleep screening or raising awareness of sleep problems within each discussion area. Parents' comments which indicated agreement or disagreement with review and exploratory study findings were logged. In addition, any new parent constructions or priorities for sleep service improvement which needed further stakeholder confirmation were recorded separately.

In preparation for the second event, a visual representation of the six discussion themes and emerging constructions from the first co-design event was prepared using a wall-sized model, based on an adapted version of the person centred planning tool: the Planning Alternative Futures with Hope 'PATH' (Pearpoint, O'Brien & Forest, 1998) (refer Figure 14). This is a planning style used with organisations and

individuals which helps people to understand complex situations and take control over change (Sanderson & Lewis, 2012) and it was felt this mirrored a participatory approach.

The overarching ethos of the PATH model is to provide a central visual focus for participants to engage with and plan for change (Figure 14). The process begins with step 1- The North Star which requires participants to dream and record how they would like life to be different. Step 2- Our Sense of the Goal invites participants to imagine that they have achieved their final goals and describe how this feels. Focusing on the ultimate outcomes first, energises the group and ensures the event begins with a positive mindset and clear shared vision. At this point the PATH model was adapted for the purposes of this study (Table 23). To ensure adequate time to discuss all six discussion themes, these two initial steps were combined and reflected a broad discussion of the ultimate outcomes. Stakeholders were asked to visualise what improved sleep looked like, and their collated responses represented their constructions around 'Quality of life improves for the child and family'. The next step –Now, brings participants back to the present and requires them to examine what life is like now. This creates a tension between the existing problem and ulitimate goals, and motivates the group to plan for change. In the co-design study, this step was replaced with a discussion of discussion theme 1- 'Parents and practitioners have a shared understanding of what a sleep problem is' which reflected the start of a family's journey towards improved sleep (Table 23). The remaining steps (Who to enrol, How to grow stronger, Next steps and Strategies) gradually tease out strategies for change which lead the group closer to the 'North Star' or ultimate outcomes. The PATH model was further adapted here and the remaining four columns were headed with discussion themes two to five, as shown in Table 23

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and sequentially deliberated by the group. So whilst some of the distinct steps of the PATH model were omitted, the overarching ethos of the PATH was maintained. The researcher was confident this adapted tool could assist co-designers in planning for change in a cohesive way, based on their experience of the PATH model in previous person centred planning work.

Figure 14 PATH model (Pearpoint, O'Brien & Forest, 1998)
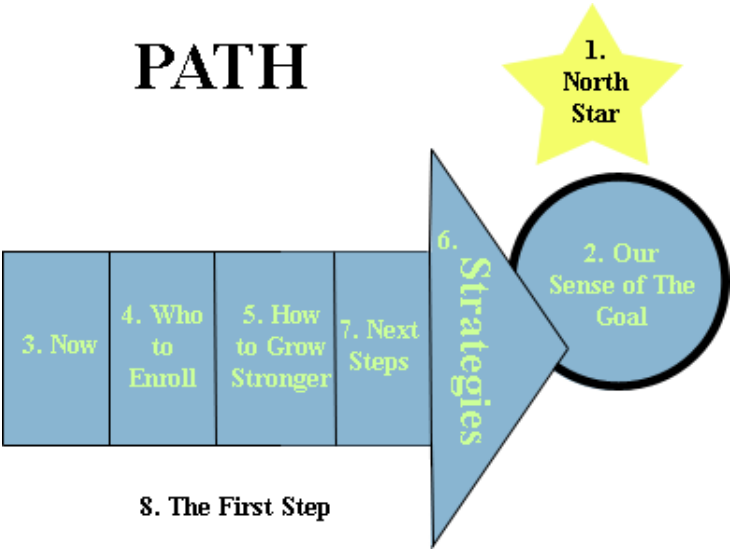
Table 23. Key to Figure 14: Adaptation of the PATH model and columns for the co-design study.

North star & Our sense of the goal- replaced with 'Quality of life improves for the child and family' outcome.

Now- replaced with 'Parents and practitioners have a shared understanding of what a sleep problem is' outcome.

Who to enrol- replaced with 'Sleep services are well publicised and accessible for parents' outcome.

How to grow stronger- replaced with 'Parents and practitioners develop a safe and supportive relationship' outcome.

Next steps- replaced with 'Parents and practitioners improve their understanding of the sleep problem' outcome.

Strategies- replaced with 'Regularity and quality of child's sleep improves' outcome.

Figure 15 shows the PATH model that was developed for the second co-design event. New parent constructions were listed in blue ink and exploratory study sleep practitioner constructions which required further confirmation were listed in red ink. This helped to provide a central focus for all stakeholders and a means of visually representing developing stakeholder constructions to support the co-design process.

Figure 15 PATH model representing developing stakeholder constructions.

5.2.17. Second co-design event (parent and sleep practitioner).

This was held on the 22 June 2015, 11am-1pm at a community centre, four weeks after the parent only event to allow for transcription and preliminary data analysis. The venue had a central location, was parent friendly and reasonable hire cost of £10 per hour. The event was attended by three parents, four sleep practitioners (refer Tables 20 and 21) and co-facilitated by a NISCHR staff member, and organised using a combination of Doodle planning systems, emails and excel spreadsheets.

The event commenced with a re-statement of co-design event aims (Table 18) then co-designers were introduced to the partly completed PATH model (Figure 15). As a warm up activity participants were asked to consider the end goals of SHE first; 'Regularity and quality of child's sleep improves' and 'Quality of life improves for the family'. The question was posed "If you were going to deliver sleep advice or receive it, how would you know it has been successful?" and co-designers were asked to describe on post-it notes how success would make them feel, which were positioned next to the 'North Star' on the PATH model. This also provided additional data to help confirm the success criteria for the ultimate outcomes of SHE.

In two separate sessions the group was played the first and second half of the audio podcast. The researcher reflected from the first event that the total 15 minute play time was too long for participants to listen passively and not actively engage. Following each seven minute broadcast participants were asked to consider their personal reflections in relation to each of the identified stakeholder issues. They were asked if there was anything that could be done differently to achieve change and responses were documented on PATH model. Levels of agreement or disagreement within the group were also documented.

5.2.18. Analysis of second co-design event.

Second co-design event video and audio recordings were transcribed verbatim and data were thematically analysed in relation to the six discussion themes. Data were coded to identify repeated debates of topics such as sleep screening or raising awareness of sleep problems within each discussion area. Stakeholders' comments which indicated agreement or disagreement with review, exploratory study and 1st co-design event findings were logged. In addition, any new stakeholder constructions or priorities for sleep service improvement which needed further stakeholder questioning and exploration were highlighted on the PATH model, ready for discussion in the final co-design event.

5.2.19. Third co-design event (parent and sleep practitioner).

This event was held on the 16th July 2015, 11am-1pm at the same community centre as the second event, four weeks after to allow for transcription and preliminary data analysis. The event was attended by five parents, four sleep practitioners (Tables 20 and 21), co-facilitated by a NISCHR professional, and organised using a combination of Doodle planning systems, emails and excel spreadsheets.

The event commenced with a warm up activity and stakeholders were again referred to the developing PATH model. Co-designers were asked to discuss the remaining discussion themes not already covered in the second event and revisit processes identified for further clarification. Participants were then presented with the six SHE advice areas identified from the review: Sleep timing, bedtime routines, communication, environment, behaviour, physiology and their individual components which were displayed on wall charts around the room. Participants were asked in groups, to select specific SHE components they considered would be

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challenging for parents/sleep practitioners to achieve/implement. The aim was not to discuss every single SHE advice point; only the advice points participants found challenging. As a whole group, participants then reported on their views regarding the acceptability of SHE advice and how implementation could be supported. This concluded the event and participants were thanked for their involvement.

5.2.20. Analysis of the third co-design event.

Third co-design event video and audio recordings were transcribed verbatim and data were thematically analysed in relation to the six discussion themes. Data were coded to identify repeated debates of topics such as sleep screening or raising awareness of sleep problems within each discussion area. Stakeholders' comments which indicated agreement or disagreement with review and exploratory study, 1st and second co-design event findings were recorded in addition to any new constructions or priorities for sleep service improvement.

Findings from the review and co-design studies were collated to produce a SHE tool for children with DD. Findings from all three studies were then synthesised in relation to the six discussion areas and early analytical correlations made with novel and mid-range (research based) theories of change. This synthesis then informed the systematic development of a programme theory underpinning SHE for children with DD and explanatory logic model.

5.3. Presentation of Stakeholder Constructions Around How SHE Should Be Delievered.

Figure 16 shows the completed visual representation (PATH model) of stakeholder constructions around how SHE should be delivered, at the conclusion of the three co-

design events. Their constructions are summarised in Table 24 and in the following

supporting narrative, which feed into a final synthesis of cumulative study findings

and development of programme theory.

Figure 16. PATH model showing findings from co-design events.

Table 24. Overview of stakeholder constructions of how SHE should be delivered.

Discussion themes	Agreement and disagreement with review and exploratory study findings	New stakeholder insights
1- Parents and practitioners have a shared understanding of what a sleep problem is.	Parents and practitioners need to recognise sleep problems	Parents can be afraid to recognise their child has a sleep problem
		Parents may not give priority to sleep against child's other multiple needs and fail to recognise sleep problems
		Parent partners and relatives need to agree and recognise there is a sleep problem
		Generic, specialist and early years practitioners should screen all children for sleep problems (*agreement & disagreement*)
		Practitioners should ask parents about sleep in a non-judgemental way
		Parents should be empowered to screen for sleep problems using the personal child health record (PCHR) or red book. (*agreement & disagreement*)
		Public awareness raising campaigns are needed around sleep
		Parents are shy to socialise at pre-school groups where they may have their awareness raised
		Parents may have literacy /communication problems preventing them from utilising screening or awareness raising initiatives
2-Sleep services are well publicised and accessible for parents.	Adequate provision of sleep services	Parents may have literacy /communication problems preventing them from engaging with sleep service publicity
	Sleep teams should be easily accessible for parents	Parents may be afraid of accessing specialist sleep teams.
	Improved publicising of sleep services	Provision of a generic sleep team (*agreement & disagreement*)
3 Parents and sleep practitioners develop a safe and supportive relationship.	Practitioners should be empathic and positive with parents	Continuity of sleep practitioner support
	Parents receive the support they need throughout their contact with sleep services	
4 Parents and sleep practitioners improve their understanding of the sleep problem	Sleep practitioners need to complete a comprehensive sleep assessment	Comprehensive assessment should involve sleep practitioners observing children at bedtimes
	Sleep practitioners need to complete a competent assessment	Parents may not give assessment information honestly

		Comprehensive assessment should involve other members of the multi-disciplinary team and partner organisations
5 Regularity and quality of child's sleep improves	Parents are supported to follow sleep advice *(agreement & disagreement)*	Sleep practitioners need to praise parents for achievements throughout their contact
	Parents receive effective sleep advice	Sleep practitioners need to liaise with others to follow sleep advice consistently across all settings
6 Quality of life improves for the family	Parents score favourably on QOL measures and evaluation questionnaires.	Descriptions of what improved quality of life looks like
		Parents feel supported to maintain progress with help from parent support groups.

5.4. Discussion Theme One- Parents and Practitioners Have a Shared Understanding of What a Sleep Problem Is.

5.4.1. Agreement/disagreement with review and exploratory study findings.

Practitioners and parents need to recognise sleep problems.

Agreement was found with exploratory study findings (4.6.2) and (4.11.1) which identified that sleep practitioners and parents both had a role to play in recognising sleep problems. Parent and sleep practitioner co-designers agreed that generic practitioners and sleep practitioners needed to attend to parents' worries about children's sleep and recognise when there was a problem. Jenny's and Sophia's narratives demonstrate perceptions of how professionals sometimes seemed unsure of discussing children's sleep or gave the impression of being disinterested.

> "*With the GPs it was 'it's just the way he was' and I don't like that attitude it's just the way he was*" (Jenny)

> "*GP wasn't interested. If they don't even give you any understanding you go away feeling "oh I shouldn't have come"*". (Sophia)

Furthermore, VSP(2) and SFSO acknowledged that generic practitioners often took an un-helpful 'wait and see' approach, which Jenny and Amy believed to be based on a low expectation of children with DD in terms of sleep. Parents argued that this belief that sleep problems were an inevitable part of DD conditions needed to be challenged. Stakeholder discussions therefore seemed to confirm the need for better

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training of generic and sleep practitioners in recognising sleep problems in this group of children.

> VSP(2): "*So it's getting the professionals then to get them to understand as well that maybe sometimes; the 'You're child will grow out of it' will not ever happen* "

> Jenny: "*I think that is part of the problem, that the not sleeping is just part of their condition, no its not!*"

> Amy: "*Yes!*"

This finding indicates problems with the legitimation of sleep problems in children with DD, whereby they are not valued or labelled as an important issue by generic practitioners. The need to provide legitimacy to sleep problems and make it standard practice to recognise and prioritise them seems an emerging priority for change. It is also posited that explanatory links can be drawn with generic practitioners' low health expectation for children with DD and belief that sleep problems are unlikely to improve.

It was also agreed that parents could hinder the process of recognising sleep problems, when they accepted their child's sleep as normal and continued to cope (which links with exploratory study findings- 4.6.2). Parents spoke about how they 'normalised' their children's sleep and adapted to their situation which prevented them from questioning or trying to understand it. This could be a particular risk if this was their only child and they did not have older siblings to compare sleep development with. Why parents chose to play down children's sleep problems could

be explained by their desire to not feel any different from other parents. This can be linked to normalisation theory (Wolfensberger & Tullman, 1982) which explains differentness as negative and sameness as positive. Explanatory links can also be made with parents' reluctance to legitimise sleep problems and general low health expectation that their child's sleep is likely to improve. The following narrative from Fadheela and Gloria demonstrates this construction.

> Fadheela: "*You might not know you've got a problem because it becomes normal*"
>
> Gloria: "*Especially if it's your first one*"

Three sleep practitioners and two parents also agreed with exploratory study findings (4.11.2) that sleep practitioners should openly discuss with parents what they consider to be a sleep problem at the start of their contact. This was to deliberate whether issues such as co-sleeping were perceived as problematic for parents or something sleep practitioners believed to be a problem. It was agreed that an open-minded conversation needed to happen when parents first engaged with sleep services to ensure sleep practitioners were clear about parent's expectations and beliefs about children's sleep. The following extract from SFSO evidences this construction:

> SFSO: *"The twelve year old is sleeping with the mum, but the social worker knows the family really well and said it's not a problem for the family, they're fine with it but the nurse has seen it as a problem (..) but what I said was well we can just have that conversation what*

> *do you want to happen? (..)And they can come back to me when they are ready if they want to change sleeping arrangements.*"

VSS(3) who was also a parent of a child with DD and historical sleep problems agreed with exploratory study findings (4.7.2), which showed that parents sometimes adopted coping strategies that masked underlying sleep problems and hindered their recognition. She described her experiences of using co-sleeping as a successful management strategy but her awareness this may not be the ideal, long term solution and her reluctance to discuss this with generic practitioners at the time. Therefore, it is important practitioners are aware of the contextual factor that parents who co-sleep with their children may be too embarrassed to discuss their children's sleep problems, resulting in them being overlooked. Explanatory links can be drawn here with the legitimation of sleep problems and a public belief that they are a personal issue for families. This evidences how parents need to feel comfortable talking with others about their experiences and beliefs, without fear of professional or wider societal judgement.

> "*My son slept with me until he was 10, because he was a terrible sleeper and he slept if he was with me (..) I wouldn't have wanted to tell people that probably at the time*"(VSS(3))

5.4.2. New stakeholder insights.

Parents can be afraid to recognise their child has a sleep problem.

Parents explained the emotional challenges of coming to terms with children's sleep problems and accepting that help was needed. When they perceived it was unusual for children to struggle with sleep, they felt fearful of openly admitting there was a problem. This could precipitate a level of denial and tendency for parents to avoid discussing the issue. Being afraid to recognise sleep problems was therefore identified as a pre-implementation barrier to engaging with sleep services and receiving sleep interventions, which is demonstrated in the following dialogue.

> Gloria: "*I feel that everybody else's child is doing something or the norm and yours isn't, you're loathed, you don't want to accept it do you?*"
>
> Amy: "*Exactly*"
>
> Gloria: "*Cause you don't want to accept that your child isn't doing what everyone else's is doing you want to live in your own little bubble*"
>
> Emily: "*I'm fine!*"

Fear of being different resonates with normalisation theory in terms of parents viewing difference as undesirable and something to be avoided. Explanatory links can also be made with the stages of change theory or transtheoretical model (TTM) (Prochasksa & DiClimente, 1983; Prochasksa & Velicer, 1997). This mid-range

theory holds that change happens over a period of time and can include a pre-stage of contemplation where people deliberate the consequences and impact of changing their behaviour.

Amy also described how she believed parents commonly lost confidence in their beliefs about sleep problems if, for example, health, social care and education professionals felt their child was not showing signs of sleep deprivation at school. In these situations parents felt judged, lost confidence and could fail to continue raising concerns. Sharon and VSP(2) indicated that they agreed with Amy's view, further evidencing the impact this contextual factor has on the early recognition of children's sleep problems. This shows how parent's belief in their ability to recognise sleep problems is important and can be affected by the responses and attitudes of professionals.

> Amy: "*A professional goes in and sees them in school-" oh they are alright in school" and you think flippin eck, hang on it's all pointing back to the parent and then you lose confidence then don't you?"*
>
> Sharon and VSP(2): *Nod in agreement*
>
> Amy: "*And you know and you can be doing your absolute hardest and researching everything but if they are still not sleeping and that's happening at home and "but they are fine at school" it's very disempowering.*"

VSS(3) and Gloria and Clarissa also discussed the impact of others' attitudes on parents' confidence to recognise sleep problems. They confirmed how parents often

felt judged by professionals or peers and embarrassed to admit they were struggling with their child's sleep. Clarissa described how she had had a negative reaction from her child's head teacher who apportioned blame on her for her son's sleep problems. Gloria also referred to other parents passing judgemental comments about sleep on social media. Therefore, admitting children have sleep problems could be very difficult in such a climate of negativity. This illustrates why some children's sleep problems may go unrecognised for some time and is a contextual factor that generic and sleep practitioners need to be aware of when supporting parents at the very start of their SHE journey. Explanatory links can be made with the legitimation of sleep problems and societal belief that children's sleep problems are an exclusively private, parenting issue; not the responsibility of others outside the family.

> Clarissa: "*I had the head teacher on the phone saying I don't know how you let your son get like this, yer you are a dreadful mother (..) admitting your child's got a problem, is fine but its other parent's perception of that you know*
> Gloria: *"They have various things on Face-book don't they so someone puts on my child's like this and it's like wow .Why you doing that?"*
> VSS(3) and Clarissa nod in agreement.

Parents may not give sleep problems priority against multiple health needs and fail to recognise sleep problems.

Parents expounded how it could be challenging to recognise sleep problems when children had multiple complex needs around health, behaviour or education. During appointments with generic practitioners it was difficult to apportion adequate time to

sleep which meant problems remained unaddressed or did not receive the early intervention that was needed. The following dialogue between Fadheela and Emily demonstrates this.

> Fadheela: *"It's not always going to be the first thing on your mind when you are trying to battle for lots of other things"*
> Emily: "*Exactly*"
> Fadheela: *"So actually whilst it might be a really big problem that's contributing to all the other difficulties that they've got, when you're there you're just trying to get what you need at that time."*

Emily expanded on this point and described how she often grappled with the prioritisation of sleep against other complex needs. Although sleep was a problem for her son, other issues took priority and there was seldom time to discuss it during appointments. This demonstrates a significant contextual factor which could affect how sleep problems achieve recognition (which sleep practitioner co-designers appeared largely unaware of). This links with ideas around the legitimation of sleep problems and how they are undervalued by society. Parents may consider other health issues such as behaviour problems as more serious, owing to a lack of understanding about the broader negative impacts of sleep deprivation, which may explain why parents were disinclined to justify time spent addressing them.

> "*I'm trying to get across my problems with him at the moment, obviously sleep is one of them (..)Sleep's at the bottom of the list and of course times run out. And I've been there an hour and half and I'm only half way down the list. So it just doesn't come up." (*Emily)

Parent partners and relatives need to agree and recognise there is a sleep problem.

Jenny, Emily and VDO who was also a parent of a child with DD and historical sleep problems acknowledged how parents' partners and relatives had a role to play in recognising children's sleep problems. Sometimes other family members may have a stoic attitude towards sleep difficulties or may be too busy to realise there might be a problem. They described in the following narrative, how such reactions and beliefs could make it harder for parents to publically acknowledge their child's sleep problem.

> Jenny: *"I mean it took some convincing with my husband, you know that we had a problem in the first place because, in his generation in his parent's generation, it goes under the carpet you forget about it, it doesn't exist, well I'm sorry but it does."*
>
> VDO: " *I also think that I know speaking from personal experience that with my husband I went to see CAMHS and things like that I don't think he quite realised cause he worked such long hours he realised quite how bad it was for me."*(..)
>
> Emily: *"It's very hard though from my personal circumstance, my in-laws are very stoic as you put it they are just very no there is nothing wrong, just get to bed. Why aren't you sleeping ,rrrrrr, they are very, it's not, they don't see it as a problem, it's very difficult for me.*"

It is therefore suggested that the attitudes of wider family members and previous generations can affect parents confidence to admit children's sleep problems, and

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sleep practitioners need to be mindful, that parents may not always have the support of close family members when communicating sleep concerns. This is a contextual factor that could affect the process of recognising children's sleep problems.

Generic practitioners and sleep practitioners should screen all children for sleep problems (agreement and disagreement).

The new priority was identified of generic and sleep practitioners screening all children (typically developing and with DD) for sleep problems. Stakeholders explicated how if everyone was asked how their children slept as standard practice, children's sleep problems were less likely to be overlooked. This seemed to be a call for raising the value of sleep within broader society and again providing legitimacy to sleep problems. Introducing sleep screening as usual practice would also help parents to normalise and accept their children's sleep problems. It was important for parents not to feel different from others when they embarked on the process of accepting there might be a problem with the way their child slept. This maybe because parents viewed difference as undesirable or something that made them feel less accepted and aspired to sameness instead. This again links with the principles of normalisation theory which holds that difference triggers stigma and social exclusion (Gilbert, 2004). The following extracts demonstrate this construction:

> "*I think how you can be supported is whichever professional you go to meet asks about sleep. (..) It's about professionals actually asking that question, as routine.*" Fadheela

> "*It's got to be the norm(..) you don't feel like you are being singled out especially*" Gloria

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> VDO: "*I think the more ways of triggering those questions the better*"

> Sharon: *Nods*

> VDO: "*Then you get help quicker*"

VFW(2) and Jenny and Amy also suggested early years practitioners should take a more active role in sleep screening. They proposed that when children access nursery settings, early years' practitioners would be well placed to introduce the topic of sleep with children and parents. Suggestions were made around encouraging children to discuss sleep using a story board or taking a 'sleep diary teddy' home at night. In addition, a training need was highlighted amongst early years' workers so they were able to look out for signs of sleep problems to promote early identification. The active involvement of early years' practitioners in the sleep screening process represented a new priority for sleep service improvement.

> Jenny: "*Maybe the nursery school teachers can pick up on something.(..). They could do a little story board or something. (..) Things like they used to have "Teddy Tuck" that used to come home and they would have to keep a diary, (..)*

> VFW (2): "*Not waiting for school to start and getting that awareness through child minders, nursery settings and early years' workers. Start picking up on, (..) needs to be in awareness raising as part of early years training.*"

VSS(3) however showed some disagreement with involving early years practitioners in the sleep screening role. In her capacity also as a parent of a child with DD and

historical sleep problems, VSS (3) expressed how she would have felt uncomfortable with this level of intrusion into her family life. She explained how she would have felt embarrassed by the questions of early years professionals. Explanatory links can be drawn here with the legitimation of sleep problems and adherence to established beliefs that sleep problems are the private affair of families. This demonstrates how the view that children's sleep should not be openly discussed exists and can pose a barrier to the early identification of problems.

> *"I wouldn't have liked it (..) probably would have been embarrassed by it"* (VSS(3))

Generic practitioners and sleep practitioners should ask parents about sleep in a non-judgemental way.

Some parents voiced the caveat that the questioning of parents about sleep needed to be done in a non-judgemental manner. Parents cautioned how the issue of sleep was closely linked to parenting ability which was a sensitive and emotive topic for parents. Therefore, generic and sleep practitioners needed to be skilful in the way they approached the subject with parents, so they did not feel they were being criticised. If parents felt practitioners were being pejorative, this could prevent them from cooperating with screening process, resulting in sleep problems being left unrecognised. This chimes with self-efficacy theory (Bandura, 1977, 1997) and the strategy of social persuasion whereby individuals who receive encouragement are more likely to develop self-efficacy. Parents may be more open to discussing sleep and confident in their agency to change things, if questioned about it in a helpful and encouraging way, as the following extract from Amy and Gloria demonstrates:

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Amy "*It's the way that they ask questions as well,*

Gloria: "*without feeling that you're being judged*

Amy*: "without putting the parent on the spot so they feel it's their parenting"*

Parents should be empowered to screen for sleep problems using the personal child health record (PCHR) (agreement and disagreement).

Parents suggested including sleep screening questions in the red book or personal child health record (PCHR) they referred to when their children were of pre-school age so they could be well informed of normal sleep expectations and identify problems independently. Parents spoke about their familiarity with the PCHR and reliance upon it as a developmental checklist which made it an appropriate channel through which to convey screening information. This positive suggestion demonstrated parents need to be active participants, taking ownership of their child's health rather than passive or powerless recipients of care. Links with empowerment theory (Perkins & Zimmerman, 1995) can be made owing to parents' belief that their actions have a direct impact on the achievement of positive sleep outcomes for their child. Explanatory links can also be drawn with the transtheoretical model of change (TTM). This identifies various experiential change processes such as 'social liberation' and the empowerment of individuals to seek alternative methods of problem identification. The following extract from Gloria and Emily demonstrates this construction.

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"*More pages within the red book to ask you or make you do your own analysis of is your child up to scratch?*" (Gloria)

"*They don't come with manuals, the only manual we've got is the red book*" (Emily)

Sleep practitioner VDO showed agreement with empowering parents to screen for sleep problems, however SFSO expressed concern. She argued that including screening questions in the PCHR may mislead parents, and that all children develop sleep patterns at different rates making it difficult to distinguish between expected and delayed development. SFSO also explained how this step may be unnecessary as all generic practitioners and sleep practitioners should be asking about sleep as a matter of course. Amy, Jenny and Sharon confirmed this was not the case in their experience, therefore, using the PCHR as a vehicle for parent's independent sleep screening was a necessary priority for sleep service improvement. This challenge may have been indicative of an imbalance of power whereby SFSO was uncomfortable facilitating the decision making powers of parents or felt parents were unable to analyse professional knowledge. This was coupled with the inaccurate assumption that generic practitioners currently screened for sleep problems.

SFSO: "*It's a difficult one really isn't it? Because every child is different and some children take longer to fall into their sleep patterns don't they than other children. But as a parent I'm sure you're asked that by your health worker as well, are you?*"

Amy and Jenny: "*No no no*" *(shake heads)*

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Sharon: "*It's hit and miss with Health Visitors*"

Public awareness campaigns are needed around sleep.

The need to raise awareness about children's sleep and potential problems was highlighted as a priority for sleep service improvement. Parents and sleep practitioners felt that better understanding of sleep problems could be facilitated through increasing the public conversation about sleep. Through simple health promotion messages, media campaigns, press and magazine articles, more people would understand the importance of children's sleep and it would become 'normal' to discuss sleep and raise concerns. It is posited that stakeholders felt sleep did not feature prominently in the media in terms of the general public's emotions and there was an established belief that it was a private, family issue not the concern of wider society. This chimes with the recurring analytical theme of legitimation and the need to change the public perception of sleep problems. Explanatory links can also be drawn with the transtheoretical model (TTM) and the experiential change process of 'consciousness raising' whereby public awareness of problem behaviours is raised through media campaigns. The following narratives evidence the need for this sleep service improvement idea.

> "*Getting more magazines for women to write articles on sleep, it's simple get it out there( ) have a sleep awareness raising day*" (Jenny).
> "*There was all that talk about how everybody needs exercise, so maybe people should realise it's as important to get enough sleep*" (Sharon).

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VFW (2) "*The eat five a day, you know maybe some of the messages around sleep (..) needs to be in a catchy sort of easy to understand message (..)some kind of really simple bullet point information that you start to get all families understanding that okay this is what we need to be aspiring to*"

Parents are shy to socialise at pre-school groups where they may have their awareness raised.

Parents raised the contextual factor that some may not feel able to socialise with other parents at pre-school groups where they may learn about sleep awareness days or messages around sleep. Therefore, sleep practitioners should be mindful that some parents are isolated and will need services to actively reach out to them. This would ensure these parents become aware of children's sleep as a potential issue and guard against unnecessary delays in seeking sleep help or sleep problems going un-noticed. Feeling shy to socialise could be attributed to the parent's personality, emotional stability or fear of being seen as different. It is posited that this chimes with normalisation theory and the challenges of fitting in with other parents whose children do not have disabilities or sleep problems. The following discussion demonstrates this construction:

Gloria: "*Maybe as an individual you are not ready to access them because you are so shy yourself so you miss it. You don't see the early years services till about 3, so you need a professional to actually get in there because you're not ready to access the little clubs that your*

child can have access to, because it's stepping through the door isn't

it? (..)"

Fadheela: "Yes"

Gloria: "But if you have issues yourself, you are not going to access

these things so it's not going to be picked up till they get to school is

it?"

Parents may have literacy /communication problems preventing them from utilising screening or awareness raising initiatives.

Literacy and communication problems were cited as a potential barrier to parents receiving important awareness raising messages around sleep or being able to independently screen for sleep problems using the PCHR. Amy described her voluntary experience of running a parent support service and how some parents have their own literacy difficulties, reducing their capacity to get involved in the public conversation about sleep and recognise problems. Amy called for sleep practitioners to be continually mindful of how such parents may slip through the net and the need to communicate sleep messages through a range of creative media that all parents could access.

> "*There will be some parents who can't use the red book so there*
>
> *needs to be addition to that for parents who can't use books*" (Amy)

5.5. Discussion Theme Two- Sleep Services are Well Publicised and Accessible for Parents

5.5.1. Agreement/disagreement with review and exploratory study findings.

Adequate provision of sleep services.

Agreement was found with exploratory study findings (4.8 and 4.11) which identified a shortfall in available sleep services to meet the needs of children with DD. Parents spoke about their experiences of over-stretched sleep services and frustration at being continually told about funding constraints which left them feeling helpless. This showed how adequate specialist sleep services firstly needed to be on offer, which were then actively publicised so parents and generic practitioners were aware of them. The following narrative demonstrates this construction:

> "*I have tried to get him some help and nobody has ever mentioned any services (..) there should be some sort of service she could have sent us to that they could assess what was going on (..)A lot of services always tell you they've got no money, it's not your fault is it?" (*Sophia*).*
> "*There's not enough people working on the ground*" (Amy).

Sleep practitioners: VSS(3) and VFW agreed that the provision of sleep services for children with DD was poor. VSS(3) talked about generic practitioners avoiding asking parents about sleep because they perceived there were no sleep teams available to help them. VFW spoke of their experience in one local county of signposting parents and how there was very little sleep provision for families.

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Therefore, it was confirmed as important that sleep service provision was capable of meeting the level of service need and such teams were well publicised to parents and generic practitioners. It is posited that this perceived inadequate resourcing of services can be linked to the need to provide increased legitimacy to the issue of sleep. The following narrative shows sleep practitioner's agreement with the need to better resource sleep services:

> VSS(3): "*I was certainly not asked if sleep was a problem and I was to hazard a guess as to why it's not happening it because if they do ask there is a need for another service that they probably haven't got(..)l there is no point in asking the question if they can't help with the problem is there?"*

> VFW*: "I don't know how other counties fair but certainly in the 12 years there wasn't any I'd heard of (..),but the rest of the county as well there wasn't really much support.*"

Sleep teams should be easily accessible for parents.

Agreement was also shown with exploratory study findings (4.8.1. and 4.11.2) which identified problems with accessing specialist sleep services. Parents agreed that improving the ease of access to sleep teams was a priority area for improvement. They reported being excluded from specialist sleep services because their children did not meet the restrictive referral criteria. This was a significant barrier for parents to overcome at the start of their journey in looking for sleep help and is evidenced in the following narratives:

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"*That in itself is really frustrating that each team has a criteria and you don't fit into*" (Fadheela).

Emily "*Why does it need to be you have to have an ASD diagnosis? You have to have this diagnosis, that diagnosis to get into it, why does it have to be so?*"

VFW: "*I'd heard of (..) and met (..) a few times, but I think I don't know how easy that was for parents to tap into , I think as well there was a criteria to meet as well so*"

Further agreement with findings which identified that sleep teams should be easily and directly accessible for parents was provided by VDO. She argued when sleep teams stipulated the validation of gatekeeping professionals such as Social Workers to gain entrance to their services, this produced an unnecessary barrier for parents to overcome. Therefore, making the route to referral as simple and direct as possible for parents was identified as a priority for sleep service improvement, as VDO's extract shows:

"*I think that having that being able to refer yourself is important because a lot of parents don't want to have to find a social worker to be part of that service, because that has all the different connotations.*" (VDO)

It is posited that sleep teams set narrow entry criteria as a way of rationing their scarcely resourced service. Or in the case of NHS teams, this practice may reflect a gaming strategy to work towards achieving Government set 14 week waiting list

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targets (Welsh Assembly, 2015). For example, if services operated a fully inclusive, openly accessible service, their waiting list would increase dramatically, negatively impacting on waiting times. Nonetheless, existing problems with extended waiting times for sleep support were voiced by parents. Emily and Gloria reported waiting for up to 18 months for sleep support and added that appointments being cancelled created further delay. The need for speed of access to specialist sleep teams was therefore identified as a priority for service improvement. This shows how parents perceived waiting times were already unacceptably high and teams may feel compelled to manipulate the terms of service entry to ensure lists do not become unmanageable and fall short of governmental targets.

> Emily: "*But with easy access cause the problem is waiting 18 months*"

> Gloria: "*Yes quick access everything is 18 months down the line isn't it? Then you are on a waiting list, and then if you have got an appointment then it's cancelled about 3 times if it's anything like mine anyway*"

Improved publicising of sleep services.

Agreement was found with exploratory study findings (4.8.1. and 4.11.1) which identified the need for sleep services to take a more active role in promoting their services. Sharon, Emily and Gloria reported how the ease of finding the contact details of specialist sleep services was important to them. They did not want this to become an unnecessary barrier to receiving appropriate help. This again explains

parents' desire to be autonomous and take the lead on sourcing sleep support rather being than passive or powerless recipients of care which links to empowerment theory and participation. Furthermore, it is posited that the poor promotion of sleep teams leads to a limited service uptake which masks the true magnitude of family need and may be indicative of a rationing and gaming strategy by services.

> Sharon "*We need more promotion that there is help available*"
>
> Emily: "*A full contact list that's the point of in the beginning you've got your midwifes and your health visitors number and that's it there's no other contacts then is there?(..)*"
>
> Gloria*: "Even if they had websites, checklist websites, does your child follow this? And then if so contact that"*

Parents also agreed that generic practitioners had a limited knowledge of available sleep services rendering them unable to signpost parents to appropriate support. Parents expressed how they were often disappointed by the lack of awareness of specialist sleep teams and they expected them to know more, as is evident from the following narrative from Jenny:

> "*They should know about this more than I do, that's their job*" (Jenny)

5.5.2. New stakeholder insights.

Parents may have literacy /communication problems preventing them from enaging sleep service publicity.

Amy again raised how some parents may have literacy or communication problems and would find it hard to engage with written forms of sleep service publicity. Therefore, a priority for service improvement was suggested that services needed to be mindful of the needs of such parents and explore alternative methods of sleep service promotion.

> *"Parents who would for instance who wouldn't be looking for a website because they have their own communication issues* (..)*We need something to support parents who don't have the same communication as other parents*" (Amy)

Parents may be afraid of accessing specialist sleep services.

One parent (Sharon) voiced how she felt daunted by the task of making contact with sleep services. Although she was aware of a specialist sleep team and wanted to seek support, she lacked the confidence to approach them. Sharon's fear of contacting sleep practitioners describes a contextual factor that could prevent some parents from receiving help. Her words remind sleep practitioners that some parents will always feel apprehensive engaging with services and steps need to be taken to ensure they are as welcoming and parent-friendly as possible. It is suggested that this links with the principles of normalisation and parent's fear of stepping into 'disabled' services which may be perceived as different and deviant. Explanatory correlations can also

be drawn with the TTM and the temporal change stage of '*preparation*' whereby the individual has made a decision to invoke change in the near future, but is still deliberating.

> "*I've not got in touch yet it's a bit scary getting in touch*" (Sharon).

Provision of a generic sleep service (agreement and disagreement).

Parents suggested providing mainstream sleep services that parents of children with DD could access as a first point of call. They expressed how this may be easier for parents to approach as a 'normal' service and could provide a less threatening route to appropriate sleep help for parents who may be apprehensive. Furthermore, parents expressed the hope that generic practitioners in such universal sleep teams could offer basic advice for simple sleep problems and signpost parents to specialist sleep teams if needed. It is posited that this links with parent's desire for sameness and the principles of normalisation. This was a new priority for sleep service improvement which could make a positive difference to parents who had made the decision to seek sleep help, as is evident from the following narratives:

> Fadheela: "*So if you have a general sleep disorder team they could then signpost you to the more specific team*
>
> Amy: "*Because if you did it that way then that means that those parents who aren't gonna, they might not go to the website(..) they'll be on the same levels as all the other parents.*"

"Why does it need to be different? (..) Kids could just have a sleep issue, that could be their only issue I don't understand why you have to have separate."(Emily)

Some concerns were however raised with the notion of a generic sleep team. Clarissa cautioned against generic practitioners having unrealistic expectations of children with DD and a lack of understanding of their specific needs. Sleep practitioners VFW and VSS(3) highlighted that generic practitioners would need an improved awareness of DD, so parents did not have to repeatedly explain the impact of their child's condition. Explanatory links for these conflicting views can be drawn with the common criticism of normalisation (Culham & Nind, 2003) which highlights the danger of overlooking the specific needs of those with disabilities in attempts to provide a mainstream service. This demonstrates the importance of ensuring a generic sleep service is resourced by generic practitioners with appropriate, additional training in the needs of children with DD.

Clarissa: *"I think that's a lot to do with mainstream everything they expect too much of children anyway an average child a child with learning difficulties they've got no understanding whatsoever of what it is or how to deal with it."*
VFW : *"They would have to have an awareness because it's the worse thing for parents I think would be going to a generalist sleep advisor and then having explain what the condition the child has ."*

VSS(3): *"And them not knowing the difference"*

5.6. Discussion Theme Three- Parents and Sleep Practitioners Develop a Safe and Supportive Relationship

5.6.1. Agreement/disagreement with review and exploratory study findings.

Sleep practitioners should be empathic and positive with parents.

There was concordance with review findings evidenced by Beresford et al. (2012a, 2012b) and exploratory study findings (4.8.2, 4.9.2 and 4.12.2) which identified the need for compassionate practitioners. Gloria and Sophia voiced how sleep practitioners needed to show empathy with the daily challenge of living with children's sleep problems, but often failed to sincerely show this level of understanding. This seemed to present a significant barrier for parents who needed to be able to trust in the advice sleep practitioners eventually gave them.

> Gloria: "*The problem is every day they may not have the experience, they haven't been in your shoes they don't know what it's like to be awake 24/7 and really at the end of your tether and why should they*"
>
> Sophia: "*I've often found in our journey the people that have got these feelings who can personalise are the ones who do best*"

One parent elaborated on how she believed sleep practitioners could demonstrate good interpersonal skills with parents. Jenny had had an exemplary experience of support from a sleep practitioner and shared what she believed to be the key to a successful supportive relationship. For Jenny it was important for sleep practitioners to reassure parents that they were doing a good parenting job. This chimes with self-

efficacy theory and the strategy of social persuasion (Bandura, 1977, 1997) whereby individuals who receive encouragement are more likely to develop self-efficacy. It was also important to work optimistically with them, raising expectations that their child's sleep can improve. It is posited that this links with the wider need to raise health expectation in children with DD.

> "*She did it very positive (..) it kept him [*son] *hoping (..)the biggest thing you want to hear is you're not a bad parent'* (Jenny).

Parents receive the support they need throughout their contact with sleep services.

Agreement was found with review findings evidenced by Beresford et al. (2012g), Johnson et al. (1995) and O'Connell and Vanaan (2008), and exploratory study findings (4.9.2 and 4.12.2) which identified that parents should receive appropriate support to follow SHE advice. Gloria expanded on her experiences of sleep support described in the exploratory study and confirmed they were largely satisfactory. However, she reported needing to see the sleep practitioner more regularly than was offered and was still in the process of trying to get them to visit her at home again to continue with sleep work. Her words conveyed a need for sleep services to be able to offer an increased intensity and frequency of parent contact. Explanatory links can be made with the TTM and the temporal stage of '*action'* whereby the individual has started to make changes and the behavioural change process of ' *helping relationships'* which relate to the need for support for desired change.

> *"I didn't see her as many times as I would have liked to (..) I'm trying to get her back"* (Gloria)

5.6.2. New stakeholder insights.

Continuity of sleep practitioner support.

It was explicated that sleep advice should be given by the same sleep practitioner continuously and how the thread of continuity was commonly lost when there was excessive staffing movement within sleep teams. Without continuity, Jenny, Sharon and Fadheela argued that the opportunity to build a relationship with sleep practitioners was also limited. Poor continuity could be explained by the under-resourcing of sleep services already alluded to (4.8.1) and linked to the need to increase the legitimacy of sleep problems in children with DD.

> Jenny: "*We should see the same person each time*"

> Sharon: "*Yes the person we have seen has moved to different departments so the consistency isn't there. They change departments quite often. I've got to try and get through to CAMHS to get this next appointment.*"

> Fadheela: "*We have been so disappointed with so many others and actually it's about having that consistency*"

5.7. Discussion Theme Four- Parents and Sleep Practitioners Improve Their Understanding of the Sleep Problem

5.7.1. Agreement/disagreement with review and exploratory study findings.

Sleep practitioners need to complete a comprehensive sleep assessment.

Agreement was found with exploratory study findings (4.7.1 and 4.12.1) which identified that sleep practitioners needed to complete a comprehensive sleep assessment involving observing the child at home and over multiple sessions. Fadheela and Gloria described how sleep practitioners needed to observe their child at home to fully understand their circumstances and sleep problems. There was a strong sense that if this time was invested, parents would have more confidence and trust in the advice sleep practitioners gave, therefore home visits should be an integral part of the assessment process.

> "*They need to come home professionals needs to come and see children in their homes because that is where you are going to get the best possible picture about them around sleep* " (Fadheela)
>
> "*They can watch what the child is doing on their own setting and see how they are obviously home is the best because they are different at home*" (Gloria)

A comprehensive sleep assessment could also be achieved by performing multiple assessment sessions according to SFSO, VSP(2), Fadheela, Amy and Jenny. They described how it was unrealistic for sleep practitioners to understand the nature of children's sleep difficulties based on a single session. Parents also described feeling

affronted when assumptions were made too quickly and which resulted in ineffective advice. It was important that parents felt sleep practitioners were investing time listening to parents before rushing to give sleep advice. It is posited that a comprehensive assessment was key to effective SHE implementation, because parents valued customised advice that was tailored and responsive to their child's individual needs. Explanatory links can be made with a developing analytical theme of customisation, which describes the importance of adapting SHE advice according to assessed need.

> *"They make conclusions about children based on forty minutes. We have spent a lifetime with these children and we are only just getting to grips with how they function so actually one-off sessions are a big 'no no' cause you don't get a true picture about how the kids are so it's about having multiple sessions"* (Fadheela)

> Jenny "*I'd rather they took the time to get to know the child, that is my main big thing is that they don't have enough time to get to know the child and parent*"
> Amy "*(..) I think sometimes there's not enough information gained before things are put into practice that might not actually be right for that family.*"

Sleep practitioners need to complete a competent sleep assessment.

Agreement was found with exploratory study findings (4.12.1) which highlighted how sleep practitioners needed to be able to competently interpret sleep assessment findings. SFSO and VSP(2) discussed the importance of performing a psychological

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formulation and working in partnership with parents to arrive at a deeper understanding of the sleep problem. This links with empowerment theory and analytical ideas around sharing professional and lay knowledge and how this may be important for intervention success. Evidence for this was however, limited to sleep practitioners only. Parents in both the exploratory and co-design study reported little experience of this sleep problem 'diagnosis' stage and potential for knowledge exchange, which may explain their lack of discussion and provision of supporting evidence.

> VSP(2):"*I always say to parents it's a process of elimination it's like a jigsaw puzzle if you don't find all the pieces it's not all going to fit together and you won't get the end result , so you have to take the time to be able to do it.*"
>
> SFSO:"*We do is we do a formulation we go through the strengths and positives, you look at the positives that the parents are doing, and the child as well and erm, you look at what the problem is what the triggers (..) so it's about really going through the assessment finely and with the parent.*"

5.7.2. New stakeholder insights.

Comprehensive sleep assessment should involve observing children at bedtime.

Fadheela, Emily and Amy reported that a comprehensive sleep assessment should include observing the child at bedtime to facilitate sleep problem understanding. It seemed important to parents that sleep practitioners took this step so they could develop a good appreciation of their individual situation. However, sleep

practitioners needed to spend time getting to know parents first before building up to this level of observation. Parents felt this was a priority for sleep service improvement which could facilitate parents' and sleep practitioners' improved understanding of children's sleep problems. Furthermore, it is posited that this continues to link with the theme of customisation and advice which has been developed following a substantial resource and time investment in the sleep assessment process.

> Fadheela: "*You may have that introductory session to somebody else in your home and then building it up sort of graded exposure to where they are actually looking at the child working with the child*"

> Emily: "Yes"

> Amy: "*Or could they possibly come to your house at bedtime*?"

> Fadheela: "*That's what's needed*"

> Amy: "*In their room*"

Comprehensive sleep assessment should involve other members of the multi-disciplinary team and partner organisations.

Parents and sleep practitioners reported that a comprehensive sleep assessment should involve collaboration with other members of the multi-disciplinary team and partner organisations. Stakeholders explained how due to the complexity of children with DD, it was unlikely that a sole sleep practitioner could unpick the multiple causes of their sleep problems and such narrow assessments often led to ineffective advice. Therefore, it was imperative that sleep practitioners sought the opinions of

other generic practitioners involved with the child such as Health Visitors or Occupational Therapists. Indeed, adopting a multi-disciplinary, collaborative approach fits with current approaches to health care and is something that services should aspire to (NHS, Wales, 2011).

> Fadheela: "*It's really about a multi-disciplinary approach(..), if you don't have a formulation that involves more than one profession when you are talking about labelling a child it's a real problem (..) unless you've got lots of people looking at it you are not going to get a true picture*"
>
> Emily "*Absolutely.*"

The importance of involving partner organisations involved in children's care such as education, transport or short break settings in the sleep assessment process was also discussed. Parents were impressed by sleep practitioners who observed children in other settings, to get a detailed picture of the child's complete day to inform their understanding of how they slept. It is posited that meaningful collaboration with others involved in the child's care was important to co-designers because of the desire to deliver and receive customised SHE advice tailored to individual need, which is evidenced in the following narrative:

> "*She watched what was going on, on numerous occasions and she went into school the school setting and was able to differentiate between the two because he's not the same at home*"

Gloria

> *"You need to look at the child holistically as well and we always look at days nights behaviour, school everything then as well so I think that's something that's really important looking at everything that's going on"*

<div align="right">VSP(2)</div>

Parents may not give assessment information honestly.

One parent (Jenny) cautioned how the achievement of a comprehensive sleep assessment may be compromised if parents were reluctant to fully engage in the assessment process. She asserted how families of children with DD and sleep problems may have a number of difficult issues to cope with, some of which they may be uncomfortable discussing with sleep practitioners. Sometimes parents may not provide all of the information necessary to ensure a thorough sleep assessment and it was important that sleep practitioners were mindful of this contextual factor which could affect the success of the intervention. It is posited that parents may give incorrect or withhold information due to a mistrust of generic practitioners which highlights the need for effective relationship building during the assessment phase of the intervention.

> *"There is so many issues that may be they wouldn't want to talk about certain things, they're could be other things underlying that are going on, that maybe they wouldn't talk to a professional"* (Jenny)

5.8. Discussion Theme Five- Regularity and Quality of Child's Sleep Improves

5.8.1. Agreement/disagreement with review and exploratory study findings.

Parents are supported to follow sleep advice.

Agreement was found with review findings evidenced by Beresford et al. (2012b, 2012g), exploratory study findings (4.12.2) which identified the need for parent buddies to support parents with following sleep advice. Amy and Emily talked about utilising 'parent experts' and described a sense of ranking such personal experience very highly. They argued such experts should exist as paid staff within sleep teams to ensure their contribution was valued and recognised. It is posited that parents placed significant value on lay knowledge and felt it should be used to complement professional knowledge. This effective sharing of professional and parental knowledge seemed crucial as something that would make the SHE intervention work. Explanatory links can be drawn with self-efficacy theory and the positive influence of vicarious experience (Bandura, 1977, 1997) to improve parents' agency to implement change. Further correlations can be made with empowerment theory and the TTM's temporal '*action*' stage and change process of '*helping relationships*' which highlights the need for appropriate support for desired change.

> Amy: "*Would it not be an idea to have parent experts? Paid parents*"
>
> Emily "*That makes a heck of a lot of sense cause the times I've sat there 'bla bla bla' 'they'll be fine' cause you go home at night*"
>
> Amy: "*Frankly who has experience, who has actually been paid to do that. My point is they never give it the value that is deserves*"

VDO, however, disagreed with employing a parent buddy to support parents in following sleep advice, and argued resources would be better spent on training sleep practitioners to deliver advice in a supportive way. It is posited that VDO may have held this view due to a vested interest in protecting her professional role. She may have perceived the giving over of professional responsibility to a paid parent buddy as a threat to her employment. Therefore, there was a disparity in stakeholder views around the concept of a paid parent buddy to support the implementation of sleep advice.

> VDO: "*I still think that to pay for someone to mediate shouldn't that money be better spent training professionals on how to build that relationship and how to behave?*"

In terms of the activity of inviting parents to bring peer supporters to all appointments with sleep practitioners to support them with following sleep advice, parent co-designers were asked in the co-design event invitation letters to bring a friend or relative if they wished. This decision was taken based on exploratory study findings (4.9.3) which indicated that parents might feel threatened by a perceived imbalance of power during sessions with sleep practitioners and the researcher. No additional verbal evidence was gathered to support the need for this action during the delivery of sleep advice. However, one parent (Sharon) chose to bring a supporter to the first co-design event. Her supporter did not actively engage with the event, but seemed to provide moral support for Sharon which enabled her to participate. Sharon's actions concurred with those of parents Sarah (parent) and the words of

Gloria in the exploratory study. This provided further agreement that parents may benefit from the active offer of bringing peer supporters to all sleep appointments which may help them engage with advice and improve their child's sleep.

Agreement was found with exploratory study findings (4.12.2) which identified that parents needed to be prepared for the hard work involved in following sleep advice. Jenny agreed parents often had the misconception that improvements in sleep would happen quickly once sleep advice was followed. She voiced how parents needed to be prepared for more gradual improvements in sleep and how their expectations needed to be realistic. VDO also talked about sleep practitioners warning parents in advance that it may take weeks before they see sleep improvements and this was important to help parents stay motivated in following the advice. Therefore, advising parents on how long sleep advice might take to work was confirmed as an important activity in supporting them to implement sleep practitioners' recommendations. Offering this supplementary information about expected work load and time frames links with ideas around sharing professional knowledge and how this is important for intervention success.

> Jenny: "*Time. Cause it takes a certain amount of time to get there. It doesn't happen overnight, cause a lot of parents think it's going to, you've got to work at it.*"

> VDO: "*You know don't as a professional say if you do this for so long, the problem will be sorted you know because every child is different, (..) it might take two weeks but for other children it might take an awful lot longer*"

"I might have stuck with it then if somebody had have said (laughs) just keep going cause it will work eventually. Cause after 3 weeks all these books are telling me he should be doing it. And it wasn't and I thought oh well." VSS(3)

Agreement was found with exploratory study findings (4.12.2) which identified the need to support families with visual prompts when delivering sleep advice. Amy spoke about how some parents and children with DD were visual thinkers and would respond well to sleep advice being represented visually through pictures. She also suggested this may make the process more enjoyable for families which would motivate them to engage with sleep practitioners. SFSO confirmed how she regularly used visual prompts to support parents with following sleep advice and how this was effective. This therefore confirms how using visual media to communicate sleep advice is an important activity for sleep practitioners to utilise at their discretion to facilitate improvements in children's sleep. Explanatory links can be drawn with the need to provide a customised service responsive to individual family need.

Amy: "A *vision board do it with them as a family. Use pictures so it's visual you know cause most are visual thinkers so"*

SFSO: "*Yes I do that sometimes with families that need a bit more visual prompts* (..)

Amy: "*You could use pictures for anything really its simple it may be we complicate things too much it could be something as simple as enjoying making a plan for how things are going to get better*"

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Parents receive effective sleep advice.

VSP(2) agreed with exploratory study findings (4.12.2) which identified that sleep practitioners should explain to parents the rationale behind sleep advice to help motivate them to follow it. VSP(2) talked about informing parents why a consistent bedtime routine was essential, to develop their understanding and convince them of the need to implement this advice. She suggested the giving of this explanatory information could also be helpful in dispelling any misconceptions that sleep practitioners give such advice to create added work for parents. Therefore, the delivering of sleep advice supported by underpinning rationale was confirmed as a priority for sleep service improvement. This again links with ideas around the sharing of professional knowledge with parents and how this may be important in making the intervention work.

> "*So that's where your bedtime routine and the reasoning behind the routines, you could then have little snippets for parents so then their understanding is being improved then as well. You don't think a bedtime routines in place just to make you have more work to do there is actually a reason behind it.*"(VSP(2))

Agreement was also found with review findings reported in Beresford et al. (2012g) and exploratory study findings (4.12.2) which recommended psycho-education was delivered to parents as part of the advice package. In a discussion between Amy and VSP(2) the example of informing parents of the sleep theory behind sleep terrors and sleep walking was highlighted. VSP(2) suggested that Amy would have benefitted from understanding the science behind these types of sleep disturbances and would have gained some reassurance and confidence to progress with following sleep

advice. Therefore, investing time in educating parents about the mechanisms of sleep was confirmed to be important in facilitating parents to follow sleep advice. This relates to the continuing theme of sharing professional and lay knowledge as a potential theory explaining what makes a SHE intervention work.

> Amy "*They were horrific night terrors and he used to scream, scream (..)*

> VSP(2): "*Were you explained why the likeliness of the sleep walking and the night terrors together, the reasons behind that?*

> Amy: *No no nothing*

> VSP(2): *I think just knowing(..) just finding out the reasons behind something can just put yourself at ease then, okay it doesn't get rid of the problem, but it puts yourself at ease then and you can start building on that.*

Agreement was shown with exploratory study findings (4.9.2 and 4.12.2) which identified that effective sleep advice should be delivered in a non-directive way. VDO who was also a parent of children with DD and historical sleep problems described how it was common for parents to feel judged throughout their contact with generic practitioners and sleep practitioners. She explained how there was a constant pressure to follow every element of advice given which could be overwhelming for parents. It was common to feel like a 'bad parent' if they failed to implement advice fully and consistently and so it was important that sleep advice was delivered in a non-judgemental manner. This relates to self-efficacy theory and

the strategy of social persuasion (Bandura, 1977, 1997). Offering advice in a supportive and positive way seems to be important in convincing parents that they have the ability to succeed and improve their child's sleep.

> "*If your child has additional needs you constantly feel that you are being judged not even just at meetings but from things that you read, you know I should be doing all this, I should be doing Makaton every single day you know all time and if you say you're not then you are not as good a parent as you should be because you are not helping your child develop you know so it's that being judged and this is just another issue.*" (VDO)

Agreement was found with review findings reported in Beresford et al. (2012f, 2012g) which identified the contextual factor of the child's home environment and the impact this could have upon parents' ability to follow effective sleep advice. Jenny confirmed that when she was trying to implement a sleep practitioner's sleep recommendations it was challenging to do so because of problem neighbours. In Jenny's case, her neighbours created a lot of noise at night which prevented her son from sleeping well. It was therefore identified as important to be aware of how the quality of a child's sleep environment could affect the success of a sleep intervention.

> "*Yes I had a problem with neighbours and they would be partying till god knows what time and it would kick off in the middle of the night, that wouldn't help.*"(Jenny)

Agreement was shown with review findings reported in Beresford et al. (2012f, 2012 g) and exploratory study findings (4.12.2) which identified the contextual factor of parents' busy day to day lives and how this can affect their ability to follow effective sleep advice. VSP(2) talked about how parents who worked full-time often struggled to fit in all the steps of a bedtime routine, and how sleep practitioners needed to be aware that sleep advice should fit with a family's daily lifestyle to be effective. This links with ideas around the customisation of sleep support and need to tailor advice to family need. Explanatory links can also be drawn with self-efficacy theory (Bandura, 1977, 1997) and people's subjective belief in their physical ability to perform behaviours.

> *"Generally you just have working parents these days. If you have two working parents you are picking your child up from nursery, you are not getting home until 7 o'clock some nights by the time you start dinner, all of that when does the wind down time happen it's so hard when you are really busy"* (VSP(2))

5.8.2. New stakeholder insights.

Sleep practitioners need to praise parents for achievements throughout their contact.

Two parents identified a need for sleep practitioners to support parents by praising them for achievements throughout their contact. Amy and Jenny felt it was common for parents to focus on impairments but could be motivated to engage in sleep support if sleep practitioners actively pointed out achievements or positive attributes. They posited that positive reinforcement would help build parent's confidence and

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trigger more positive activity. Jenny qualified this with her own experience of exceptional support from a sleep practitioner which convinced her she had the power to improve her son's sleep. Therefore, ensuring sleep practitioners adopt an encouraging approach when supporting parents to follow sleep advice was confirmed as an important factor in achieving sleep improvements. This links with self-efficacy theory and the strategy of social persuasion (Bandura, 1977, 1997).

> Amy: "*The massive thing that has come out for me is concentrating on all the positive things that are happening in that family because the more we concentrate on positive things the more positive things they attract and if that starts to happen in their life then starts to build confidence isn't it?*"
>
> Jenny: "*Yes.*"
>
> Amy: "*As parents, we are constantly having to look at the negative about your child and what you're doing wrong, and it brings you right down doesn't it? (..)*
>
> Jenny: "*That's what (..) are good at they pick up on your good points and they make you feel really positive like you are not doing things wrong*"

Sleep practitioners need to liaise with others to follow sleep advice consistently across all settings.

SFSO, VSS(3) and Jenny agreed that partner organisations involved in a child's care needed to be involved in the consistent implementation of sleep advice. For example,

it was important to advise school transport staff of strategies to keep children awake during their journey, to support parents in their efforts to improve children's sleep at night. Therefore, ensuring sleep practitioners work closely with partner organisations in ensuring sleep advice is implemented consistently across all settings was identified as a new priority for sleep service improvement which could help facilitate improvements in children's sleep. This links with the theme of sharing professional knowledge as an important factor in intervention success.

> SFSO: "*Parents might not think they are allowed to ask taxi escort to keep them awake, but a professional writing a letter to the taxi firm working with the mum, saying maybe if you distract the child by doing this they won't fall asleep.*"
>
> Jenny: "*Yes*"

Agreement was shown with the need for a consistent approach to sleep work across all settings where a child spends their time. VSS(3) talked about ensuring grandparents were informed of sleep advice if they looked after the child during the evenings or overnight. She explained how it was important grandparents supported parents by following sleep advice consistently to ensure the child's sleep could be improved. Amy also spoke more generally about the need for consistency when supporting parents with children's sleep problems and how it was easy to undo progress made if there were gaps in the way sleep strategies were followed. Therefore, ensuring parents are supported to follow sleep advice consistently by collaborating with others involved in children's care was confirmed as an important activity for sleep practitioners to perform. This continues to link with themes around

the constant exchange of professional knowledge and its importance in making the intervention work.

> VSS(3): "*Certainly if they are having any kind of caring responsibility for the child because it has to be consistent, so if they are staying at grandparents once a week and they are doing something completely different to what mums doing then it's not going to work. (..)*"

> Amy: "*it all falls to pieces again so you don't carry on with the routine you have been building so it needs consistency.*"

5.9. Discussion Theme Six- Quality of Life Improves for the Child and Family.

5.9.1. Agreement with review and exploratory study findings.

Parents score favourably on quality of life measures and evaluation questionnaires.

Agreement was found with review findings reported in Beresford et al. (2012c, 2012d, 2012e) and Malow et al (2013) and exploratory study findings (4.12.1) which identified that sleep services should evaluate quality of life (QOL) improvements at the end of their interventions. SFSO affirmed her earlier comments that the sleep team she represented, always administered QOL outcome measures to quantify improvements but highlighted the challenges involved in persuading parents to complete and return them. VSP(2) also talked about poor return rates of evaluation questionnaires and how the resourcing of a final evaluation home visit would be helpful to establish QOL improvements with more families. Therefore, resourcing

sleep practitioner time to administer QOL measures was identified as an important contextual factor. Links can be made with self-efficacy theory and the concept of mastery experiences (Bandura, 1977, 1997) whereby positive effects are quantified to help build people's sense of self-efficacy and belief that they can sustain changes made.

> SFSO: "*We have five different measures we ask parents to fill in (.) they don't always come back if you've ended an intervention (..)so you do miss a few like that but most parents are brilliant filling them in and its good for them to see the pre and post as well(..)*"

> VSP(2): "*Not quality of life ones we tend to have our feedback forms are poorly returned I think it would be a lot better if we could go out and see the families again and actually fill it out with families.*"

5.9.2. New stakeholder insights.

What improved quality of life looks like.

Parents and sleep practitioners described how parents might feel if a sleep intervention had been successful and the knock-on effect of improved quality of life for the family. VDO, Jenny, Amy and Sharon agreed that they would expect to see an improvement in the child's daytime behaviour and reduction in parents' stress. Amy and Sharon also described the positive side effect of parents' sleep improving. Stakeholders' descriptions were indicative of a clear appreciation of the broader effects of children's sleep problems and how they affect multiple areas of family life.

VFW(2) stated that parents may feel a sense of empowerment and feel able to support other families with sleep problems. This relates to the parent buddy idea and links with the theme of sharing and valuing lay and professional knowledge. All of these factors helped to describe what improved quality of life may look like for families once their child's sleep had been improved.

> Sharon: "*You stop feeling like a zombie*"
>
> VFW(2): "*I think families are then ready to help other families.*"

Parents feel supported to maintain progress with help from parent support groups.

VDO, Jenny and Amy suggested that parents could be supported to sustain sleep improvements in their children, by attending parent support groups. It was posited that parents need peer support and an opportunity to connect with others in the same position, to help them maintain sleep progress and see eventual improvements in quality of life. This chimes with the theme of valuing and sharing lay and professional knowledge as an important factor that makes the intervention work. Links can also be drawn with self-efficacy theory (Bandura, 1977, 1997) and the positive influence of vicarious experience to improve parents' agency to sustain sleep improvements. Furthermore, explanatory relationships can be drawn with empowerment theory and the TTM's temporal stage of '*maintenance*' whereby change is sustained and the behavioural change process of '*stimulus control*' which highlights the role of self-help groups in reducing relapse.

> "*Parents need to know that there is this group, this group that and the other out there just so they can go "arggh!". That is really important.*" (Jenny)

In discussing the important role of parent support groups with helping parents to maintain sleep improvements, stakeholders however voiced how parents who run them needed more support. Amy and VDO emphasised that running a parent support group could be very time consuming and challenging and often groups discontinued if those running them were poorly supported. The need for sleep practitioners therefore to collaborate closely with parent support groups and value their role in maintaining sleep progress was identified as a new priority for sleep service improvement. This links with ideas around the legitimation of sleep problems and the value society and professionals place on sustained sleep improvements for families.

> Amy: "*But the self-help groups the parents who run it need to be supported because what can happen is you're a parent of a disabled child or whatever, and you end up helping everyone else*"

> VDO: "*Yes, I look after the parent support groups (..) many had fallen by the wayside and now we are building them back up again.*"

5.10 Development of SHE Tool

Co-designers were invited in the third co-design event to explore the acceptability of SHE advice components identified in the scoping review, in order to finalise the development of a SHE tool (Table 23). Findings are presented that evidence

decisions made around the inclusion/exclusion of components in the final tool. Advice components were included if they were supported by a minimum of two review citations or one citation plus additional evidence or stakeholder agreement, to ensure tool validity. Furthermore, it should be noted that wordings of components incorporate additional verbs such as 'encourage' or 'consider' to reflect the need for non-directive language as identified in the exploratory study (4.12.2). Stakeholder discussions are now presented in relation to each advice area.

5.10.1. Sleep timing (Figure 3, section 2.7.7.)

Amy and Gloria showed agreement with the advice to '*set consistent bedtimes and wake times (including holidays and weekends)*'. Gloria described how this was effective in improving her son's sleep pattern, although it was challenging to wake him consistently early on Sundays because of a desire to lie-in on weekends. This highlights how parents may need additional support to adhere to this component during weekends and holiday times.

> "*We all wake up at 6.30 on Sundays (..) even if it's a day off so it's a little bit annoying, but if I stick to the routine then come school time I don't have to change the routine.*" (Gloria)

Emily disagreed with the advice to '*go to bed only when tired*' and felt it would be challenging to implement for her son who never seemed to show signs of tiredness. In the review, this component was cited by Stores and Wiggs (2001) and no further, more recent SHE tools had included it. Therefore, on the basis of limited and dated review evidence and stakeholder challenge this component was excluded.

> *"That wouldn't work (..) the go to bed when tired , he's never tired"*
>
> (Emily)

No further comment was made on the remaining four components of '*set age appropriate bedtimes*', '*encourage age appropriate daytime napping*', '*avoid late afternoon napping*' and '*avoid excessive time awake in bed*' which were each evidenced by multiple review citations, and included in the final tool.

5.10.2. Bedtime routines (Figure 3, section 2.7.7)

Gloria, Clarissa, Amy and VSS(3) showed support for the advice to '*set relaxing routine*' but acknowledged that parents needed extra support to enable them to consistently follow a new routine. This demonstrates how parents need sustained guidance from sleep practitioners when implementing SHE advice which was reflected in the programme theory. Amy demonstrates the need for additional support in the following extract:

> *"It's keeping that routine and that's the hardest thing and it puts pressures on you, but if you can get through it"* (Amy)

Emily, Gloria and VSS(3) challenged the view that some children would be very resistant to the advice to '*discourage television and blue light emitting devices at bedtime*'. SFSO agreed that families often needed extra support to limit screen time, and realistic goals needed to be set rather than 'carte blanche' bans. Amy, Jenny, Emily and VSP(2) made links to the component: '*consider blue light blocker sunglasses*' and agreed this could support parents with limiting the effects of blue light; therefore both these components were collapsed into one. Additional evidence

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from Sasseville, Paquet, Sevigny and Herbert (2006) also explicates the decision to include the suggestion of blue-blocker sunglasses, which was supported by a single review citation (Handsel Project, 2012). Furthermore, this discussion evidences how SHE advice should be tailored to families' needs and backed by appropriate implementation support. The following extracts from Emily and SFSO demonstrate how parents may struggle to remove television and blue-light emitting devices at bedtime:

> Emily: "*Trying to take it (*the kindle*) away from him it's like oh my god it's like the world's ended and we have a massive meltdown every single night about this (..)*"

> SFSO: "*Professionals say oh you need to get his telly out of the room (..) I'm not here to take anybody's telly anywhere it's about controlling how much he watches.*"

Concerns were raised by Emily with the advice to '*limit rituals*' as she cautioned that her son needed rituals and would strongly resist any attempts to reduce them. Amy, Clarissa and VSP(2) agreed that this could be challenging, however, purported it was a necessary component, achievable with the right support, therefore this was included in the final tool. The following extract from Emily and Amy highlights the discussion which took place around the advice to '*limit rituals*'.

> Emily: "*But they need that ritual and you have got absolutely zero chance of* (son's name) *now if he doesn't do it he will have a complete meltdown and that will be it his day will be ruined.*"

> Amy: "*But you've got to work on that because with (*Son's name*) had rituals and I can't begin to tell you what they were (..) now we are down to just some noises and stuff it's taken a long time but we have managed to access the right kind of therapy.*"

Amy also described how she had found mindfulness relaxation an effective tool in promoting sleep for her son. In her dialogue she linked the '*include relaxation techniques*' and '*consider alternative therapies*' components. Similarity between these components therefore prompted the decision to combine them. No comment was made about the advice to '*ensure routine activities are consistently ordered and timed*' and '*ensure routine is 20-45 minute duration*' which were supported by multiple review citations and therefore included. However, the component '*child completes a step independently*' was removed due to limited review evidence. The following extract from Amy shows her support for relaxation and alternative therapies.

> "*I think alternative therapies are good like mindfulness relaxation techniques are good cause I thought none of those had gone in with (son's name) he actually uses the breathing that I taught him now more than he ever did*"   (Amy)

5.10.3. Behaviour management (Figure 4, section 2.7.7)

VSP(2), VSS(3), VFW and Amy expressed agreement for the component '*ensure bedroom is not used as a punishment setting*' thus supporting its inclusion in the

final tool. SFSO, VSP(2) and Jenny cautioned that the advice to '*avoid soothing to sleep with a bottle/breast after 6-12 months old*' was challenging for some families; further evidencing the level of support needed by parents to follow SHE advice. Stakeholders also argued that this component should be part of a wider treatment package incorporating the need for attention to weaning or eating difficulties. Therefore, the caveat *(# care re: weaning/feeding difficulties*) was added to this component. Support for this is evidenced in the following extract:

> SFSO: "*I'm meeting a lot of younger children and children who are 3 and still using the bottle to soothe at night and it's very difficult cause it's one thing that does soothe them* (..)
>
> VSP(2): "*Yer the bottle is hard especially if you have got a child who is not a good eater in the day.*"
>
> SFSO: "*That's right*"
>
> VSP(2): "*They worry if you remove the feeds in the night, cause they are already under weight.*"

Emily, Clarissa and Jenny showed agreement with the advice to '*incorporate rewards*' and VSP(2) added "*It has to be a reward that works for your child*" therefore the phrase '*which are meaningful to the child*' was added to this component. This further evidenced how SHE advice should be tailored to specific needs. No comment was made on components: '*ensure child falls asleep and sleeps alone in own bed*', '*set and stick to limits*', '*put child to sleep drowsy*', '*give minimal interactions during night-time feeds or night awakenings*' and '*encourage child to think about problems/plans before going to bed*' (appropriately re-positioned from

physiological advice area); however, these were all included in the final tool based on adequate review evidence.

5.10.4. Environment (Figure 6, section 2.7.7)

VFW, VSS(3), Clarissa and Emily challenged the advice to '*ensure quiet noise levels at sleep times'* arguing it could be hard for some families to control noise external to the house. This emphasised again how SHE should be tailored to the individual circumstances of the family, to ensure realistic goals are set and the decision was made to include this component based on multiple review citations. The following extract from VFW shows how not all families would be able to maintain quiet noise levels when needed:

> "*I think quiet noise levels is going to be a difficult one cause I can just imagine you can't control some of the noise outside of the house."*
>
> (VFW)

VSS(3) expressed concerns with the acceptability of the component '*consider room temperature 16-20c, bedding and sleep clothes to maintain comfortable body temperature'*. She contended that low-income families may struggle to keep their houses warm in the winter. This deepened understanding around this component and highlighted a need for the sensitive selection of SHE advice points and appropriate collaboration with partner organisations. Moreover, based on adequate review evidence the decision was made to include this component in the final tool.

> "*So many houses that are financially fuel poor that that just wouldn't be possible for some houses."* (VSS(3))

Stakeholder agreement was expressed for advice to '*ensure a darkened room (black-out blind)*', '*ensure bedroom has a familiar layout and calm déc*or' and '*allow security object to promote self-soothing*' which were all included in the final tool. Agreement was also shown for the component: '*consider sensory sensitivities of the child*' and the final tool combined this with '*mask background noise with white noise*' to ensure the needs of children with auditory sensitivities were appropriately addressed. Advice points: '*Ensure bed is comfortable (consider sleep systems)*' and '*remove or hide stimulating toys from the bedroom*' did not receive stakeholder comment but were included based on adequate review evidence. '*Use a nightlight*' (also supported by adequate review evidence) and '*consider a red-modelling bulb*' were collapsed into one component because both offered acceptable alternatives to children who may be scared of darkened bedrooms. Additional evidence from Wright, Lack and Kennaway (2004) supported inclusion of the reference to a red modelling bulb which was originally underpinned by evidence from Handsel Project (2012) only.

5.10.5. Physiological factors (Figure 5, section 2.7.7)

VSP (2) agreed with advice to '*ensure child has plenty of light in the day*' and Amy agreed with the components '*encourage healthy diet, limit fat and sugar intake*' and '*encourage daily exercise*' which due to similarity were combined with '*avoid late evening exercise*' (adequately supported by review evidence) in the final tool. No stakeholder comment was received on advice to '*avoid smoking and alcohol*', '*light meals only near bedtime*', '*limit caffeine intake in hours before bedtime*' and '*use toilet before bed*'; however each component was supported with more than two scoping review citations, hence their inclusion.

Also, stakeholders did not discuss the adequately evidenced component of '*drink milk before bedtime*', which in the final tool was merged with '*encourage tryptophan/melatonin rich foods combined with carbohydrates at suppertime*' because of similarity between these components. However, SFSO described how it was commonly challenging to encourage tryptophan/melatonin rich foods, as children typically did not like them. This demonstrated the importance of customising SHE advice rather than giving out standardised to all families which may be inappropriate for some children. The following extract from SFSO describes how children may dislike tryptophan/melatonin rich foods:

> "*It's difficult you know the melatonin rich foods things like porridge, things that children would not eat*" (SFSO).

Jenny, Gloria and VSS(3) dealt with the uncertainty around conflicting advice points: '*avoid fluids at bedtime and night*' and '*encourage child to stay hydrated*' by proposing that sleep practitioners and parents needed to make an individual judgement based on the needs of the child. Stakeholders agreed that following a comprehensive assessment, the specific situation and needs of the child would be clear and advice could be tailored accordingly. Jenny stated how she would not restrict fluids for her child at bedtime due to links between dehydration and migraines. Gloria agreed and described how problems with toileting at school meant fluids were important in the evenings for her son, to encourage appropriate bowel and bladder function at home. This corroborates exploratory study findings that SHE advice needs to be customised following a thorough assessment and the final wording of this component reflects this '*ensure the child's individual hydration*

needs are met'. The following discussion between Jenny, VSS(3) and Gloria demonstrates the need for this refinement to component wording.

> Jenny: "*You have to judge the child cause (*child's name) *has to drink a lot*"

> VSS(3): "*Yes*"

> Jenny: "*Cause he suffers with migraine so you know sometimes it's just dehydration that brings it on if he's been running around all day or whatever, so you've just got to judge it, but that would go with the sleep diary and the food diary and anything and if you'd done all those things in the first place it would help*"

> Gloria: "*Mine won't wee or poo during the day so (..)there is no way I can stop him from drinking cause as soon as he gets in at 4 o-clock it's like there is your 2 bottles with all your medicine in, drink them and by the time it gets to 10 o'clock at night he wants to wee.*"

Finally, advice to '*avoid blackcurrant juice in the evenings'* was supported by a single review citation (Early Support, 2010) and did not receive any stakeholder discussion. Therefore, additional evidence provided by Cohn (2010) and Salisbury NHS Foundation Trust (2010) rationalises the inclusion of this advice point.

5.10.6. Communication adaptations (Figure 4, section 2.7.7.)

The components of '*give clear expectations, prompts and cues', 'incorporate augmentative communication strategies' 'encourage routine timing of all meals'* and

'*consider visually modelling routine using a doll*' received no stakeholder discussion but were all included based on adequate review evidence. Advice to '*ensure bedroom only used for sleep and calm activities*' was also included due to an adequate evidence base, however, stakeholders did express concerns with its acceptability. Emily and VFW voiced how adolescent children may struggle to achieve this, because their bedroom may be the only private place they can use when they are in need of solitary time. This highlights issues around customisation and implementation support when delivering SHE advice. The following extract demonstrates stakeholders' concerns around reserving the bedroom for sleep and calm activities:

> "*That bedroom is only used for sleep would not happen in my house*" (Emily)

> "*I think that particular issue would be a challenge for (..) especially teenagers , your bedroom needs to be a private place as well.*" (VFW)

In summary, the completed SHE tool incorporates the original six advice areas and 45 individual components for sleep practitioners to select from.

Table 25. Final SHE Tool.

1.Sleep timing	2.Bedtime routines	3.Behaviour management	4.Environment	5.Physiological	6. Communication adaptations
1.1. Set consistent bedtimes and wake times (including holidays and weekends)	**2.1.**Set relaxing routine	**3.1.** Ensure bedroom is not used a punishment setting	**4.1.** Ensure quiet noise levels at sleep times	**5.1**. Encourage healthy diet, limit fat and sugar intake.	**6.1.** Give clear expectations, prompts and cues.
1.2. Set age appropriate bedtimes	**2.2.** Discourage television or blue light emitting devices at bedtime. (consider blue light blocker sunglasses if resistant)	**3.2.** Avoid soothing to sleep with a bottle/breast after 6-12 months old. (#care re: weaning/feeding difficulties)	**4.2.** Consider room temperature 16-20c, bedding and sleep clothes to maintain comfortable body temperature	**5.2.** Encourage daily exercise (but avoid this late evening)	**6.2.** Incorporate augmentative communication strategies
1.3. Encourage age appropriate daytime napping	**2.3.**Consider alternative therapies and relaxation techniques	**3.3.** Incorporate rewards which are meaningful to the child.	**4.3.** Ensure a darkened bedroom (black-out blind)	**5.3.** Ensure child has plenty of light in the day	**6.3.** Encourage routine timing of all meals.
1.4 Avoid late afternoon napping	**2.4.** Limit bedtime rituals	**3.4.** Set and stick to limits	**4.4.** Ensure bedroom has a familiar layout and calm decoration.	**5.4.** Avoid smoking and alcohol	**6.4.** Consider visually modelling routine using a doll.
1.5 Avoid excessive time in bed	**2.5** Ensure routine activities are consistently ordered and timed	**3.5.** Ensure child falls asleep and sleeps alone in own bed	**4.5.** Allow security object to promote self-soothing	**5.5.** Light meals only near bedtime.	**6.5** Ensure bedroom is only used for sleep and calm activities.
	2.6. Ensure routine is 20-45 minute duration	**3.6.** Put child to sleep drowsy	**4.6.** Consider sensory sensitivities of the child	**5.6.** Limit caffeine intake	
		3.7. Give minimal interactions during night time feeds and night awakenings	**4.7.** Ensure bed is comfortable (consider sleep systems)	**5.7.**Ensure child uses toilet before bed	
		3.8. Encourage child to think about problems/plans before going to bed	**4.8.** Remove or hide stimulating toys in bedroom	**5.8.** Encourage milk and eat tryptophan/melatonin rich foods with complex carbohydrates at suppertime.	
			4.9. Use nightlight or red modelling bulb if preferred	**5.9.** Ensure child's individual hydration needs are met	
				5.10. Avoid blackcurrant juice in the evenings	

5.11. Conclusion.

This chapter has presented the findings of the co-design study. An overview and discussion of stakeholder constructions of how SHE should be delivered has been provided. In addition, a co-designed SHE tool for children with DD has been presented. The following chapter now explains how scoping review, exploratory and co-design study findings were synthesised to produce a programme theory to underpin the SHE tool.

CHAPTER 6. SYNTHESIS OF FINDINGS.

6.1 Introduction

This chapter describes the synthesis of findings from the scoping review, exploratory and co-design studies and methods employed to develop a programme theory and logic model which explains how SHE is supposed to work to improve sleep. Firstly, a detailed description will be given of how a theory of change shown as an outcomes chain was developed. The development of the theory of action presented as a programme theory matrix will then be described. This chapter concludes with the presentation of a logic or conceptual model of the combined theory of change and action (programme theory) which also represents emerging mid-range and novel theoretical underpinnings.

6.2. Programme theory development.

Guided by the work of Funnel and Rogers (2011), scoping review, exploratory and co-design study findings were synthesised to produce a programme theory consisting of a theory of change and action as follows.

6.2.1. Development of the theory of change.

The important features of a theory of change were drawn out from synthesising review, exploratory and co-design study findings. As guided by Funnell and Rogers (2011) such inquiry methods help to further clarify what is known about the problem under investigation, its causes and consequences. In developing the theory of change, the decision was taken to consider the six discussion themes (Table 18) as problems,

causes and consequences and reframe them as intended outcomes for a SHE intervention as follows:

- The problem is parents, generic practitioners and sleep practitioners do not agree on what a sleep problem is. The intended outcome is they develop a shared understanding of sleep problems.

- The problem is parents and generic practitioners do not know where to get sleep help. The intended outcome is sleep services are well publicised and accessible for parents.

- The problem is children with DD have sleep problems. The intended outcome is regularity and quality of sleep improves for the child.

- The cause of sleep problems is a lack of understanding of the sleep problem. The intended outcome is sleep practitioners and parents improve their understanding of the sleep problem.

- The problem is parents do not feel safe and supported by sleep practitioners. The intended outcome is parents and sleep practitioners develop a safe and supportive relationship.

- The consequence of children's sleep problems is reduced quality of life for the child and family. The intended outcome is quality of life improves for the child and family.

These outcomes were then plotted onto an outcomes chain (Figure 17) (the centrepiece of a developing programme theory) and displayed in a logical sequence, by establishing high and low level outcomes as end points. To show how the achievement of outcomes may not always be sequential and how vicious circles

often apply to SHE programmes, feedback loops were also incorporated into the chain. These feedback loops were depicted by dotted lines in the outcomes chain, although it is acknowledged that not all possible examples of when parents may disengage may be represented.

Funnell and Rogers also recommend that the boundaries of the programme are established by systematically focusing and scoping outcomes. This means grading outcomes in terms of how much they can be directly attributed to the intervention, to ensure programme results are not under or over claimed and ensures that intervention complexity and contextual factors are not overlooked. Outcomes were placed into one of three categories as follows (N.B. focus is a subcategory of total scope).

- **Direct focus outcomes:** The direct result of programme effort :

  'Sleep services are well publicised and accessible for parents', 'Parents, generic practitioners and sleep practitioners improve their understanding of the sleep problem' and *'Regularity and quality of child's sleep improves'*.

- **Within scope but of focus outcomes:** Not within the direct influence of programme effort but within scope :

  'Parents and practitioners have a shared understanding of what a sleep problem is and *'Parents and sleep practitioners develop a safe and supportive relationship'*

- **Out of scope outcomes:** Way beyond the scope of the programme:

  'Quality of life improves for the child and family.'

The process of scoping and reviewing outcomes was helpful in showing how much outcomes could be credited to SHE, for example recognising that improved quality of life may happen as a positive side effect rather than a direct result of SHE. It also acknowledged the important but less explicit contributions of developing a shared understanding of the sleep problem and a supportive relationship with parents which highlighted additional issues (not the direct focus of the intervention) that sleep practitioners needed to address.

Figure 17: Proposed Outcomes Chain for SHE
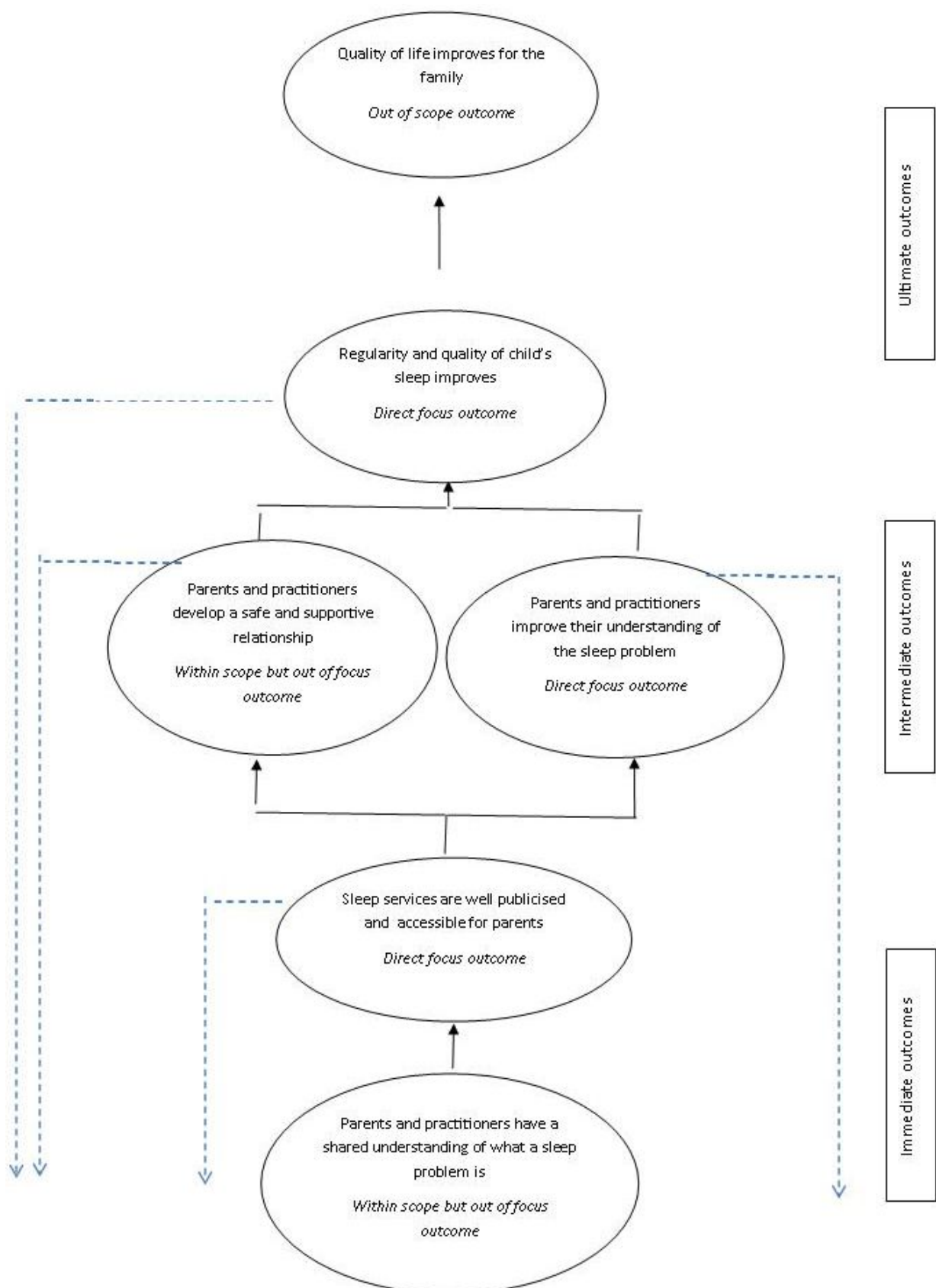
6.2.2. Development of the theory of action.

A theory of action explores what an intervention does to activate the outcomes espoused in a theory of change (Funnell & Rigers, 2011). It elucidates what activities the programme does and what resources it needs to bring about change whilst also clarifying the rationale for these operations. Evidence from the scoping review, exploratory and co-design study was used to populate the theory of action which is displayed alongside the theory of change in a matrix (Figures 18-24). The matrix incorporates the following components (Donabedian, 1966; Funnell & Rogers, 2011; Schurr et al., 2014):

Success criteria. This is a detailed description of the broadly defined outcomes in the outcomes chain. It defines the attributes and the success criteria for each outcome and what would represent effective intervention performance.

Programme factors describe what should occur to achieve these outcomes. These are factors which are mostly within the control of the programme and will affect the achievement of results. They describe how well programme activities should be carried out to achieve intended outcomes

Non-Programme factors are outside of the direct control of programmes but can affect the success of an intervention. These dimensions describe the wider context of programme delivery and highlight how professionals can attempt to manage the external risk factors which could hamper intervention success.

Activities, Processes and Principles describe the actual process or actions that make up the intervention. It includes everything the intervention does to produce desired outcomes and links are made between the activities and each outcome feature. Some activities are also about managing non-programme factors.

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<u>Resources and inputs (financial and human)</u> are the easily identifiable inputs required to carry out the programme such as training, staffing, funding, facilities and time. This dimension is also referred to as 'structure' or 'context' and describes the factors which control how sleep practitioners and parents act within an intervention.

<u>Outputs and throughputs</u> describes the direct outputs and general productivity that can occur if the activities are implemented as intended.

In combing the theory of change and theory of action, the matrix represents the proposed programme theory underpinning SHE. The matrix will now be presented.

.

Figure 18: SHE programme theory matrix –key.

Exploratory study themes:
Parent 1-'"*Well this is how it is" beliefs about sleep problems*.' Parent 1A- ' *"It was a battle" the challenges of sleep problems*.' Parent 1B- '*Resignation and acceptance*.' Parent 1C ' *"The sleep problems compound everything else" broader impacts*.'

Parent 2- '"*The biggest problem is knowing where to get the help" getting professional help*.' Parent 2 A '"*They couldn't quite understand the severity of it" validation of sleep concerns*.' Parent 2B '"*Nobody's willing to take responsibility" challenges around profession*al *responsibility*.'

Parent 3 '*Ways of knowing about sleep*.' Parent 3A '*Understanding sleep problems*.' Parent 3B '*Forms of coping*.'

Parent 4 '"*I'd love Supernanny just come and spend some time with us" visions of sleep support*.' Parent 4A ' *"Their answer was sleeping tablets and I refused" disappointment with medication*.' Parent 4B ' *"You just tick off the boxes" the need for customised support*.' Parent 4C '"*At home you can just relax more" the importance of place*.'

Practitioner 1 –'*Sleep service accessibility*.' Practitioner 1A '*Expectancies and awareness of sleep help'*. Practitioner 1B ' *"Some families seem to fall through the net" negotiating referral systems*.'

Practitioner 2- '*Inconsistent sleep support'* Practitioner 2A '*Nature of assessment*.' Practitioner 2B '*Nature of treatment*.'

Co- design event findings: CD1=Co-design event 1.CD2= Co-design event 2. CD3-=Co-design event 3

Scoping review findings= references coloured blue.

Figure 19: SHE programme theory matrix – Parents and practitioners have a shared understanding of what a sleep problem is.

Outcomes Chain (Outcomes)	Success Criteria	Programme factors (Context)	Non-Programme factors (Context)	Outputs and Throughputs	Activities, Processes, Principles (Process)	Resources and Inputs- Financial; Human.(Context)
Parents and practitioners have a shared understanding of what a sleep problem is. *Within scope but out of focus outcome.*	Child has problems falling asleep, staying asleep, waking too early, sleeping too much or disturbed episodes interfering with sleep (Stores, 2009) Problems have occurred at least 3 nights a week for at least 3 months (Montgomery et al., 2004) Practitioners and parents <u>recognise</u> the child has sleep problems and is in need of help. Parent 2A, Practitioner 1A Agreement: Sophia, Fadheela, Jenny CD(1), Agreement: VSP(2), SFSO, Amy, Jenny CD(2) Emily,Gloria, Clarissa, VSS(3)CD(3) All children are <u>screened</u> for sleep problems Agreement: Fadheela, Gloria, Emily, Jenny.CD(1), Jenny, Amy, Sharon, SFSO, VDO, VSP(2) VFW(2)CD(2)	**Whether:** Generic and sleep practitioners have a consistently positive attitude towards <u>recognising</u> sleep problems and raise parents' expectations that sleep can improve. Parent 2A Practitioner 1A) Agreement: Sophia, Fadheela, Jenny CD(1), Agreement: VSP(2), SFSO, Amy, Jenny CD(2) In the <u>screening</u> process all practitioners ask parents about sleep in a non-judgemental way. Amy, Gloria, Fadheela, CD(1). *"Often what happens for all of us is our parenting is brought into question"* Amy, CD(1)	**Whether:** Parents have come to accept their child's sleep problem as "normal" and <u>recognise</u> the sleep problem Parent 1B.Agreement: Fadheela, Gloria., Sharon, CD(1) Parent gives priority to sleep against child's other multiple needs and <u>recognises</u> the sleep problem Fadheela , Emily CD(1) Emily CD(3) *"It's not always going to be the first thing on your mind when you are trying to battle for lots of other things"* Fadheela CD(1) Parent has literacy problems /no internet access preventing them from <u>screening</u> via PCHR or having sleep <u>awareness</u> raised. Amy CD(1) Parent is shy to socialise with other parents at pre-school groups where they may have their <u>awareness</u> of sleep problems raised Gloria, Fadheela CD(1)	Children's sleep problems and requests for help are <u>recognised</u> by parents and practitioners. Parent 2A Practitioner 1A Agreement: Sophia, Fadheela, Jenny CD(1), Agreement: VSP(2), SFSO, Amy, Jenny CD(2), Parents acknowledge their child has a sleep problem through <u>screening</u> questions from generalist and specialist practitioners and early years practitioners.: Fadheela, Gloria, Emily, Jenny (CD1) Agreement: Jenny, Amy,Sharon, SFSO, VFW (2) CD(2) Disagreement:VSS(3)CD(3). Parents <u>screen</u> for problems through checking PCHR. Fadheela, Gloria, Emily, Jenny CD(1), Disagreement: SFSO (CD2), Agreement Amy, VDO CD(2) Parents become <u>aware</u> that their child has a sleep problem through media campaign Sharon, Jenny, Sophia, (CD1), Jenny, Amy, VFW (2) CD(2)	Generic and sleep practitioners listen to parents take sleep concerns seriously and <u>recognise</u> sleep problems. Parent 2A Practitioner 1A Agreement: Sophia, Fadheela, Jenny CD(1), Agreement: VSP(2), SFSO, Amy, Jenny CD(2) Generic and sleep practitioners <u>screen</u> for sleep problems and actively ask ALL parents about sleep problems. Fadheela, Gloria, Emily, Jenny.CD(1). Agreement: Jenny, Amy, Sharon, SFSO, VDO, VSP(2) VFW(2) CD(2)Development And early years practitioners. Jenny, Amy, VFW(2) CD(2) *"Talk with the children about what happens when they go to sleep (..) maybe the nursery school teachers can pick up on something."* Jenny CD(2)	Financial and human resourcing to train generic and sleep practitioners in <u>recognising</u> sleep problems Parent 2A, Practitioner 1A Agreement: Sophia, Fadheela, Jenny CD(1), Agreement: VSP(2), SFSO, Amy, Jenny CD(2) Financial and human resourcing to train generic and sleep practitioners to <u>screen</u> for sleep problems. Fadheela, Gloria, Emily, Jenny (CD1),Agreement: Jenny, Amy, Sharon, SFSO, VDO, VSP(2), VFW(2) CD(2) Development And early years practitioners Jenny, Amy, VFW(2) CD(2), VFW VSP(2)Clarissa CD(3) Disagreement: VSS(3) CD(3). Financial and human resourcing of creating PCHR sleep <u>screening</u> tools Fadheela, Gloria, Emily, Jenny (CD1) Amy, VDO CD(2). Disagreement: SFSO CD(2)

	There is a good public <u>awareness</u> of sleep problems in children with DD Sohpia, Jenny, Sharon, CD(1). Jenny, Amy, VFW(2) CD(2)VFW, VSP(2) CD(3)		Parent is afraid to admit & <u>recognise</u> their child has a sleep problem (Gloria, Amy, Emily CD(1). Development: Parent feels judged by practitioners and has poor confidence: Amy,Sharon, VSP(2) CD(2). Gloria , Clarissa, VSS(3) CD(3) Parents have adopted coping strategies such as co-sleeping, which prevent them from <u>recognising</u> sleep problem Parent 3B.:VSS(3), Gloria, CD(3) Parent partners and relatives agree & <u>recognise</u> there is a sleep problem Jenny, VDO (CD(2). Agreement Emily CD(3)	*"Professional goes in and sees them in school "oh they are alright in school" (..) it's all pointing back to the parent and then you lose confidence"* Amy CD(2) *"We can just have that conversation what do you want to happen? And they can come back to me when they are ready"* SFSO CD(2)	Policy makers run a media campaign raising <u>awareness</u> on sleep , Sharon, Jenny, Sophia (CD1) Jenny, Amy, VFW(2) CD(2) In <u>recognising</u> sleep problems all practitioners openly discuss with parents what they consider to be a sleep problem. Practitioner 1A Agreement: VSP(2), VDO, Sharon, Amy, SFSO CD(2) Clarissa, VSS(3) CD(3) Parents can refer to sleep checklist in PCHR, to <u>screen</u> for children's sleep problems. Fadheela, Gloria, Emily, Jenny CD(1)(CD2), Agreement:Amy, VDO,CD(2). Disagreement: SFSO CD(2)	Financial and human resourcing of raising public <u>awareness</u> of sleep and sleep problems (media, facebook, leaflets) Sohpia, Jenny, Sharon, CD(1). Jenny, Amy, VFW(2), CD(2) *"There was all that talk about how everybody needs exercise so maybe people should realise it's as important to get enough sleep"* Sharon (CD1) *Every child is different and some children take longer to fall into their sleep patterns."* SFSO CD(2)

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6.2.3. Narrative summary of 'Practitioners and parents have a shared understanding of what a sleep problem is.' (Figure 19).

This immediate, pre-implementation, within scope but out of focus outcome of SHE describes how a common understanding should be developed between parents and practitioners of what constitutes a sleep problem in a child with DD. Scoping review evidence firstly broadly defines sleep problems as problems falling asleep, staying asleep, waking too early, sleeping too much or disturbed episodes interfering with sleep (Stores, 2009) which have occurred at least three nights a week for at least three months (Montgomery et al., 2004). Success criteria are then described by three categories: the successful recognition of sleep problems, successful sleep screening and a good public awareness of sleep, which will now be presented.

The SHE intervention facilitates the recognition of sleep problems in children with DD by parents and practitioners. This is achieved through training generic and sleep practitioners in sleep problem recognition, and practitioners adopting a positive attitude towards sleep, listening and openly discussing with parents the nature of the sleep problem. This can help parents to: prioritise and admit the sleep problem without feeling judged, address coping strategies such as co-sleeping which can mask the problem, avoid normalising the sleep problem and feel supported when other family members do not agree there is a sleep problem. Some parents however, may still struggle with these non-programme factors, and will deny or delay recognition of their child's sleep problem; this will be beyond the control of the SHE intervention.

The intervention also aims to screen children with DD for sleep problems through screening all children as standard practice. This is achieved through training early

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years, generic and sleep practitioners to sensitively screen for children's sleep problems and encouraging more frequent conversations about sleep between parents and the professionals they come into contact with to ensure the early detection of sleep problems. It is important that parents of children with DD do not feel singled out during this screening process and that this is a standard procedure carried out for all children. This helps to 'normalise' inquiries about sleep problems for parents and influences their receptiveness to acknowledging the existence of sleep problems. The need to be aware also that some parents may be uncomfortable discussing the personal issue of their child's sleep is acknowledged. In addition, it is posited that the development of PCHR sleep screening tools empower parents to independently identify sleep problems; however, they may be misleading for some children whose patterns of sleep development are outside the norm. Furthermore, it is acknowledged that some parents may not benefit from the option of self-screening if they have literacy issues and practitioners need to be mindful of their heightened need to receive formal sleep screening.

Lastly, the intervention works to promote a good public awareness of sleep problems in children with DD. The aim is for parents to become aware that their child has a sleep problem through media awareness raising campaigns resourced by policy makers. Increasing public awareness of the importance of sleep through health promotion slogans, posters, leaflets and social media for example can help most parents identify if their child has a sleep problem; however, it is again acknowledged that those with literacy difficulties may not fully benefit from such a campaign. Once a child's sleep problem has been identified, agreed on and acknowledged by parents and practitioners, a search for sleep support can then be instigated which leads onto

the next immediate outcome of '*Sleep services are well publicised and accessible for parents*'.

Figure 20: SHE Programme theory matrix- Sleep services are well publicised and accessible for parents.

Outcomes Chain (outcomes)	Success criteria	Programme factors (context)	Non-programme factors (context)	Outputs and throughputs	Activities, processes, principles (process)	Resources and inputs (Financial, human (context)
Sleep services are well publicised and accessible for parents *Direct focus outcome*	Adequate sleep services with trained sleep practitioners to support children aged 0-18, with any DD and sleep problems wherever they live. Parent 2B, Practitioner 1B Agreement: Fadheela, Amy, CD(1),VSS(3), VFW CD(3). Parents can source sleep help through a generic sleep service. Fadheela, Amy CD(1). Emily CD(3) Challenge: Generic practitioners don't understand DD: .VFW, VSS(3), Clarissa CD(3) Parents and generic practitioners have a good awareness of sleep services through publicity activities. Parent 2B, Practitioner 1A. Agreement: :Sharon, Emily, Gloria, Sophia CD(1) Parents can easily access sleep teams in a timely manner. Parent 2B. Practitioner 1B Agreement: Emily,Gloria, Fadheela CD(1) VDO CD(2)	Whether: Adequate sleep services are available with trained sleep practitioners are available to meet the needs of children aged 0-18, with any DD and sleep problems wherever they live. Parent 2B. Practitioner 1B Agreement: Fadheela, Amy,CD(1),VSS(3),vFWCD(3). There is agreement and commitment amongst policy makers to provide a generic sleep service Fadheela , Amy, CD(1)Emily CD(3)Challenge: Generic practitioners don't understand DD: .VFW, VSS(3), Clarissa CD(3) Sleep teams are committed to actively publicising their service to parents and generic practitioners. Parent 2B, Practitioner 1A. Agreement: :Sharon, Emily, Gloria , Sophia CD(1). Sleep teams understand that parents need to easily access their service Parent 2B. Practitioner 1B Agreement: Emily,Gloria, Fadheela CD(1) VDO CD(2)	Whether: Generic practitioners engage with sleep service publicity. Parent 2B. Agreement: Jenny, CD(1) VSS(3), CD(3 Parent has literacy problems/no internet access preventing them from engaging with sleep service publicity Amy CD(1) Parents are afraid of accessing specialist sleep teams because of low confidence Sharon CD(1) *"I've not got in touch yet it's a bit scary getting in touch"* Sharon, CD(1) *"The worst thing for parents I think would be going to a generalist sleep advisor and then having explain what the condition the child has"* VFW CD(3)	Parents have the prospect of having children's sleep problems addressed through adequate sleep services for children aged 0-18, with any DD and sleep problem wherever they live, Parent 2B, Practitioner 1B Agreement: Fadheela, Amy, CD(1),VSS(3), VFWCD(3). Parents source sleep help through a generic sleep service. Fadheela, Amy CD(1). Emily CD(3) Challenge: Generalist practitioners don't understand DD: .VFW, Gloria, VSS(3) CD(3) Parents become aware of sleep services and how to access them through publicity activities Parent 2B, Practitioner 1A Agreement: Sharon, Emily, Gloria, Sophia CD(1) Parents can easily access sleep services. . Parent 2B. Practitioner 1B Agreement: Emily,Gloria, Fadheela CD(1) VDO CD(2)	Adequate sleep services children aged 0-18, with any DD and sleep problems wherever they live. Parent 2B Practitioner 1B Agreement: Amy, Sophia CD(1),VSS(3)VFWCD(3). Parents are offered a generic sleep service. Fadheela, Amy CD(1). Emily CD(3) Challenge: Generalist practitioners don't understand DD: .VFW, VSS(3), Clarissa, CD(3) Sleep services are publicised on a website and listed in an on-line directory. Practitioner 1A Sharon, Emily, Gloria , CD(1) Sleep services actively publicise services through posters and leaflets. Parent 2B, Practitioner 1A .Sophia CD(1) Direct referrals for ease of access . Parent 2B. Practitioner1B.AgreeFadheela CD(1) VDO CD(2) Short waiting times for ease of access. Parent 2B. Practitioner 1BAgreement: Emily,Gloria,SophiaCD(1)	Financial and human resourcing of adequate sleep services with trained sleep practitioners to meet the needs of all children with DD. Parent 2B, Practitioner 1B.Agreement: Amy, Sophia CD(1) , VSS (3),VFW CD(3) Financial and human resourcing of a generic sleep service Fadheela , Amy, CD(1)Emily CD(3) Challenge: Generic practitioners don't understand DD: .VFW, VSS(3), Clarissa, CD(3) Financial and human resourcing of publicising available sleep services. Parent 2B, Practitioner 1A. Agreement: Sharon, Emily, Gloria , Sophia CD(1). Financial and human resourcing of sleep teams which are easily accessible with short waiting times. Parent 2B. Practitioner 1B Agreement: Emily,Gloria, Fadheela CD(1) VDO CD(2)

6.2.4. Narrative summary of 'Sleep services are well publicised and accessible for parents.' Figure 20.

This immediate, pre-implementation, direct focus outcome of SHE effort describes how available sleep services should be actively publicised and easily accessible to parents. Success criteria is organised into four categories: the provision of adequate sleep services, provision of generic sleep services, good accessibility of sleep services and active publicity of sleep services which will now be presented in turn.

The SHE intervention aims to provide adequate specialist sleep services to meet the needs of families of children with DD. The output of parents having their children's sleep problems addressed whatever the child's type or level of DD, age and wherever they live is achieved through the appropriate funding and resourcing of sleep services to meet family's needs. This incorporates the need for specialist sleep services to offer good geographical coverage and broad referral criteria which does not exclude children on the basis of age or type of DD.

The intervention also works to provide a generic sleep service, for parents to start their engagement with practitioners through. This is achieved through a resourcing commitment from policy makers. When parents first approach generic practitioners for help, it is important they can 'normalise' the sleep problem by accessing a mainstream sleep team first, so they do not feel different from other parents. If needed, signposting to specialist sleep services can occur afterwards. The option of a generic team was identified as significant in bringing about the change of parents deciding to access the sleep help that they need. However, the need for mainstream practitioners within generic teams to have a good understanding of the specific needs

of children with DD was highlighted as a contextual factor that could affect potential success.

The intervention also aims for specialist sleep services to be easily accessible for parents. This is achieved by services accepting direct referrals from parents rather than needing the endorsement of gatekeeping professionals which helps to keep the access route simple. In addition, sleep teams should offer support in a timely way for families and operate only short waiting lists that fall within national guidelines. It is acknowledged that some parents may still be afraid to access specialist sleep teams and practitioners should be mindful of being as approachable as possible to attempt to counteract this contextual, external factor.

Finally, the intervention works to ensure specialist sleep services are widely publicised to raise awareness amongst parents and generic practitioners. This is achieved through the appropriate funding of publicity initiatives which promote sleep teams on websites, on-line directories, posters and leaflets for example. Non-programme factors which could affect the success of such activities include reluctance from generic practitioners to engage with sleep service publicity, and parents with literacy problems who may not be able to access the promotional literature. These elements may be largely outside of the intervention's control, but it is remains important to be aware of them. When parents are aware of well resourced, available sleep services and can easily access them, this leads to the next two intermediate outcomes which can be addressed simultaneously: '*Parents and practitioners develop a safe and supportive relationship*' and '*Parents and practitioners improve their understanding of the sleep problem*'.

Figure 21. Programme theory matrix –Parents and practitioners develop a safe and supportive relationship.

Outcomes Chain (Outcomes)	Success Criteria	Programme factors (Context)	Non-programme factors (Context)	Outputs and Throughputs	Activities, Processes , Principles (Process)	Resources and Inputs- Financial; Human.(Context)
Parents and sleep practitioners develop a safe and supportive relationship ***Within scope but out of focus outcome.*** *"She just put me at ease, she reassured me it wasn't my fault"* Jenny, CD(1)	Parents feel sleep practitioners use <u>good interpersonal skills</u> throughout their contact with them (Beresford et al., 2012a, 2012b) Parent 2A, practitioner 2A and 2B.Agreement: Amy, Jenny,Gloria CD(1) Parents feel they have received the amount of <u>support</u> they need throughout their contact with services. (Adkins, Molloy et al., 2012; O'Connell & Vanaan, 2008) Parent 4C, Practitioner 2B. Agreement: Gloria, Jenny, Sharon CD(1), *"The biggest thing you need to hear is that you're not a bad parent"* Jenny, CD(1)	**Whether:** Sleep practitioners have developed <u>good interpersonal skills</u> and use these with parents (Beresford et al.,2012a, 2012b).Parent 2A, practitioner 2A and 2B.Agreement: Amy, Jenny,Gloria CD(1) Sleep practitioners are committed to delivering a continuity of contact so parents feel well <u>supported</u> Jenny, Sharon, Fadheela, Amy CD(1) Sleep practitioners are commissioned to offer the <u>support</u> parents need (Adkins, Molloy et al.,2012; O'Connell & Vanaan, 2008) Parent 4C, Practitioner 2B. Agreement: Gloria, Jenny, Sharon CD(1), Sleep practitioners are supported and committed to offering home visits so parents feel <u>supported</u> (Beresford et al., 2012b). Parent 4C.Practitioner 2A and 2B.	**Whether:** Parents perceive sleep practitioners have a lack of empathy and feel they do not have <u>good interpersonal skills</u> Parent 1. Parents have had previous positive experiences with practitioners in terms of <u>good interpersonal skills</u> (Bartlet & Beaumont, 1998) *"We should see the same person each time"* Jenny CD(1) *"I didn't see her as many times as I would have liked"* Gloria CD(1)	Parents receive sleep practitioner input that is delivered using <u>good interpersonal skills</u> (Beresford et al., 2012a, 2012b) Parent 2A, practitioner 2A and 2B. Agreement: Amy, Jenny Gloria, CD(1) *"Building a relationship with parents(..) make that parent feel safe"* Amy CD(1) Parents are <u>supported</u> by the same sleep practitioner continuously. Jenny, Sharon, Fadheela, AmyCD(1) Parents are offered the amount of <u>support</u> they need throughout their contact with services. (Adkins, Molloy et al., 2012; O'Connell & Vanaan, 2008) Parent 4C, Practitioner 2B. Agreement: Gloria CD(1) Parents are offered home visits to ensure they feel safe and <u>supported</u>. (Beresford et al., 2012b). Parent 4C.Practitioner 2A and 2B	Sleep practitioners demonstrate <u>good interpersonal skills</u> when supporting families with sleep. (Beresford et al., 2012a, 2012b) Parent 2A, practitioner 2A and 2B. Agreement: Amy, Jenny, Gloria CD(1) Services provide continuity of sleep practitioner contact to ensure parents feel <u>supported</u>. Jenny, Sharon, Amy, Fadheela CD(1) Sleep practitioners offer the amount of <u>support</u> the parent needs during implementation (number of home visits, phone calls (Adkins, Molloy et al., 2012;O'Connell & Vanaan, 2008) Parent 4C, Practitioner 2B. Agreement: Gloria CD(1) Services offer home visits to ensure parents feel safe and <u>supported</u>. (Beresford et al., 2012b). Parent 4C.Practitioner 2A and 2B.	Financial and human resourcing to ensure sleep practitioners are trained to develop <u>good interpersonal skills</u> (Beresford et al., 2012a.2012b). Parent 2A, practitioner 2A and 2B. Agreement: Amy, Jenny, Gloria CD(1) *"She did it very positive, it kept him hoping"* Jenny CD(1) Financial and human resourcing to facilitate sleep practitioners in giving parents the <u>support</u> they need throughout their contact. (Adkins, Molloy et al., 2012; O'Connell & Vanaan, 2008).Parent 4C , Practitioner 2A, 2B. Jenny, Sharon, Amy, Fadheela, Gloria CD(1).

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6.2.5. Narrative summary of 'Parents and sleep practitioners develop a safe and supportive relationship'. Figure 21.

This intermediate, within scope but out of focus outcome of SHE effort describes the need for parents and sleep practitioners to develop a trusting working relationship before parents feel able to follow SHE advice. Success criteria is organised into two categories: sleep practitioners use good interpersonal skills with parents throughout their contact, and parents receive the amount of support they need throughout their contact, which will now be presented in turn.

The SHE intervention works to ensure sleep practitioners demonstrate good interpersonal skills when working with parents. This is achieved through the resourcing of training sleep practitioners to develop these skills and utilising them during their contact with parents. Parents feel it is important that sleep practitioners show an appreciation of their situation, communicate sensitively and offer reassurance, which creates confidence in the practitioner's ability to support the family and give sound advice. It is acknowledged that some sleep practitioners may be less approachable and understanding than others and some parents may perceive them to have a lack of empathy. Furthermore, some parents may have developed a mistrust of sleep practitioners due to previous negative experiences of care. These contextual factors may affect the success of this outcome and it is important sleep practitioners are aware and make attempts to manage them.

The intervention also aims to provide parents with the amount and type of support they need throughout their contact with sleep services. This is achieved through the adequate funding of services so they have the time and resources to offer parents the level of support they need to engage with SHE. This incorporates parents

experiencing continuity in the sleep practitioner support offered, so that advice is consistent and they do not have to develop additional working relationships with new practitioners. In addition, parents should be offered home visits as part of the intervention because this is where they feel most comfortable and safe discussing the issue of sleep. The quantity of support required by parents in terms of number of home visits or phone-calls should also be driven by parents' need. Programme factors which may affect success are whether sleep practitioners are committed to delivering a continuity of contact, the level of support required by individual families and offering home visits as an alternative to clinic appointments. These contextual factors require acknowledgement and should be managed to ensure success. The development of a safe and supportive relationship can take time and this can be built whilst the sleep problem is being assessed which is described in the next outcome '*Parents and sleep practitioners improve their understanding of the sleep problem'*.

Figure 22: Programme theory matrix- Parents and sleep practitioners improve their understanding of the sleep problem.

Outcomes Chain	Success Criteria	Programme factors	Non-programme factors	Outputs and Throughputs	Activities, Processes , Principles	Resources and Inputs- Financial; Human.
Parents and sleep practitioners improve their understanding of the sleep problem. *Direct focus outcome*	Parents and sleep practitioners understand the nature and causes of the sleep problem through a comprehensive sleep assessment Parent 4C, Practitioner 2A Agreement Fadheela, Emily, Amy, CD(1) SFSO, VSP(2), Jenny, Amy CD(2) Parents and sleep practitioners understand the nature and causes of the sleep problem through a competent sleep assessment ((Beresford et al., 2012b, 2012g), Practitioner 2A. Agreement VSP(2), SFSO. CD(2) Parents and sleep practitioners understand the nature and causes of the sleep problem through a supportive sleep assessment process . Parent 3A, Practitioner 2A	**Whether:** Sleep practitioners have an effective working relationship with other members of the multi-disciplinary team to ensure a comprehensive sleep assessment. Fadheela, CD(1) VSP(2), Amy CD(2) Sleep practitioners overlook co-occurring conditions which may explain the sleep problem and fail to complere a supportive sleep assessment.(Malow et al, 2013) Practitioner 2A. Sleep practitioners are supported to invest time in a supportive sleep assessment process for parents. Parent 3A, Practitioner 2A Sleep practitioners understand the need to offer a supportive sleep assessment to parents. Parent 3A, Practitioner 2A	**Whether:** Parents give assessment information honestly and accurately ensuring a comprehensive sleep assessment. Jenny CD(2) *"There could be other things underlying that are going on, that maybe they wouldn't talk to a professional"* Jenny CD (2) The parent has a positive attitude toward the sleep assessment process and engages ensuring a comprehensive and competent sleep assessment Practitioner 2A Parents have had positive experiences in past of practitioners and engages in a comprehensive and competent sleep assessment. (Bartlet & Beaumont, 1998) The sleep assessment is clouded by medication and skews a competent assessment. Practitioner 2A	Parents are visited at home as part of a comprehensive sleep assessment process Parent 4C, Practitioner 2A Agreement Fadheela, Emily, Amy, CD(1) *"Come into my home and work with me, learn about my family and how things function"* Amy CD(1) Development: Children are observed at bedtime by sleep practitioners Fadheela, Emily, Amy, CD(1) *"Could they possibly come to your house at bedtime?* Amy CD(1) Parents receive multiple sessions with sleep practitioners to ensure a comprehensive sleep assessment process. Practitioner 2A. Agreement Fadheela CD(1). Amy,Jenny CD(2) *"I'd rather they took the time to get to know the child"* Jenny CD(2)	Sleep practitioners assess the child at home to complete a comprehensive sleep assessment .Parent 4C, Practitioner 2A Agreement Fadheela, Emily, Amy, CD(1) Development: Sleep practitioners need to observe the child at bedtime Fadheela, Emily, Amy, CD(1) Sleep practitioners need to perform multiple sessions to complete a comprehensive sleep assessment. Practitioner 2A. Agreement Fadheela CD(1), Jenny, Amy CD(2) *"Not just one session (.) how they make conclusions about children based on 40 minutes"* Fadheela , To complete a comprehensive sleep assessment, sleep practitioners involve other members of the multi-disciplinary team and partner organisations. Gloria, Amy, Fadheela, CD(1), VSP(2), Amy CD(2)	Financial and human resourcing of sleep practitioner's time to complete a comprehensive sleep assessment. Parent 3A, Practitioner 2A. Agreement Fadheela, Emily, Amy, CD(1) SFSO, VSP(2),Jenny, Amy CD(2) Financial and human resourcing of sleep training for practitioners to ensure they can complete a competent sleep assessment (Beresford et al., 2012b, 2012g), Practitioner 2A. Agreement VSP(2), SFSO CD(2) Financial and human resourcing for sleep practitioners time to offer a supportive sleep assessment process to parents.. Parent 3A, Practitioner 2A *"All of these multi agency people that need to be part of the assessment"* Fadheela, CD(1)

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| | | | | Other members of the multi-disciplinary team and partner organisations are involved in the <u>comprehensive sleep assessment</u> process. Gloria, Amy, Fadheela, CD(1)SFSO, VSP(2) CD(2)

Children receive a <u>competent sleep assessment</u> and are screened for co-morbidities and have their sleep problems assessed using sleep histories, sleep diaries and validated outcome measuresAdkins, Molly et al,2012; Cortesi et al, 2012. Parent 3A Practitioner 2A

Sleep assessment findings are interpreted in partnership with the family to achieve a <u>competent sleepassessment</u>Practitioner 2A.Agreement SFSO, VSP(2), CD(2)

Parents receive <u>a supportive sleep assessment</u> are helped to complete sleep assessment documentation and are offered a variety of sleep recording methods Parent 3A Practitioner 2A | To perform a <u>competent sleep assessment</u> sleep practitioners screen for co-morbidities, utilise sleep histories, sleep diaries and validated sleep outcome measures. Adkins, Molly et al,2012; Cortesi et al, 2012. Parent 3A. Practitioner 2A

To perform a <u>competent sleep assessment</u> sleep practitioners interpret sleep assessment findings in partnership with parents and identify causes/strengths (psychological formulation)Practitioner 2A.Agreement SFSO, VSP(2) CD(2).

To perform a <u>supportive sleep assessment</u> parents receive guidance in completing sleep assessment documentation and are offered a variety of sleep recording methods Parent 3A Practitioner 2A | *"I always say to parents it's a process of elimination it's like a jigsaw puzzle"* VSP(2) CD(2) |

6.2.6. Narrative summary of 'Parents and sleep practitioners improve their understanding of the sleep problem.' Figure 22.

This intermediate, direct focus outcome of SHE effort describes the need for parents and sleep practitioners to improve their understanding of the nature and causes of the child's sleep problem. Success criteria is organised into three categories: a comprehensive sleep assessment is completed, a competent sleep assessment is completed and a supportive sleep assessment process is facilitated which will now be described in turn.

The SHE intervention works to provide a comprehensive assessment of the child's sleep problems through the adequate resourcing of sleep practitioner's time to observe the child at home at bedtime, perform multiple assessment sessions and liaise with other members of the multi-disciplinary team (MDT) and partner organisations. This helps parents to feel that sleep practitioners have invested sufficient time getting to know their child and will be basing their advice upon evidence collected. The need for sleep practitioners to have a good working relationship with other MDT members and partner organisations to elicit the required information is acknowledged as a programme factor that could affect success. Non-programme contextual factors which could also influence success are whether parents give assessment information honestly, positively engage in sleep assessment activities such as sleep diary completion and have had past, positive experiences of sleep practitioner support. These non-programme factors may be largely outside the control of the intervention, however, it is still important to acknowledge how they could hinder achievement of intended outcomes.

The SHE intervention works to provide a competent sleep assessment through the financial and human resourcing of training sleep practitioners in the sleep assessment process. This prompts sleep practitioners to firstly organise screening for physical and psychological co-morbidities, which may be causing the sleep problem. If such co-occurring conditions are over-looked this can affect the success of the SHE intervention. Sleep training also equips sleep practitioners with the skills to utilise sleep histories, sleep diaries and validated outcome measures to uncover multiple causal factors, understand the nature and establish a baseline recording of the sleep problem. Sleep practitioners will also be skilled at interpreting sleep assessment findings in partnership with parents and using a psychological formulation to summarise the child's strengths and sleep problem causes. Non-programme factors which could hinder success are the ability and willingness of parents to engage with the sleep assessment process and whether the child is already taking sleep medication such as melatonin which could cloud the findings. These factors may be largely outside the control of sleep practitioners; however, it is important to be explicit about external elements which could affect intervention success.

Finally, the intervention works to provide a supportive sleep assessment process through the resourcing of sleep practitioner's time, to enable them to offer guidance to parents in completing sleep assessment documentation. This may include advising parents how to complete sleep diaries or providing alternative methods of recording sleep information using visual prompts or easy read versions. Whether sleep practitioners appreciate the need to invest time supporting parents through the sleep assessment process is acknowledged as a programme factor which may affect intervention success. In summary, achieving an improved understanding of the sleep problem is a crucial step towards tailoring SHE advice and identifying the support

needs of families which feeds into the ultimate outcome of SHE '*Regularity and quality of child's sleep improves*'.

Figure 23: Regularity and quality of child's sleep improves.

Outcomes Chain	Success Criteria	Programme factors	Non-programme factors	Outputs and Throughputs	Activities, Processes , Principles	Resources and Inputs- Financial; Human.
Regularity and quality of child's sleep improves *Direct focus outcome*	Parents achieve set goals which determine an acceptable level of improvement in sleep rather than ideal sleep. (Beresford et al., 2012c, 2012d, 2012e; O'Connell & Vaanan, 2008). Agreement VSP(2) CD(3) *"The child is sleeping in some form that is right for that child"* VSP(2) CD(3) Improved sleep outcomes as measured by repeated sleep outcome measures (Adkins, Molloy et al., 2012; Cortesi et al., 2012: Malow et al., 2011) Practitioner 2B.Agreement SFSO , AmyCD(3). Parents receive effective sleep advice. (Beresford et al., 2012g). Parent 4B and 4C. Practitioner 2B.Agreement VSP(2), VDO CD(2) Agreement: VSS(3), VFW, VSP(2) CD(3)	**Whether:** Sleep practitioners deliver effective sleep advice in a non-directive, non-judgemental way. Parent 2A and 4B, Practitioner 2B Agreement VDO CD(2). *"I should be doing all this (..) and if you say you're not then you're not as good a parent, so it's that being judged"* VDO CD(2) Sleep practitioners have the time to re-run sleep outcome measures following implementation of effective sleep advice to measure sleep improvements.(Cortesi et al., 2012; Malow et al., 2011; Adkins, Molloy et al., 2012) Practitioner 2B. Agreement: Amy, SFSO CD(3) *"It just gets you down, there could be depression involved in that"* Gloria CD(3)	**Whether:** Parents understand and agree with the effective sleep advice and family follow it consistently (Beresford et al., 2012f, 2012g). Parent 4B Other stressful life events prevent acceptance of effective sleep advice. (Bartlett & Beaumont, 1998; Beresford at al., 2012g).Practitioner 2A. Parents have mental health issues affecting acceptance of effective sleep advice. (Beresford et al., 2012g). Parent 1A, Practitioner 2A Agreement Gloria CD(3). The home environment is conducive to sleep and the following of effective sleep advice. (Bereseford et al., 2012. Chapters 17 & 18). Agreement Jenny CD(2)	Parents receive customised effective sleep advice. (Beresford et al., 2012g). Parent 4B and 4C. Practitioner 2B. VSS(3), VFW, VSP(2) CD(3). Parents are given the rationale behind the effective sleep advice Practitioner 2B. Agreement VSP(2) CD(2) Parents are given psycho-education underpinning effective sleep advice (Beresford et al., 2012g) Practitioner 2B Agreement VSP(2) CD(2) The child's physical and psychological co-morbidities are addressed before effective sleep advice is given. (Adkins, Molly et al.,2012; Cortesi et al., 2012) Parent 3A. Practitioner 2A Sleep outcome measures are re-run following implementation of effective sleep advice Practitioner 2B. Agreement: SFSO CD(3)	Effective sleep advice is customised (Beresford et al., 2012g) Parent 4B and 4C. Practitioner 2B. Agreement:, Agreement VSP(2), VDO CD(2) VSS(3), VFW, VSP(2) CD(3) Rationale behind effective sleep advice is given Practitioner 2B. Agreement VSP(2) CD(2) Psycho-education underpins effective sleep advice (Beresford et al. ., 2012g) Practitioner 2B Agreement VSP(2)CD(2) Sleep practitioners address co morbidities before giving effective sleep advice. (Adkins, Molly et al.,2012; Cortesi et al., 2012) Parent 3A. Practitioner 2A Sleep outcome measures are re-run following effective sleep advice . (Cortesi et al., 2012; Malow et al., 2011; Adkins, Molloy et al., 2012) Practitioner 2B. Agreement: SFSO, Amy CD(3)	Financial and human resourcing for disability specific sleep training for sleep practitioners to ensure they can deliver. (Beresford et al., 2012g)Parent 4B and 4C. Practitioner 2B.Agreement VSP(2), VDO CD(2) Agreement: VSS(3), VFW, VSP(2) CD(3) Financial and human resourcing for sleep practitioners to offer the support required by parents to implement sleep advice. (Beresford et al., 2012g; Johnson et al., 1995; O'Connell & Vanaan, 2008) Parent 4B and 4C. Practitioner 2B.Agreement Amy, Emily , Sharon CD(1) , SFSO , Jenny, Amy CD(2), Jenny CD(3) Financial and human resourcing of a paid parent buddy to support parents in following SHE advice. (Beresford et al., 2012b, 2012g) Practitioner 2B. Agreement Amy, Emily CD(1) Jenny, Amy CD(2) Disagreement VDO(CD2)

	Parents are supported to follow sleep advice Beresford et al. (2012g), Johnson et al 1995, O'Connell & Vanaan, 2008. Parent 4B and 4C. Practitioner 2B.Agreement Amy, Emily , Sharon CD(1) , SFSO CD(2), VSS(3), VSP(2) Jenny , Amy, Clarissa CD(3) *"Praise does go a long, long way for the tiniest thing"* Amv CD(3)	Sleep practitioners support parents by praising them for achievements along the way Jenny, Amy CD(2) Amy CD(3) Sleep practitioners are committed to working with parent buddies to support parents to follow sleep advice Beresford et al. (2012b, 2012g). Practitioner 2B. Agreement Amy, Emily CD(1) Jenny, SFSO CD(2)Disagreement VD0 (CD2) Sleep practitioners have a good working relationship with partner organisations and others involved in the child's care to ensure they support parents by following advice consistently SFSO, Jenny CD(2)) Agreement VSS(3), Amy CD(3)	Child has competing complex health needs which prevent parents from following effective sleep advice. Beresford et al. (2012f, 2012g). Parents struggle to follow effective sleep advice due to busy day to day lives Beresford et al. (2012f, 2012g).Parent 4B. Agreement: VSP(2) CD(2) The child has already been offered sleep medication which may reduce their motivation to follow effective sleep advice. Practitioner 2B. Parent 4A. Partner organisations support parents by following advice consistently.SFSO, Jenny CD(2) Agreement VSS(3), Amy CD(3) *"I might have stuck with it then if somebody had have said just keep going cause it will work eventually"* VSS(3) CD(3)	Parent buddy supports parents to follow sleep advice. Beresford et al. (2012b, 2012g). Practitioner 2B. Agreement Amy, Emily CD(1) Jenny, Amy CD(2)Disagreement VD0 (CD2) Parents are invited to bring peer supporters to all appointments to support them. Parent 4C Agreement Sharon CD(1) Multi-disciplinary team and partner organisations are asked to support parents by carrying out advice consistently SFSO, Jenny CD(2) Parents receive the amount and type of support they need Beresford et al. (2012g) Johnson et al , 1995, O'Connell & Vanaan, 2008. Parent 4B, Practitioner 2B Families are supported to follow advice through visual media/communication adaptations Practitioner 2BAgreement SFSO, Amy CD(3) Parents are prepared for the hard work involved in following advice and are supported by this Practitioner 2B Agreement VSS(3), VSP(2), Jenny CD(3)	Parent buddy is employed to support parents with following sleep advice. Beresford et al. (2012b, 2012g). Practitioner 2B. Agreement Amy, Emily CD(1) Jenny, Amy CD(2) Disagreement VDO (CD2) Sleep practitioners invite parents to bring peer supporters to all appointments to support them. Parent 4C Agreement Sharon CD(1) Sleep practitioners support parents by preparing them for the hard work involved in following advice. Practitioner 2B Agreement Jenny, VDO CD(2) VSS(3), Amy,Gloria CD(3) Sleep practitioners support parents by ensuring advice consistently followed across all settings. SFSO, Jenny CD(2) Agreement VSS(3), Amy CD(3) Sleep practitioners give parents amount and type of support needed. Beresford et al. (2012g), Johnson et al , 1995, O'Connell & Vanaan, 2008. Parent 4B, Practitioner 2B Parents are supported with written/visual media Practitioner 2BAgreement SFSO, Amy CD(3)	*"To pay for someone to mediate, wouldn't that money be better spent on training professionals how to build that relationship?"* VDO CD(2)

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6.2.7. Narrative summary of 'Regularity and quality of child's sleep improves.' Figure 23.

This ultimate, direct focus outcome of SHE effort describes how the regularity and quality of the child's sleep can be improved. Improved sleep is firstly defined in a way that is personal to the family whereby they are involved in setting goals which are acceptable to them. It is also defined in terms of sleep outcomes measured by repeated outcome measures such as sleep diaries or validated tools. Success criteria are also described by two categories: parents receive effective sleep advice, and parents are supported to follow sleep advice which will now be described in turn.

The intervention works to deliver effective sleep advice through resourcing the training of sleep practitioners in SHE, which skills them to tailor sleep advice (selected from the SHE tool) according to the assessment findings and needs of the family. This may also include making appropriate referrals to other generic practitioners or support organisations based on assessed needs. Sleep practitioners are competent at delivering psycho-education around sleep and explaining the rationale behind advice to motivate parents to follow it. Sleep practitioners are also skilled at ensuring that physical and psychological co-morbidities are addressed prior to giving SHE advice, and re-running sleep outcome measures at the end of the intervention to evaluate sleep improvements. Programme factors which could affect intervention success are whether sleep practitioners deliver the SHE advice in a non-directive, non-judgemental way, and whether they have sufficient time to re-run outcome measures at the close of the intervention. Numerous contextual factors largely outside the controls of the intervention are identified: firstly, whether parents understand and agree with the SHE advice and all family members follow it

consistently; whether parents have mental health issues, are stressed or too busy to accept and follow SHE advice; whether their home environment is conducive to sleep or the child has competing complex health needs that can prevent them from adhering to sleep advice. Finally, whether the child has already commenced on or been offered sleep medication which may reduce their motivation to follow SHE recommendations. This makes explicit the complexity of SHE and the significant potential for external factors to hinder achievement of this ultimate outcome of intervention effort.

The intervention also works to support parents in following SHE advice through the resourcing of sleep practitioner's time to offer the level of support required by individual families. Sleep practitioners offer parents the amount of home visits and telephone calls they need, communicate sleep advice through written or visual media if required and liaise with other organisations involved in the child's care to ensure they also follow advice consistently. Sleep practitioners support parents by preparing them in advance for the hard work involved in following SHE recommendations and actively invite parents to bring peer supporters to all appointments to make them feel safe. Finally, financial and human resourcing is provided for a paid parent buddy service to support parents with following sleep advice. Programme factors that could affect success are identified as whether sleep practitioners agree with and support a parent buddy initiative, whether they support parents by praising them for achievements throughout their contact and if sleep practitioners have established good working relationships with partner organisations who they ask to commit to SHE advice. At the boundary of the programme is also the contextual factor of whether partner organisations accurately follow sleep advice or jeopardise progress made by parents by failing to do so. This demonstrates the complexity of the support

needed by parents to follow SHE advice which is complemented by the intervention's efforts to also achieve the '*Parents and sleep practitioners develop a safe and supportive relationship*' intermediate outcome. In summary, when the regularity and quality of the child's sleep is improved, quality of life may then improve for the child and family which the next outcome represents.

Figure 24: Quality of life improves for the child and family

Outcomes Chain	Success Criteria	Programme factors	Non-programme factors	Outputs and Throughputs	Activities, Processes, Principles	Resources and Inputs- Financial; Human.
Quality of life improves for child and family *Out of scope outcome.* *"You stop feeling like a zombie"* Sharon CD(2) *"I think families are then ready to help other families."* VFW (2), (CD2)	Child's behaviour improves and parent's stress reduces Agreement: Jenny, Amy, Sharon, VDO,(CD2) Parent's sleep improves Amy, Sharon (CD2) Parents feel empowered to help other parents VFW(2), SFSO, VSS(3) (CD2) Parents feel supported to maintain progress via continued support from sleep teams (Beresford et al.,2012g; O'Connell &Vanaan, 2008) Parents score favourably on quality of life outcome measures or evaluation questionnaires. (Beresford et al.,2012c, 2012d, 2012e; Malow et al., 2013) Practitioner2A.Agreement VSP(2) , SFSO CD(3) Parents feel supported to maintain progress with help from parent support groups Amy, Jenny, VDO CD(2)	**Whether:** Sleep teams are supported to offer continued support to families following intervention. (Beresford et al., 2012g; O'Connell & Vanaan, 2008). Sleep practitioners are committed to running and repeating quality of life outcome measures or evaluation questionnaires (Beresford et al., 2012c, 2012d, 2012e; Malow et al., (2013) Practitioner 2B Agreement : SFSO, VSP (2) CD(3) Sleep practitioners have a good relationship with parent support groups Amy, Jenny, VDO CD(2) .	**Whether:** Parents complete quality of life outcome measures at the start and end of an intervention or return evaluation questionnaires. (Beresford et al.,2012c, 2012d, 2012e; Malow et al.,2013) Practitioner 2B Agreement: VSP (2)SFSO CD(3) Parent support groups are well maintained to support parents in maintaining sleep progress. Amy, Jenny, VDO CD(2) Parents are comfortable with accessing parent support groups for continued support Practitioner 1A.	Parents receive continued support from sleep teams (Beresford et al.,2012g; O'Connell & Vanaan, 2008) Parents have their quality of life evaluated through outcome measures or follow up questionnaires. (Beresford et al., 2012c, 2012d, 2012e; Malow et al.,2013) Practitioner2B.Agreement:VSP (2)SFSO CD(3) Parents receive continued support to maintain progress at parent support groups. Amy, Jenny, VDO CD(2) *I ran the support group on facebook. (..) in the end I just couldn't cope with it, it was just too much"* Jenny CD(2)	Sleep teams offer parents continued support (Beresford et al., 2012g; O'Connell & Vanaan, 2008) Sleep practitioners run and repeat quality of life outcome measures or follow up evaluation questionnaires. (Beresford et al., 2012c, 2012d, 2012e; Malow et al., 2013), Practitioner 2B Agreement: VSP (2)SFSO CD(3) Sleep practitioners collaborate with and support parent support groups. Amy, Jenny, VDO CD(2) *"The self-help groups the parents who run it need to be supported"* Amy CD(2) *"You need lots of back-up in there."* Jenny CD(2)	Financial and human resourcing for sleep teams to offer continued support to families (Beresford et al., 2012g; O'Connell & Vanaan, 2008) Sleep practitioner time to run and repeat quality of life outcome measures or follow up evaluation questionnaires (Beresford et al., 2012c, 2012d, 2012e; Malow et al., 2013) Practitioner2B.Agreement:VSP (2)SFSO CD(3) *"Feedback forms are poorly returned, I think it would be a lot better if we could go out and see the families again and actually fill it out with families"* VSP(2) CD(3) Financial and human resourcing of sleep practitioner time to collaborate with and support parent support groups. Amy, Jenny, VDO CD(2)

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6.2.8. Narrative summary of 'Quality of life improves for child and family.' Figure 24.

This ultimate, out of scope outcome describes how quality of life can be improved as a bi-product of successful SHE implementation. Improved quality of life is firstly defined as noticing improvements in the child's behaviour, parent's own sleep and reduced stress levels. It is also described as a feeling of empowerment to help other families experiencing children's sleep problems. Success criteria are further defined by three categories: parents score favourably on quality of life outcome measures and evaluation questionnaires, parents receive on-going support from sleep teams, and parents receive ongoing support from parent support groups; each will be presented in turn.

SHE works to measure improvements in quality of life through running quality of life outcome measures at the start and end of the intervention. Alternatively, qualitative evaluation questionnaires may be administered as a follow up to the intervention to capture how parents' feel their quality of life has been improved. The completion and chasing up of this documentation requires resourcing of practitioner's time and additional commitment from sleep practitioners. Whether parents have the time and motivation to complete the documentation is also acknowledged as an external factor which could impact on the success of this outcome.

The intervention also works to help parents feel supported to maintain sleep progress made by offering follow up support for families. It is important that parents feel reassured by the offer of continued sleep practitioner support if needed once the intervention has been completed, which helps them to sustain sleep improvements in

their child. It is acknowledged that adequate financial and human resourcing of sleep teams is required to meet this commitment to families, to ensure quality of life improvements are sustained.

Finally, the intervention works to help parents feel supported in sustaining sleep progress by ensuring parent support groups are available and that these are supported by sleep practitioners. The ongoing support and encouragement that can be offered by parents in a similar situation can be a powerful tool to help parents continue with SHE advice and avoid returning to previously unhelpful coping strategies. Attending parent support groups after the intervention has finished can therefore have a positive impact on maintaining quality of life improvements. However, it is noted that sleep practitioners need to invest time collaborating with parents who run these groups to ensure they are supported to continue. Furthermore, some parents may not be comfortable with accessing these groups which represents a non-programme contextual factor beyond the intervention's control, which may affect achievement of this outcome.

6.3 . Conceptual representation of the programme theory outcomes chain logic model

An outcomes chain logic model (Figure 25) is presented which visually represents the SHE programme theory and emerging conceptualisation of this study's original contribution to knowledge. Each intervention outcome is displayed in a blue box in the centre of the diagram. The lightest blue boxes represent the immediate outcomes, mid-blue boxes represent intermediate outcomes and the final two darkest blue boxes show the ultimate outcomes of SHE. This model shows the sequence of results leading to the ultimate outcomes of improved sleep and quality of life and is circular

to emphasise the holistic approach and cyclical rather than linear process of the intervention.

Typically, parents and practitioners begin at the immediate, pre-implementation outcome of 'Parents and practitioners have a shared understanding of what a sleep problem is' and move in a clockwise direction achieving intermediate and ultimate outcomes. Each black arrow represents the intended progression of one outcome to the next. The mechanisms by which each outcome is achieved are explained in the theory of action, programme theory matrices (figures 18-24) and are represented in the first encompassing purple ring. However, as depicted in the circular representation of the model, parents may backtrack to previously attained outcomes or exit the intervention at various time-points and after a period of time re-join at the stage of 'Parents and practitioners have a shared understanding of what a sleep problem is'. To represent this, red arrows show how outcomes are sometimes not achieved as originally intended, due to contextual factors (programme and non-programme factors) and the intervention fails or takes longer to work.

Emerging conceptualisations with mid-range theories of change which demonstrate how the SHE intervention presents plausible solutions to sleep problems are depicted in the second encompassing green ring. This shows how the broad social evidence based theories of Normalisation, Self-efficacy, Empowerment and the Trans-theoretical model of change underpin the intervention across all outcomes. Novel analytical themes which further explain the complexities of SHE (legitimation, customistation, knowledge exchange, health expectation and rationing and gaming) are denoted in the outer encapsulating yellow ring, which again shows how they have relevance across the broad spectrum of intervention outcomes. These novel

themes represent this study's original contribution to knowledge. A detailed

discussion of these mid-range and novel analytical themes is presented in the

following chapter.

Figure 25: SHE Outcomes chain logic model.
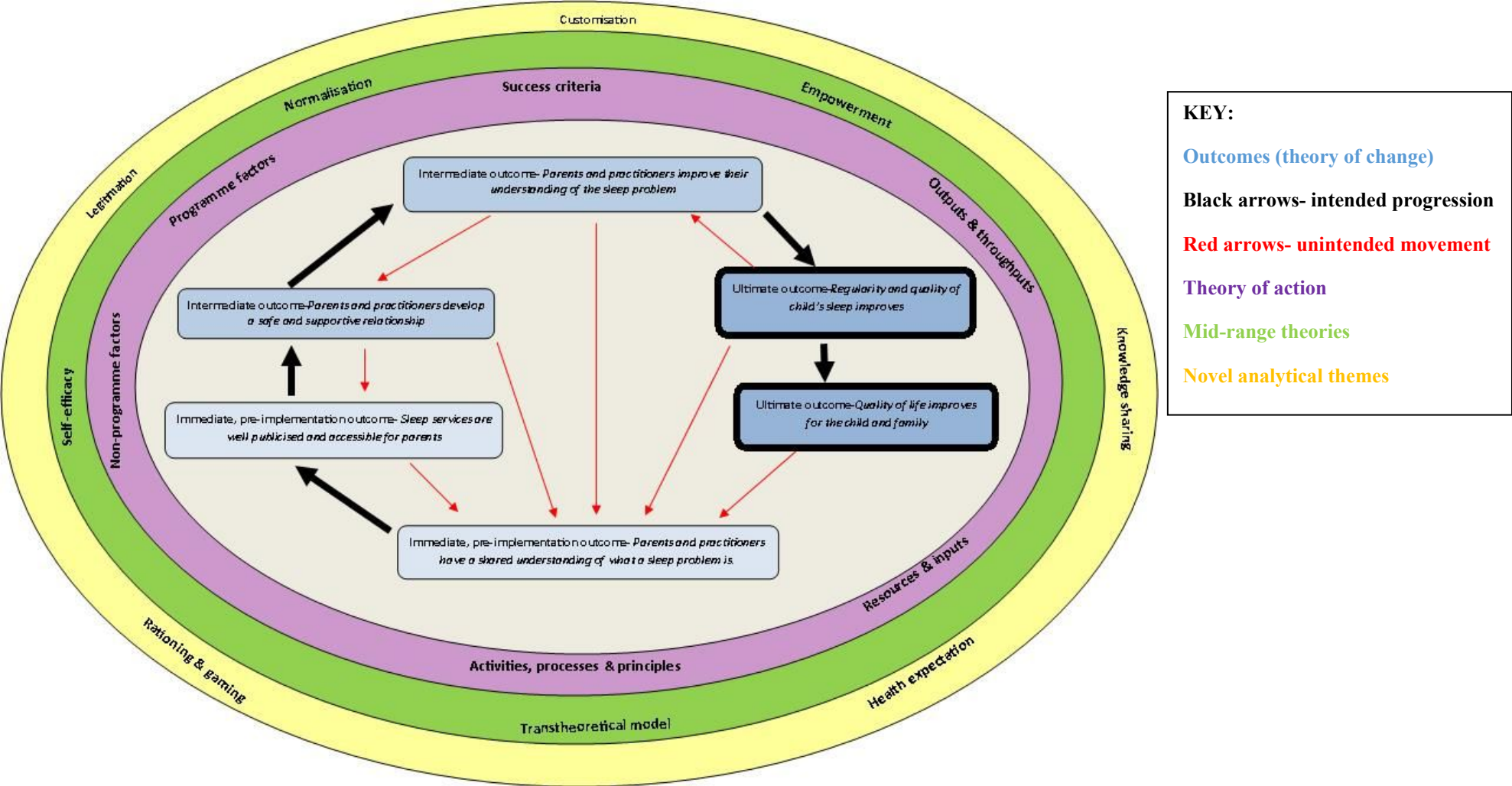
CHAPTER 7. DISCUSSION

7.1. Introduction

This chapter begins with discussion of how the systematically developed SHE programme theory relates to mid-range theories of change. This is followed by a discussion of how the programme theory links to the underpinning novel analytical themes. Novel themes represent this study's original contribution to knowledge: an explicit understanding of the complexity embedded in an SHE intervention. Most importantly, this study shows how the legitimation of sleep problems is a foundation on which SHE implementation should be based. Findings also demonstrate a greater understanding of the nature of customisation, health expectation, knowledge exchange and impact of rationing and gaming on implementation success. These themes are discussed in relation to existing evidence and broader theoretical perspectives. Implications for practice, policy and research are also outlined. Presented next is a section on reflexivity to show how the researcher's learning developed throughout the study and PhD process.This is followed by study limitations and a critical appraisal of thesis findings. This chapter then concludes with some final remarks on this study's overall findings and orginal contribution to knowledge.

7.2. Abstraction to Wider Evidence Based Theories of Change

7.2.1. Self-efficacy theory.

This study has found that the attitudes of other family members, generic practitioners and peers are significant in affecting parents' ability to both recognise sleep

problems and effectively engage with a SHE intervention. This links with the principles of the sociological theory of self-efficacy (Bandura, 1977; 1997). This refers to a person's subjective belief that they are capable of engaging in a particular behaviour and holds that a strong sense of self-efficacy frequently produces success, whereas those who doubt their capabilities often give up sooner when faced with challenges. The four main strategies that build self-efficacy are mastery experiences (successes achieved through perseverance), vicarious experiences (witnessing the success of social models), social persuasion (receiving active encouragement) and modifying individual's perceptions of their physical and emotional states.

Self-efficacy is often extrapolated to parenting self-efficacy which considers not what parents know or do, but instead what they believe they can do to influence children's development (Giallo, Wood, Jellet & Porter, 2013; Sevigny & Loutzenhiser, 2009). In a review of research on parenting self-efficacy (PSE) Jones and Prinz (2005) showed a robust evidence base linking PSE to parental competence and support for the impact of PSE on child development. A lack of research was identified however, exploring the predictors of PSE and related variables such as the influence of family generations and employment factors; this reveals limitations in Bandura's theory and suggests potentials for further development. This study's findings would support this, as parent data indicates how the attitude of other family members (and also generic practitioners and peers) is significant in affecting parent's ability to recognise sleep problems. Furthermore, findings of a study of mothers of children with autism (Chong & Kua, 2016) supported the premise that mastery experiences were the most significant factor in building PSE, followed by physiological and affective states of parents. However, vicarious experiences and

social persuasion were not found to be critical in shaping PSE indicating a potential limitation in the application of the theory.

Conversely, in this study, the strategies of social persuasion and vicarious experiences were found to be the most impactful on parents' self-efficacy to engage with SHE. Findings showed how social persuasion was influential during the sleep screening process and implementation of SHE advice, and the value of vicarious experiences was clearly demonstrated in stakeholder debates around paid parent experts and parent support groups. Evidence was also found in support of the impact of mastery experiences (motivating parents using outcome measures) and physical and affective states of parents; however it is acknowledged that these strategies could have been explored in greater depth, which raises a potential area for improvement in the conceptualisation of SHE programme theory. Notwithstanding, espousing links to self-efficacy theory further qualifies intervention complexity and shows how there can be numerous contextual factors affecting the ultimate success of a SHE intervention.

7.2.2. Empowerment theory.

Study findings show how parents want to be empowered to independently identify their children's sleep problems and access sleep services. Furthermore, findings demonstrate the need to place greater value on the knowledge and experience of parents in relation to sleep problems. These findings chime with the tenants of empowerment theory which describes how individuals take control over their lives and emphasises self-determination and participation (Perkins & Zimmerman, 1995). It assumes that problems are best addressed by those experiencing them, and individuals have valuable insights into their own needs and should be empowered to

develop their strengths to become independent decision makers. The model has been widely applied in the care of individuals with disabilities (Fleming-Castaldy, 2013; van Nijnatten & Heestermans, 2012) and empowerment practices have been strongly associated with positive outcomes for families of children with disabilities (Dempsey & Dunst, 2004; Dempsey & Keen, 2008; Fazil, Wallace, Singh, Ali & Bywaters, 2004).

Explanatory links were made with the construction of empowering parents to independently screen for children's sleep problems which was put forward largely by parents. Parents expressed a desire to take self-determining action to identify problems which demonstrated belief in their power to affect sleep outcomes for their child. The construction of sleep teams taking a more active role in promoting their services also chimed with the tenants of empowerment theory. This was suggested in response to findings that sleep teams were often poorly advertised, creating an unnecessary barrier for parents who were motivated to seek support. Lastly, links were made with calls to value parent's knowledge and experience in the assessment process, the involvement of paid parent experts and parent support groups in supporting parents with SHE implementation.

However, the limitations of applying this theory in terms of inclusivity were addressed in programme theory development. As Funnell and Rogers (2011) point out, it can be unhelpful to assume that all individuals have a desire to play an active role in addressing problems. For example, the contextual factors of some parents being too busy to participate, lacking confidence to approach sleep teams and parental literacy difficulties all demonstrate how empowerment theory cannot be applied to all parents. Therefore, safety nets were incorporated, such as practitioners

being mindful of parent's literacy problems and offering the amount of support needed by families on an individual basis.

7.2.3. Normalisation theory.

Study findings have shown the impact of parents 'normalising' their child's sleep problems and desire not to feel different from other parents of typically developing children. Parents' expressions of wanting all children to be screened for sleep problems and accessing generic rather than specialist sleep services have also indicated a desire for normalcy. It is posited that these findings relate to the theory of normalisation (Barnes, 1996; Gilbert, 2004; Wolfensberger & Tullman, 1982), which regards difference as deviant, which causes stigmatisation and social exclusion triggering increased stigma. Social stigma is defined in the works of Goffman (1963) as the extreme disapproval of a person or group on the grounds of difference from broader society. According to the principles of normalisation, when people are not integrated and remain 'different' they may find it difficult to feel valued by others. Society may view their 'differentness' negatively and perpetuate these devaluing attitudes.

Explanatory links are made with various contextual factors in the programme theory. Firstly, whether parents 'normalise' their child's sleep and deny that a problem exists, could be explained by a desire towards not feeling any different from other parents. The emotion of fear of admitting a child's sleep problem and being singled out as different from other parents was also explained by the principles of normalisation. Similar findings were shown in Kimberly & Keiley (2003) who emphasised how parents commonly used denial as a defence against the stigma attached to disability. Fear of accessing pre-school groups where parents may have

their awareness of sleep problems raised was linked to anxieties around their child appearing different from others. Lastly, a fear of accessing specialist sleep teams and stepping into disabled services was also linked with a notion that difference was undesirable and something to be anxious about. Similar findings were shown in studies by Vogel and Wade (2009) and Dempster, Wildman and Keating (2013), which acknowledged the significant role of stigma on help-seeking behaviours. These studies showed the need to consider stigma in service design, to ensure those who needed advice were supported to access it in a timely manner.

Analytical links were also made with the following processes: The activity of screening all children for sleep problems was suggested to help parents feel more at ease with discussing their children's sleep. This reflected the assumption that reducing difference and stigma promotes inclusion. Similarly, the process of offering parents access to a generic sleep service was surmised to increase the likelihood of parents making contact with practitioners by 'normalising' the act of asking for sleep help. Through providing services that are perceived as 'normal' for all children regardless of disability, it is posited that the intervention should work to enhance parents' behaviour in positively engaging with sleep services. These assumptions are strengthened by grounding them in normalisation theory.

Criticisms of normalisation focus on its denial of difference and value of conformity (Culham & Nind, 2003; Gilbert, 2004, Bronston, 1974). Although opponents accept the basic proposition that oppression can stifle change, it is argued that in normalising services, the individual needs of the individual or experience of disability can be overlooked. This criticism is reflected in the challenge presented by three co-designers to the provision of a generic sleep service, who argued that

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generic practitioners may lack understanding of the specific needs of children with DD which would be unhelpful for parents. Therefore, whilst abstraction to the ideological position of normalisation theory is helpful in some cases, limitations do exist in its application.

7.2.4. Stages of change theory (transtheoretical model).

Study findings have indicated that parents may go through a series of pre-implementation, implementation and maintenance stages of change in their quest to improve their child's sleep problems. Although evidence can be found of distinct progression points, it was also apparent that advancement towards the ultimate outcomes is not always linear or uniform in time. However, broad links can be made with the stages of change theory or transtheoretical model (TTM) (Prochaska & DiClemente, 1983; Prochaska & Velicer, 1997). This has been widely used in interventions to address health issues such as obesity (Mastellos, Gunn, Felix, Car & Majeed, 2014) and smoking cessation (Aveyard et al., 1999). It posits that behaviour change occurs through stages and is structured by three core sub constructs (Funnell & Rogers, 2011; Taylor et al., 2006). First, the temporal dimension holds that change happens over a succession of six stages, which individuals enter at different time points and move up or down thus following a linear or spiral course. The first three stages represent an individual's behavioural intention*: 'pre-contemplation'* (the person is not considering changing their behaviour in the foreseeable future); '*contemplation'*, (the person is thinking about changing their behaviour in the next six months) and '*preparation'* (change is planned in the coming month). These are followed by the '*action'* stage (the person has made a behaviour change within the last six months), '*maintenance'* stage (behaviours have been sustained for at least six

months) and '*termination*' (behaviours are fully established for five or more years).

Second, the TTM model holds that individuals experience intermediate outcomes as a series of change processes which include decisional balance and self -efficacy. Third, 10 processes of change are suggested categorised into behavioural and experiential activities that have varying levels of influence at different stage transitions.

Broad links can be made between the temporal dimension relating to the behavioural intention of individuals, and the immediate outcomes of SHE (Figure 17, p315). These outcomes describe important pre-implementation elements and acknowledge that parents and practitioners go through a process of deciding whether there is a sleep problem and when/how to access help, before SHE is fully implemented. Intermediate outcomes and the ultimate outcome of '*Regularity and quality of child's sleep improves*' link to the action stage whereby changes are made to sleep hygiene practices. Lastly, the ultimate outcome of '*Quality of life improves for the family*' abstracts to the maintenance stage where sleep improvements are sustained and the family experiences positive knock-on effects. Although the temporal dimension was useful to help describe the different stages at which parents enter and move through a SHE programme, problems did arise with its applicability. It was difficult to neatly match each outcome with each change stage and specify the time points suggested by the TTM, because in reality parents and practitioners varied in their time spent achieving each outcome. This reflects a popular criticism of the TTM which argues that the categorisation of change into a series of distinct stages rather than a continuous process can be unrealistic (Armitage, 2009; Nigg, Geller, Horwath, Wertin & Dishman, 2011). Furthermore, individual stages are poorly

defined, difficult to differentiate and the time frames between them could be considered arbitrary (Funnell & Rogers, 2011; Sutton, 2001).

SHE programme theory is also linked to the second sub-construct of TTM which posits that the detail in intermediate outcomes should be identified, ensuring these are measured rather than just final outcomes. This is represented by the range of success criteria specified in each outcome. This also helps to measure parents' pattern of development if relapse occurs at any time point. For example, the contextual factor of whether parents have other stressful life events happening and are ready to accept advice links with the TTM's construct of individuals having confidence and self-efficacy beliefs to effect change. (This also shows how TTM links to self-efficacy theory, and how mid-range theories commonly share some elements). Also, the construction of whether parents understand and agree with advice being given relates to parents weighing up the 'pros and cons' of implementing SHE. Therefore, detailing all of the outcomes helps to measure a parent's journey toward improving their child's sleep rather than just their status at a given time or success at achieving the final result.

Finally, explanatory links are made to the processes of behaviour change posited by the TTM. During the behavioural intention stages, the experiential activity of running a media campaign can be classified as TTM's 'consciousness raising' about the problem. Also, the activity of parents screening for sleep problems using their PCHR, links to the concept of 'social liberation' which focuses on the empowerment of individuals. During the action stage, the activities of practitioners demonstrating good interpersonal skills and the parent buddy initiative link with 'helping relationships' processes. Finally, during the maintenance stage, collaboration with

parent support groups links with 'stimulus control' processes also stipulated by the model. However, the potential to include elements of the six other change processes specified by TTM: dramatic relief, environmental re-evaluation, self-re-evaluation, counter conditioning, reinforcement management and self-liberation is noted which may require further exploration to enhance intervention effectiveness.

Whilst it was helpful to make explanatory, theoretical links with the model, account was taken of a final criticism which argues that inadequate detail is given to the effects of external social, environmental and economic factors (Funnell & Rogers, 2011; Taylor et al., 2006). Care was therefore taken to consider non-programme factors, such as whether parents mask problems with coping strategies such as co-sleeping, and how this may mean they take longer to recognise the sleep problem, causing it to become entrenched and the parent to progress more slowly through the stages of change. This shows how application of TTM is not entirely sufficient in explaining all the nuances of complex interventions such as SHE.

7.2.5. Abstraction to mid-range theories, concluding remarks.

The process of making analytical links with mid-range theories has encouraged critical reflection of programme theory, strengthened rationale for constructions presented and raised questions about potential effectiveness. Links with wider, evidence based theories which transcend local theories have been helpful in demonstrating how the intervention presents plausible solutions to sleep problems in children with DD, however, a discussion of their strengths and weaknesses has highlighted potential limitations in their application.Therefore, whilst abstracting to theories of change was useful in demonstrating how the programme theory was

grounded in wider evidence, they were viewed as heuristic devices rather than strict formulas to adhere to.

7.3. Abstraction to Novel Analytical Themes- The Original Contribution to Knowledge.

7.3.1. Legitimation.

This study posits that there is a need to legitimate sleep problems in children with DD, to ensure they are appropriately prioritised, funded and resourced. To legitmate means to make "reasonable, sensible or valid" Collins English dictionary (2016). Constructions throughout the programme theory demonstrated stakeholders' perceptions of how children's sleep problems were under-valued and viewed as a private parenting issue, by parents, practitioners, policy makers and wider society. Increasing the legitimacy of sleep problems is therefore presented as an over-arching philosophy and foundation upon which SHE implementation needs to be based.

Findings indicate that when parents do not view sleep problems as a justifiable concern, this impacts on their ability to acknowledge them and seek professional help. Contextual factors of parents stoically coping with sleep problems and struggling to give precedence to sleep against competing health issues, demonstrate the influence of legitimacy on help seeking behaviours. Concordance is found with Robinson and Richdale's (2004) exploratory study which concluded that "many parents are poor at recognizing that a sleep problem exists. Treatment is frequently not sought even if the problem is recognized" (p.149) and attributed this to parents' knowledge of parenting and beliefs around sleep. Wiggs and Stores (1996b) similarly reported reluctance from parents to seek help which they linked to stoicism

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and a belief they could carry on without professional help. Wider literature therefore supports the premise that parents need to be encouraged to give more value and currency to children's sleep problems to ensure they receive the help they deserve. This study makes a number of recommendations that could help to raise the legitimacy of sleep problems in the minds of parents and firstly points to the role of practitioners.

It is posited that the extent to which practitioners legitimise sleep problems influences their early interactions with parents. Findings indicate that practitioners are sometimes dismissive of parent's initial sleep concerns and this can delay sleep problem identification and receipt of appropriate support. In a qualitative investigation into the experiences of parents of children with Rett syndrome (a neurodevelopmental disorder) McDougall, Kerr and Espie (2005) also reported negative experiences of generic practitioners failing to take sleep problems seriously; however, the authors did not examine how this may impact on parents' decisions to seek help. The Tired all the Time report (Family Fund, 2013) also evidenced how some practitioners were dismissive of parents' concerns, and broadly acknowledged this had a damaging effect on exhausted parents. Thesis findings add to this body of evidence and make explicit the link between the level of legitimacy afforded to sleep problems by generic practitioners and parents' ability and decision to seek help. It is recommended that future direction is required at the professional level to develop knowledge and understanding of sleep problems, so that their legitimacy can be cemented in the minds of practitioners and subsequently parents. This includes educating generic and sleep practitioners to consistently adopt positive attitudes towards sleep problem recognition and openly discuss sleep problems with parents in

a non-judgemental way at the outset, to ensure they are recognised and addressed in a timely manner.

Furthermore, this study adds to existing evidence by positing that the extent to which society legitimises sleep problems, can influence the pace at which parents recognise and seek support for them. Parents reported how the contextual factors of feeling judged by older generations, peers and wider society could prevent them from requesting help. They described a public perception that children's sleep problems were a private parenting issue not worthy of professional input. This signifies a level of social stigma (Goffman, 1963) or shame that parents feel when they decide to involve generic practitioners. Furthermore, the unhelpful impact stigma has on help seeking behaviours is well documented in the works of Vogel and Wade (2009) and Dempster, Wildman and Keating (2013). Therefore, there is a need to 'normalise' the act of seeking help, which aligns with the principles of normalisation (Barnes, 1996; Gilbert, 2004; Wolfensberger & Tullman, 1982). It is suggested this can be addressed by legitimising sleep problems in the minds of parents, practitioners and wider society through increasing the public conversation about sleep and running media campaigns which promote understanding. Parallels can be drawn with the recent Time to Change campaign (MIND & Rethink Mental Health, 2007) which was reportedly successful in improving public attitudes and reducing social stigma around mental health. In addition, the recommendation of this study to introduce standardised sleep screening also reflects efforts to make it usual practice to enquire about sleep, which could help to reduce stigma around professional involvement.

Parents and sleep practitioners showed how they also perceived funders currently poorly prioritised sleep problems in children with DD. Stakeholders reflected this in

their requests for better resourcing of sleep teams to meet service need, provide continuity of parental support, produce comprehensive assessments and champion parent support groups. This concurs with the Tired all the Time report (Family Fund, 2013) which reports that "sleep difficulties need to be given a higher priority by health, education and social care professionals and listed in the local offer (England)" (pp.13).

It is important to reflect upon why sleep problems in this group of children are poorly prioritised by organisations and policy makers. Beresford et al. (2012h) suggest that poor service provision can be linked to the fact children with DD "are a small minority in a much larger population. Thus, there is a risk of them being overlooked by mainstream parenting activities at a local level" (pp.271). Broad links can also be made with reports which have demonstrated how people with learning disabilities are a marginalised group who receive inequitable and inadequate healthcare (Atkinson et al., 2013; Department of Health, 2015; Disability Rights Commission, 2006; Emerson, Baines, Allerton & Welch, 2012; Mencap, 2007). Therefore, the minority status of children with DD may explain why their sleep problems may go unnoticed by policy makers. This raises the continued need to highlight the experiences of families through the dissemination of findings and further research which promotes wider understanding of sleep problems and the need for them to be addressed.

It is also posited that the 'invisibility' of sleep problems to others outside the family home, may result in organisations placing less priority on the need for professional input. In contrast to other health issues (such as challenging behaviour or continence issues), sleep problems largely occur at bedtime or during the night, so their

immediate impact is limited to the family. Reports that show how generic practitioners still need to treat parents of people with DD with greater respect and value their caring role (Department of Health, 2009; British Institute of Learning Disabilities (BILD), 2013) indicate how this explanation of "out of sight, out of mind" may be correct. In addition, a wealth of evidence that describes the negative impacts of sleep deprivation on family health (Contact a Family, 2009; Family Fund, 2013; Handsel Trust, 2009) and links with daytime challenging behaviour (Rzepecka, McKenzie, McClure & Murphy, 2011) further shows the need to give greater legitimacy to children's sleep problems, and to ensure adequate service provision is available.

In conclusion, this study presents a strong evidence base in support of the need to consider the legitimation of children's sleep problems as an underpinning philosophy to guide SHE implementation. It is posited that raising the profile, value and validity of sleep problems in the minds of parents, generic practitioners, policy makers and society can positively impact on SHE implementation, across all intervention outcomes. This has implications for practice in terms of generic and sleep practitioners needing to access sleep training to enhance their understanding and appreciation of sleep problems. For policy this means a commitment to sleep screening initiatives, media campaigns and re-prioritisation of sleep provision to ensure families' needs are met. Implications also include a need for continued research into the experiences of families to ensure children's sleep problems are given enhanced legitimacy and are current in the minds of policy makers.

7.3.2. Customisation.

This study demonstrates a greater understanding of the nature of customisation within SHE implementation. Within the broader context of healthcare, customisation relates to a shift away from standardisation to being responsive to the individual needs of patients/service users (deBlok, Meijboom, Luijkx & Schols, 2012; Minvielle, Waelli, Sicotte & Kimberly, 2014; Thompson & Nussbaum, 2000). As Minvielle et al. point out "customisation is not totally absent in healthcare, but its link with the intervention it qualifies is often not explicit" (p.217). This study makes explicit the need to customise SHE advice according to assessed need, rather than giving poorly informed or standardised advice. It is posited that if parents perceive advice is responsive to their child and families' needs, they will feel motivated to follow it which positively impacts on implementation success.

Broad parent and sleep practitioner evidence indicates how customisation is central to parents and the following recommendations explicitly link it to SHE: the need for sleep practitioners to perform multiple assessment sessions, at the child's home, at bedtime and to involve other members of the MDT and partner organisations in the sleep assessment. These activities demonstrate how parents need to perceive that sleep practitioners have invested sufficient time in understanding their child's individual needs before any advice is given. The requirement to tailor SHE advice according to assessment findings also directly reflects the notion of customisation. Similarly, the elements of factoring in parents' busy daily lives on their capacity to follow advice and introducing written or visual supportive documentation where necessary, are both reflective of an intervention that should be different for each family and responsive to individual implementation needs. This implies a need for

sleep practitioners who are well trained in sleep and SHE and highlights a training need at a practice level.

The comparison of broad based versus customised SHE advice was largely unexplored in the scoping review, however, the findings of Adkins, Molloy et al. (2012) showed that standardisation within SHE was ineffective. More broadly in respect of behavioural sleep interventions, Wiggs and Stores (1996b) found that "blanket" advice often failed and was inappropriate for children with DD. However, other evidence suggests that standardisation can be effective (Beresford et al., 2012e; Bramble, 1997; Christodulu & Durand, 2004; Montgomery et al., 2004) which contradicts this study's overall findings. It is posited that the participatory methodology and co-design methods employed in this research, delved deeper into the perceptions of stakeholders and revealed their dissatisfaction with standardisation. This resulted in a more nuanced understanding of customisation and revealed standardisation to be largely ineffective and patronising for parents.

Customisation also fits with modern learning disabilities philosophies which focus on person centred care (Department of Health, 2001, 2009; Sanderson & Lewis, 2012). 'Person centred care' means tailoring services to the needs and preferences of the person. It shifts service provision for people with learning disabilities and their families, away from a service led to a person centred ethos (Mansell & Beadle-Brown, 2004). Although the literature is mainly focused on adults with learning disabilities, evidence which links person centred care to positive health outcomes for families of children with DD is growing (Black, McConkey, Roberts & Ferguson ,2010; Davis & Gavidia-Payne, 2009). Such connections to person centred care

further evidence why policy makers need to consider customisation within service design.

To ensure a balanced discussion, it is important to reflect upon why SHE is sometimes standardised. Firstly, the strong evidence base which supports some SHE advice such as bedtime routines and sleep timing (Jan et al., 2008), could imply this needs to apply to ALL children which may support standardisation. Secondly, as Minvielle, Waelli, Sicotte and Kimberly (2004) argue, it may represent the most cost effective and least labour intensive approach in healthcare delivery. Furthermore, customisation can be associated with a significant investment of resources and often "budgetary control mechanisms are used to undermine the individualised tailored nature of planning" (p.1) (Mansell & Beadle-Brown, 2004). Although the immediate cost and resource implications of customisation may reflect reality, the findings of this study show parents place more value on advice that is tailored than that which is broad-based and possibly inappropriate. This links with the principles of prudent healthcare (Bradley & Willson, 2014) which holds that patients should receive the minimum appropriate intervention to achieve health outcomes rather than advice which is superfluous and poorly considered. Therefore, study findings recommend that policy is supportive of a customised approach within SHE.

In summary, this study presents a greater understanding of the nature of customisation with SHE implementation. For practice this means ensuring sleep practitioners are well trained in sleep and SHE to ensure they have the skills to customise. It has also been shown that customisation is supported by the principles of prudent healthcare and person centred care, and policy makers need to consider its potential benefits when planning services.

7.3.3. Knowledge sharing.

The benefits of two-way knowledge exchange between care providers and the recipients of care, in increasing patient involvement and enhancing positive outcomes are well documented (Coulter, 2005; Davis & Meltzer, 2007; Tobiano, Bucknall, Marshall, Guinane & Chaboyer, 2016). The concept is based on a two-way dialogue between practitioners and service users, rather than traditional models which focus on a monologue of information transfer that privileges professional over lay knowledge (Lee & Garvin, 2003). This study makes explicit the nature of knowledge sharing within SHE and makes recommendations for how this can be executed in practice.

Firstly, it is helpful to note that much of the literature focusing on patient engagement and knowledge exchange links with the principles of empowerment theory, which encompasses self-control, self-determination and participation (Perkins & Zimmerman, 1995). Pelletier and Stichler (2014) describe how a patient-centred model of care empowers patients through education to become active rather than passive recipients of care. Coulter and Ellins (2007) associate patient engagement initiatives with empowerment and highlight the need to consider health literacy so individuals can understand information (which mirrors SHE contextual factors focusing on parents' communication needs). More specific evidence around supporting families of children with DD, such as the Early Support family partnership model, also chimes with empowerment theory. The model cautions if parents' knowledge and ability to participate in children's care is not valued this "reduces their control of the process, decreases the understanding of problems and possible effectiveness of intervention; it does not enable them to understand the processes of helping and may disempower them" (p.5) (Davis & Meltzer, 2007). It is

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therefore posited that linking knowledge sharing with empowerment theory helps to further validate its incorporation into a SHE intervention.

The execution of knowledge exchange within SHE is made explicit in the following ways. Firstly, parent and practitioner data evidences how parents should be encouraged to share their expertise, through engagement in sleep assessment interpretation and the co-creation of customised intervention plans. Whilst broader patient engagement literature evidences patient preference and desire for such active involvement in care planning (Little et al., 2001; Tobiano et al., 2016), others caution how not everyone wants decision making responsibility at all times and social, cultural and emotional factors could affect this (Davis & Meltzer, 2007; McDonnell, Lohan, Hyde & Porter, 2009). This is also a common criticism of empowerment theory (Funnell & Rogers, 2011) and SHE contextual factors which account for issues such as parents' busy day to day lives and emotional capacity, act as safety nets to ensure opportunities for parental involvement are maximized. Supporting parents to actively contribute to decision making in SHE, guided by their individual capacity and desire is therefore a key message for practitioners.

Next, the knowledge sharing recommendations of employing a parent buddy and championing parent support groups to facilitate SHE implementation were also largely well supported by parent and practitioner evidence. However, conflict in the data did exist around practitioner acceptability of paid lay experts, indicating how the role could be perceived as threatening to sleep practitioners. Broader literature examining the role of lay health workers highlights similar implementation barriers such as difficulties managing boundaries, uncertainty and confusion around exact roles and their relationships with generic practitioners (Glenton et al., 2013;

Kennedy, Milton & Bundred, 2008). Whilst espousing potential improved outcomes, these studies highlight the need to adequately conceptualise the parent expert role. This seems prudent in the context of SHE and implies a need for future research into the parent buddy role and to further develop the emerging discipline of involving lay helping within care.

Knowledge exchange processes that focused on the responsibilities of sleep practitioners to freely share their expertise with parents are also presented. Parent and practitioner data indicates that sleep practitioners need to have disability-specific sleep knowledge and take steps to prepare parents in advance for implementation challenges. However, only practitioner evidence supported recommendations of providing parents with rationale underpinning advice and need for psycho-education; implying limited parental experience of such initiatives. Intervention studies included in the review incorporated some element of parental education, validating its incorporation into SHE (Christodulu & Durand, 2004; Cortesi et al., 2012). Furthermore, a recent evaluation of parents' experiences of psycho-educational sleep management interventions involving SHE, found that increasing parental knowledge and understanding about sleep was instrumental improving children's sleep (Beresford, Stuttard, Clarke & Maddison, 2016). Authors also noted the need to account for contextual factors such as consistency of approach and adequacy of housing conditions, which have been similarly included in programme theory development. Consequently, based on the strength of the evidence, incorporating activities which encourage the liberal sharing of professional knowledge represents a key recommendation for practice.

In summary, study findings have demonstrated a greater understanding of knowledge exchange within SHE and how this links with the theoretical perspective of empowerment. For practice this means a commitment to actively involving parents in assessment and decision making processes, where their capacities and preferences allow, championing parent support groups and incorporating SHE rationale and psycho-education into interventions. For research a need to conceptualise the role of a paid parent expert within SHE implementation is also identified.

7.3.4. Health expectation.

Study findings demonstrate the added complexity of health expectation which is embedded in SHE interventions. In addition to receiving customised, co-created advice, parents also need to feel hopeful that their child's sleep can improve. It is posited that if parents and practitioners maintain a positive health expectation and keep their confidence high in the ability of children with DD to sleep better, this will positively impact on implementation success. Janzen et al. (2005) define a health expectation as "a prediction about the consequences of certain health-related phenomena (behaviours and conditions, both internal and external), on the psychological condition of the body." (p.45). Janzen et al. suggest that expectations can relate to interventions and health status and are highly important in relation to the experience of health and health care.

Primarily, parent and practitioner data shows how pre-implementation, low health expectations impact on sleep problem recognition. It was reported that practitioners sometimes began with a negative outlook and limited conviction in sleep improvement, and this discouraged parents at start of their SHE journey by invoking feelings of hopelessness. Evidence is also presented of parental decision making to

carry on coping regardless, beliefs that sleep problems are inherent in DDs, or professional sleep help is ineffective or unavailable. This is consistent with existing evidence which reflects how parents and practitioners sometimes perceive that sleep problems in children with DD are inevitable and untreatable (Family Fund, 2013; McDougall, Kerr & Espie, 2005; Robinson & Richdale, 2004; Wiggs & Stores, 1996b). Parallels can also de drawn with the 'diagnostic overshadowing' bias whereby a person's underlying health needs are overlooked and attributed to their learning disability (Mason, 2007; Mason & Scior, 2004). In the context of this study, it would seem both parents and practitioners sometimes demonstrate this in their belief that sleep problems are inherent in DD and resulting low health expectation.

Broader similarities can also be found with reports of health professionals adopting complacent or fatalistic attitudes towards people with learning disabilities (Department of Health, 2015; Disability Rights Commission, 2006; Mencap, 2007), and calls for a cultural shift in health expectation, to ensure individuals do not experience inequitable care when compared with non-disabled peers. These reports also suggest that low health expectation is indicative of health professionals being poorly informed of the health needs of this group. This implies a training need for practitioners in sleep, SHE and developing encouraging relationships with parents. For policy, this makes explicit the need for media campaigns to raise parental expectations around effective sleep interventions which improve children's sleep. Links with the theoretical perspective of legitimation can also be made, in ensuring policy makers are committed to prioritising the sleep needs of this group of children and resourcing adequate service provision.

It is important however, to also consider the reality that parent's high expectations of sleep help may not always be met, due to poor service availability. Although findings show that sleep support is available from third sector organisation and social services sleep teams in addition to health provision, parents did report experiences of being unable to source support. In these situations, parent support groups and social media forums are accessed for advice, or alternative strategies such as co-sleeping practices or weighted blankets are adopted. Although some of these strategies help parents cope in the short-term, they are not always consistent with SHE and can be inappropriate. However, in the absence of sufficient local service availability, the supportive role of parent support groups is recommended as an implication for practice.

Parent and practitioner data also indicates how it is important to maintain parents' hope that sleep can improve during SHE implementation. It is posited that practitioners need to adopt an enduring positive and reassuring attitude, which encourages parent's conviction in their abilities to effect change. This concurs with the theoretical perspective of self-efficacy (Bandura, 1977, 1997) and social persuasion whereby individuals who receive encouragement are more likely to develop self-efficacy, which validates incorporation of the health expectation theme within SHE. This continues to imply a training need on a professional level to ensure positive working relationships are developed with parents that instil optimism.

In summary, this study makes explicit the nature of health expectation within SHE at the start of a parent's journey and during implementation. It is recommended that SHE should be delivered within a positive climate of hope. This should be instilled through a commitment by policy makers to adequately resource local provision and

run media campaigns giving positive messages about sleep. For practice, this implies a sleep training need to ensure sleep practitioners implement the wealth of sleep knowledge that is available and raise expectations that the sleep of children with DD can improve.

7.3.5. Rationing and gaming.

This study makes explicit how the adoption of rationing and gaming strategies by sleep teams can affect implementation success. In particular, within the NHS, governance and performance management by target-setting is accepted as a reality of service provision; however, underlying issues relating to gaming are sometimes overlooked. Bevan and Hood (2006) define gaming as "hitting the target and missing the point." (p.521) or the practice of organisations changing their behaviour when they know the results they report will be used to control them. Nedwick (2012) also discusses how services sometimes resort to gaming to give the appearance of achieving performance targets, and points out that "the government prefers to hear that the NHS is improving. To this extent, there are "institutional" pressures to report good statistical results" (p. 168). It is posited that health (and social service) sleep teams reduce the accessibility of their service, to deter excessive referrals and keep waiting times or referral to treatment times within acceptable targets. Support for this interpretation is also drawn from the researcher's clinical experience in NHS community teams, where initial screening appointments to families gave the impression of referrals being attended to in a timely manner.

Parent and practitioner data shows how health and social service sleep teams provide limited geographical coverage and restrict entry criteria to specific age ranges and diagnoses. It is posited that this reflects attempts to ration scarcely resourced services

and ensure performance targets are achieved, but this negatively impacts on families trying to source support. Problems with accessing professional sleep support are also reported as a concern for families of children with DD in the Sleep? What's That? report (Handsel Trust, 2007). In addition, such strategies can mask the true level of service need; an implication mirrored by Stores and Wiggs (2001a) who suggest "services will not improve until a demand for better provision is felt"(p.6). To address this, it is recommended that sleep teams offer a fully inclusive service for children aged 0-18, with any DD and with a broad geographical spread. This links with the theoretical perspective of legitimation and the need to prioritise the funding of sleep services for children with DD.

Findings also demonstrate how a third sector sleep team operated with a wide ranging referral criteria and broad geographical coverage. Whilst parents agreed this ease of access is very helpful, data also shows the wider implications of offering a fully inclusive service. In reality this meant that sleep practitioners were 'spread thinly' and could only offer minimal implementation support to families. Whereas health and social services teams who engaged with fewer families provided comparatively more intense implementation support, which parents also said was needed by some families. This shows how broader access can precipitate a support light model of implementation; effective for some parents but not all and further highlights the need for policy makers to legitimise and adequately resource service provision.

Parent and practitioner data also shows how some teams stipulate the prior endorsement of gatekeeping professionals when parents are attempting to access support. It is suggested that this added complexity reflects another rationing and

gaming strategy which restricts parent access to sleep services. The recommendation is therefore made to mirror the open access criteria operated by the third sector team represented in this study; a practice which has been shown to be very beneficial in recent Physiotherapy quality improvement initiatives. As shown by Middleton (2016), direct patient access has not lead to services being overwhelmed by inappropriate referrals; rather self-referral patients need fewer appointments as their problems are tackled sooner. It is possible that similar benefits could be seen in the care of sleep problems and children with DD, for which early intervention is always advocated (Family Fund, 2013; Robinson & Richdale, 2004; Stores & Wiggs, 2001a).

Recent policy guidance indicates that sleep support should be easily accessible. The Tired all the Time report (Family Fund, 2013) called for "clear pathways for accessing sleep advice, support and practical help at an early stage" (p. 13). NHS England (2015) guidance for commissioners also states "everyone should have access to integrated, community based, specialist multi-disciplinary health and social care support for people and/or autism in their community that is readily accessible when needed by children" (p23). This gives added support for the recommendation on a practice and policy level to simplify entry routes to sleep service provision.

In summary, findings which highlight narrow referral criteria and entry routes might be explained by rationing and gaming strategies. The wider implications of this include added barriers for parents trying to access sleep support and delays in sleep problem treatment. Furthermore, it is hypothesised that such practices mask the true level of service need. If the magnitude of sleep problems in children with DD is not visible through the volume of parents contacting sleep teams or lengthy waiting lists,

the prospect of services being adequately resourced continues to be improbable. Therefore, it is important policy makers are aware of the implications of rationing and gaming on SHE implementation and that the recommended steps are taken to enhance parents' experience of accessing professional sleep support.

7.4. Summary of recommendations.

Study findings have highlighted a range of implications for policy, practice and research as follows:

Policy:

- There is a need for national sleep screening initiatives, media campaigns spreading positive messages about sleep, reprioritisation and adequate resourcing of sleep service provision. This will foster a climate of positive health expectation and ensure children's sleep problems are legitimised.
- More understanding is needed of how the customisation of SHE fits in with the prudent healthcare agenda (Bradley & Willson, 2014) and person centred care (Sanderson & Lewis, 2012).
- A greater awareness is needed of the implications of rationing and gaming practices on SHE implementation and commitment to enhance parents' experiences of accessing professional sleep support.

Practice:

- Adequate sleep training for practitioners is needed, so they can fully understand sleep problems, customise SHE and apply up-todate sleep knowledge to practice.

- In SHE implementation, more parental involvement is needed in sleep assessment and decision making processes to faciliate knowledge exchange.

- There is a need to embedd SHE rationale and psycho-education into SHE interventions to facilitate knowledge exchange.

- Practitioners need to be encouraged to champion parent support groups to ensure parents are supported with SHE implementation.

Research:

- Continued research is needed into the experiences of families, to ensure children's sleep problems are afforded enhanced legitimacy and are current in the minds of policy makers.

- Research is needed to conceptualise the paid parent role within SHE implementation to further develop the emerging discipline of lay helping within care.

7.5 Reflexivity

Reflecting on the process of research and attempting to comprehend how one's own beliefs and views may shape findings, should be inherent in any qualitative inquiry (Jootun, McGhee & Marland, 2009). Reflexivity involves considering how our thinking came to be, how pre-existing assumptions fit with new understandings and how this influences our research. Haynes (2012) posits that "Reflexivity enables research to be insightful, questioning, intersubjective and transparent on a number of different levels" (p.77) but also argues it can perceived as narcissistic or self-indulgent where the researcher's voice is given disproportionate attention. However, it was considered very important to engage in reflexivity because participative

inquiry especially, is often influenced by politics and authority (Orr & Bennett, 2009) and there was a need to reflect on how this may have impacted on the research process.

To facilitate the reflexive process a personal journal was kept to record observations, reflections and developing ideas throughout the course of the PhD study. A report of reflexivity will now be presented through the use of the first person.

7.5.1. Nursing background.

As a researcher with a clinical nursing background in delivering and teaching about SHE, I was aware it would be difficult to separate out these roles. It was important to be aware of how my own professional background could influence the research process. From my experiences, I was always mindful of some of the limitations of SHE and challenges faced by parents and practitioners. For example, I could recall parents' disappointed reactions to standardised SHE advice and requests for additional home visits that were not always possible due to resource restraints. From my Sleep Practitioner Trainer role, I was aware of different practitioners' attitudes towards SHE and could recollect how sleep interventions were commonly implemented prior to an adequate sleep assessment. It was important to be mindful of my own such experiences and perceptions throughout the duration of study. Indeed, my clinical background sparked my interest in this study, influenced its inception and development of the research proposal. It also influenced the design of the discussion area guides and prompted additional areas of question I wanted to delve deeper into. The impact of my prior experience during the data collection phase will be explored in more detail in the next section and it is also acknowledged that my own experience and prior knowledge helped me to interpret findings. I

reflect this specialist knowledge has helped to make this research more insightful, but it has been difficult to remain as impartial as possible throughout the research process.

7.5.2. Data collection.

Keeping a reflexive journal allowed me to record my feelings after the interviews, focus groups and co-design events and consider how my skills as a researcher were developing. After reading the first interview transcription, it was clear that I spoke too much and did not give the participant enough time to respond to questions. Upon reflection during supervision, I realised I needed to work on my interview technique and become more comfortable with silence during the interview process. Subsequent journal entries reflected how I began to address this and became increasingly confident as an interviewer as I facilitated more interviews.

In developing my interview technique it was also important to reflect on issues which commonly affect nurse researchers in qualitative research such as role conflict and the wish to provide clinical advice (Jack, 2008). Jack advises researchers who are also nurses to consider a range of professional issues such as the impact of revealing their role to participants, and whether interventions should be delivered during the interview process.

At the start of each data collection event I decided to disclose my nurse background, because I felt it would be difficult to suppress my professional identity during interactions with participants. However, as Jack (2008) highlights, it is then important to reflect on the impact of this disclosure on the data collected. An extract from my reflective journal reads:

"I think because the parent knew I was a Learning Disability Nurse, she was hesitant to describe how she had co-slept with her child."

I was aware that I had to gain parents' trust in the interview process and tried to reassure them I did not intend to judge their decision making. However, I reflected that maybe some parents may have felt differently speaking with a non-nurse researcher and more able to share their experiences openly.

No life-threatening or child safe-guarding issues were raised in any of the interviews, focus or co-design groups. However, I perceived at times that some parents were in search of sleep advice, which presented me with a dilemma. In the case of Gemma and Stella who described experimenting with weighted blankets, I advised them to seek Occupational Therapist guidance to ensure correct use, once the interview and recording had stopped. Also in response to Kim's account of struggling to find professional sleep help, I felt compelled to direct her to appropriate sleep resources, again once the interview and recording had ended. It is acknowledged that if Gemma or Kim had continued their involvement in subsequent data collection events, my advice may have influenced their additional contributions, however, they were unable to participate in a second interview or co-design event and potential bias was therefore avoided.

Lastly, it is important to reflect on the tension between my current Sleep Practitioner Trainer role and the emergence of study findings. As new insights were being unearthed throughout the study, I remonstrated with the best and most appropriate way to share this information with generic practitioners attending my sleep courses. As a trainer it would have been difficult to deliver original course content and omit

to share the new knowledge I was acquiring. I decided to share some well evidenced preliminary findings, such as the functions of sleep history tools identified in the review, however other emerging findings in need of continued exploration such as SHE content, were not shared with course participants. Although challenging, this was deemed a responsible course of action.

7.5.3. The co-design method.

I found EBCD to be an effective participatory research method and experienced the 'trigger' podcast as particularly powerful in eliciting participant responses and feeding back parents' views to practitioners. However, the King's Fund toolkit gave limited guidance on how to facilitate co-design groups and keep participants focused. Similar limitations were reported by Locock et al (2014) who introduced additional formal tools such as Quality Circles (developed by the National Society of Quality through Teamwork) to ensure the groups stayed on track. In the first co-design event, parents often digressed and seemed to view it as an opportunity to discuss broader issues around disability diagnosis or education. The PATH tool was therefore introduced in subsequent events as a constant visual reminder of purpose and aims, to ensure the collection of only relevant data. This was largely successful however, co-designers still required some encouragement to limit discussions to the topic of SHE.

I also reflected on the extent to which EBCD allowed for the 'authentic' expression of the parents' voice and was concerned about them feeling unable to speak candidly in the presence of practitioners. A strength of the approach was that parents could be prepared in advance for working alongside practitioners in the first event, which may have enhanced their confidence to meaningfully participate. In addition, efforts were

made to facilitate events in a supportive way, to minimise the effects of psychosocial factors such as obedience, dominance and conformity on group processes. However, it is acknowledged that separate stakeholder groups can be more effective in identifying genuine participant views (Wainwright, Boichat & McCracken, 2014), which highlights a potential limitation of the EBCD method.

Finally, I am aware that other participatory approaches such as the Dephi method often involve service commissioners to ensure that service improvement ideas are based on cost/ resource considerations as well as clinical expertise and service user opinion (Clyne, White & Mclachlan, 2012; Snape et al.,2014). As this project was only representative of parent and practitioner views, future studies should incorporate consultation with commissioners and policy developers to enhance the feasibility of the SHE tool and programme theory.

7.6. Study Limitations

It is important to reflect on the characteristics of study participants. It could be argued that more parents, who had experienced professionally considered rather than ad-hoc SHE advice, would have provided a more informed insight into how SHE should be delivered. The inclusion criteria stated "some experience of a SHE intervention" which achieved a sample of parents with a broad variety of SHE experiences. However, it is posited that the depth of SHE experience was substituted for breadth, as the views of parents who struggled to secure sleep support were also captured which richly informed the development of pre-implementation outcomes.

Furthermore, practitioner involvement was slightly skewed towards signposting teams (six signposting and five consultation teams took part) and although their

input was vital, consultation team representatives were more vocal (as the majority of data extracts show). The involvement of more consultation teams may have produced different findings however, the mix of practitioners finally recruited, was a reflection of project time and resource constraints as well as BCUHB's decision not to participate in the co-design study.

7.7. Critical Appraisal of Thesis Findings

In qualitative research the principles of credibility, dependability, conformability and transferability are used to evaluate the trustworthiness of findings (Guba & Lincoln, 1989; Hannes, 2011; Lacey, 2010). A report now follows on the specific strategies used in this study to address these core criteria for quality and rigour.

Credibility evaluates whether or not findings truly represent the views and perceptions of participants studied. This was addressed through member checking whereby emergent findings were discussed in the supervision process and validated by participants in the second round of exploratory study interviews and co-design events. Credibility was also strengthened by the use of participant verbatim quotes during thematic analysis in all three studies presented in this thesis.

Transferability evaluates whether findings have applicability in other specific settings. The techniques were employed of providing contextual background and demographic information about study participants, to enable the reader to make judgements about the transferability of findings to different contexts. Transferability was also demonstrated through linking SHE programme theory to broader, evidenced based theories of change.

Dependability evaluates whether findings are consistent and can be repeated. This was demonstrated through the clear description of research methods employed and decisions made by the researcher. Multiple methods of data collection were utilised to ensure triangulation and an evidence trial was developed through video and audio taping and use of discussion area guides.

Confirmability evaluates how findings are influenced by the participants and not researcher bias or interest. To address this, a section on reflexivity acknowledging the effects of the researcher's perspective and clinical background on thesis findings has been presented.

7.8. Concluding remarks.

The aim of this study was to advance the knowledge base supporting SHE as a treatment for behavioural sleep problems in children with DD. Execution of the scoping review, exploratory and co-design study has provided rich findings which add to what is already known about this topic. Research objectives have also been achieved, which were to design an evidence based, systematically developed SHE tool to treat behavioural sleep problems in children with DD and develop an improved understanding of why SHE is supposed to work to improve sleep. Furthermore, the improved understanding has been presented in the form of a systematically developed programme theory linked to and evidenced by mid-range theories of change. This represents the development phase of the MRC (2000, & update 2008) framework for evaluating complex interventions. Piloting and feasibility work is now required to ensure the intervention can be carried out as intended, before a main evaluative study can be designed. The original contribution to knowledge however, is reflected in the way findings make explicit the nature of

the complexity embedded in a SHE intervention. Findings indicate how the legitimation of sleep problems is a foundation on which SHE implementation should be based. In addition, it demonstrates the nature of customisation, knowledge exchange, health expectation and the impact of rationing and gaming on implementation success. Lastly, a discussion has been presented of how these findings can now influence research, policy and practice and ultimately enhance the delivery of SHE for families of children with DD and behavioural sleep problems. In conclusion, this represents a novel investigation which pulls together a scoping review and different stakeholder perspectives to systematically develop a SHE tool and underpinning programme theory. It is sincerely hoped thesis findings will be picked up, utlised in future practice, policy and research and help to improve sleep provision for families of children with DD.

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APPENDICES

Appendix 1 – Included studies (n=52).

1	Adkins, Goldman et al.(2012)	Sleep assessment study
2	Adkins, Molloy et al. (2012)	Randomised controlled trial
3	Allison, Burke and Summers (1993)	Quasi experimental single case design study
4	Autism Society (2012)	Bespoke SHE tool for children with ASD
5	Bartlet and Beaumont (1998)	Before and after study
6	Beresford, Stuttard, Clarke, Maddison and Beecham (2012)	Research report. Pilot randomised controlled trial, two uncontrolled pre-post studies, qualitative focus group and 1:1 interviews.
7	Bramble (1996)	Observational cohort study
8	Bramble (1997)	Before and after study
9	Capability Scotland (2012)	Bespoke SHE tool for children with DD
10	Catherall and Williams Jones (2011)	Bespoke SHE tool for children with DD
11	Cerebra (2011)	Bespoke SHE tool for children with DD
12	Christodulu and Durand (2004)	Multiple baseline across participants experiment
13	Chronic Care for Sleep (2012)	Bespoke SHE tool for children with DD
14	Contact a Family (2007)	Bespoke SHE tool for children with DD
15	Cortesi, Giannotti, Ivanenko and Johnson (2012)	Randomised controlled trial
16	Disabled Living Foundation (2012)	Bespoke SHE tool for children with DD
17	Down's Syndrome Association (2008)	Bespoke SHE tool for children with Down's syndrome.
18	Durand (2011)	Book chapter detailing a bespoke SHE tool for children with DD
19	Durand (2013a)	Book chapter detailing a bespoke SHE tool for children with DD
20	Durand (2013b)	Book chapter detailing a bespoke SHE tool for children with DD
21	Early Support (2010)	Bespoke SHE tool for children with DD
22	Goodlin-Jones, Sitnick, Tang, Lui and Anders (2008)	Sleep assessment study
23	Handsel Project (2012)	Bespoke SHE tool for children with DD
24	Hewitt (1985)	Case series study
25	Johnson, Wise and Jimmerson (1995)	Bespoke SHE tool for children with DD
26	Johnson, Turner, Foldes, Malow and Wiggs (2012)	Sleep assessment study
27	Khan and Underhill (2006)	Sleep assessment study
28	Leeds NHS Community Healthcare (2010)	Bespoke SHE tool for children with DD
29	Lester (2012)	Bespoke SHE tool for children with ASD
30	Maas et al.(2011)	Sleep assessment study
31	Malow et al.(2009)	Sleep assessment study
32	Malow et al.(2013)	Randomised controlled trial
33	Middlesborough Moving Forward (2011)	Bespoke SHE tool for children with DD
34	Montgomery, Stores and Wiggs (2004)	Randomised controlled trial
35	National Autistic Society (2008)	Bespoke SHE tool for children with ASD
36	O'Connell and Vanaan (2008)	Pilot study
37	Peak Interventions (2012)	Bespoke SHE tool for children with ASD
38	Petta, Pilestead, Green and Gresley-Jones (2012)	Bespoke SHE tool for children with ASD
39	Piazza, Fisher and Sherer (1997)	Randomised controlled trial
40	Raising Children Australia (2012)	Bespoke SHE tool for children with DD
41	Redwood (2012)	Bespoke SHE tool for children with DD
42	Reed et al.(2009)	Uncontrolled pre-post test study, single group design
43	Research Autism (2007)	Bespoke SHE tool for children with ASD

44	Scope (2012)	Bespoke SHE tool for children with DD
45	Sense (2012)	Bespoke SHE tool for children with DD
46	Sitnick, Goodlin-Jones and Anders (2008)	Sleep assessment study
47	Sleep Practitioners (2012)	Bespoke SHE tool for children with DD
48	Stores and Stores (2004)	Randomised controlled trial
49	Stores and Wiggs (2001)	Book chapter detailing a bespoke SHE tool for children with DD
50	Teel (2012)	Bespoke SHE tool for children with DD
51	Weiskop, Matthews and Richdale (2001)	Case study
52	Weiskop, Richdale and Matthews (2005)	Multiple baseline across participants study.

Appendix 2 – Search strategies.

Cinahl with full text via ebsco 11.1.13

1. exp. Sleep/ (8103)
2. dyssomnias CL, DI, NU, PC, PF/ (20)
3. sleep disorders CL, DI, NU, PF/(1046)
4. ("sleep problem*" or "sleep disturbance*" or "sleep disorder*" or "sleep hygiene").ti, ab (3963)
5. **or/1-4 (combines sleep terms) (11572)**
6. exp child/ (268076)
7. child*. ti, ab (160403)
8. young adult(unable to explode)/ (24740)
9. "young adult*". ti, ab (8380)
10. **or 6-9 (combines child terms) (336875)**
11. developmental disabilities (unable to explode) /(3714) or exp mental retardation/ (12236) or exp child developmental disorders pervasive/ (8413) or exp child development disorders/(1451) (24276)
12. asperger syndrome (unable to explode) /(   ) or autistic disorder(unable to explode)/ (7734) or cerebral palsy(unable to explode)/ (5282) or down syndrome(unable to explode)/(2769) (   )
13. ("learning disab*" or retard* or "intellectual* impair*" or "special needs" or "multiple disab*") ti, ab (9879)
14. (asperger* or autis* or "cerebral palsy" or "down*syndrome") ti, ab (   )
15. **or/11-14 (combines disability terms) (36202)**
16. **5 and 10 and 15 (182)**

Psychinfo (proquest) 17.1.13

1. exp sleep/ (17149)
2. exp sleep deprivation/ (2944)
3. exp sleep disorders/(9682)
4. ("sleep problem*" or "sleep disturbance*" or "sleep disorder*" or "sleep hygiene").ti, ab (9078)
5. **or /1-4 (combines sleep terms)(28936)**
6. exp childhood development/(63239)
7. child*. ti, ab (484576)
8. exp adolescent development/(27603)
9. "young adult*". ti, ab (25138)
10. **or /6-9 (combines child terms) (533546)**
11. exp developmental disabilities/(10560) or exp intellectual development disorder/(34232) or exp pervasive development disorders/ (23001) (63681)

12. exp autism/ (18432) or exp cerebral palsy/ (3096) or exp down's syndrome/ (4743) (25779)
13. ("learning disab*" or retard* or "intellectual*impair*" or "special needs" or "multiple disab*").ti, ab (58768)
14. (autis* or "cerebral palsy" or "down*syndrome"). ti, ab (29808)
15. **Or /11-14 (combines disability terms) (106812)**
16. **5 and 10 and 15 (444)**

ASSIA (Applied Social Science Index and Abstract) via Proquest 24.1.13

1. exp sleep/(1838)
2. exp sleep deprivation/(100)
3. exp sleep disorders/(735)
4. ("sleep problem*" or "sleep disturbance*" or "sleep disorder*" or "sleep hygiene")ti, ab (928)
5. **Or/1-4 (combines sleep terms) (2452)**
6. exp child development/(824)
7. child* ti, ab (85150)
8. exp adolescence/(1252)
9. "young adult*" ti, ab (5028)
10. **Or/6-9 (combines child terms) (89868)**
11. exp developmental disorders/(300)
12. exp autistic spectrum disorders/(1351) or exp cerebral palsy/(608) or exp down's syndrome/(694) (2637)
13. ("learning disab*" or retard* or "intellectual* impair*" or " special needs" or "multiple disab*") ti, ab (5956)
14. (autis* or "cerebral palsy" or " down* syndrome") ti, ab (5521)
15. **Or/11-14 (combines disability terms) (11940)**
16. **6 and 12 and 15 (44)**

Cochrane Database of Systematic Reviews- searched 28.2.13

1. Sleep deprivation (1) – not relevant
2. Sleep disorder (14) – none relevant
3. Sleep hygiene (2) – none relevant
4. Sleep and children (25) – none relevant
   42

Cochrane Central Register of Controlled Trials 28.1.13

1. Sleep and children 2011 onwards – none relevant

Ovid Nursing Full Text Plus. 24.1.13

1. exp sleep/ (4930)
2. exp dyssomnias/(2114)
3. exp sleep disorders/(3204)
4. ("sleep problem*" or "sleep disturbance*" or "sleep disorder*" or "sleep hygiene") ti,ab (1109)
5. **Or/1-4 (combines sleep terms) (6430)**
6. exp child/(113371)
7. child* ti, ab (57964)
8. exp young adult/(3044)
9. "young adult*"ti, ab (2266)
10. **Or 6-9 (combines child terms) (122823)**
11. exp developmental disabilities/(2703) or exp mental retardation/ (5220) or exp pervasive child development disorders/(1131) or exp child development disorders/ (1174)(7598)
12. exp autistic disorder/(1024) or exp cerebral palsy/ (3320)or exp down syndrome/(973) (5238)
13. ("learning disab*" or retard* or "intellectual*impair*" or "special needs"or "multiple disab*") ti, ab (3795)
14. (autis* or "cerebral palsy" or "down*syndrome")ti,ab(3513)
15. **0r/11-14 (combines disability terms) (12906)**
16. **5 and 10 and 15 (85)**

Web of Knowledge search 4.2.13

1. sleep topic (354403)
2. dysomnia* topic (10)
3. ("sleep problem*" (6273) or "sleep disturbance*" (20891) or "sleep disorder*" (43539) or "sleep hygiene*"(822)) topic
4. **or/1-3 (combines sleep terms) (354406)**
5. child* topic (3563506)
6. "young adult*" topic (398559)
7. **or/2-3 (combines child terms) (3866890)**
8. ("developmental disab*" (22896) or "intellectual disab*" 60260() or "intellectual impair*" (1783) or "child development disorder* (3223) or "learning disab*" (24169) "mental retard*" (103000) or "special needs"(7121) or "multiple disab*" (1022) topic
9. (autis* (69119)or "cerebral palsy*"(43400) or "down* syndrome" (54443)topic

10. **or/ 8-9 (combines disability terms) (54443)**
11. **4 and 7 and 10 (2053)**
12. **Limit 11 to 2011 and 2012 pub year (328)**

Conference paper search web of science, 4.2.13

1. Sleep (conference) refined by child (865) and "disab*" (3) (1 relevant)
2. Sleep (conference)refined by child (865) and web of science categories – psychology and paediatrics (21) (none relevant)
3. Sleep (conference)refined by child (865) and autis*(41)(19 relevant)
4. Sleep (conference)refined by child (865) and retard* (4) (1 relevant)
5. Refined by all other disability terms no results

Google Scholar search 4.2.13 – limited to 2009 onwards

1. "sleep hygiene" and "disabled children" (54)

Google Scholar search 4.2.13

1. "sleep hygiene" child* disab* conference papers limited to 2011 onwards(70)

Appendix 3 – Data charting form.

Abbreviation Key: **HV**- Home visit, **TSP**- Telephone support, **CDC** – Child development centre, **CBT**- Cognitive behavioural therapy, **CSHQ** – Child sleep habit questionnaire, **TCSHQ** – Total CSHQ score– **ACT-** Actigraphy, **GAS** –Goal attainment scores, **FISH** – Family inventory of sleep habits, **ASPS –** Albany sleep problems scale, **PSOC** – Parenting sense of competence.

No.	Study details	Population & Setting	Methods	Intervention	Outcomes	Results
1	Adkins, Goldman et al (2012) Sleep assessment study. Compares shoulder and wrist location of ACT to assess sleep problems in ASD children.	US study 8 ASD children aged 6-10 yrs – 7 boys and 1 girl. Participants not similar at start of trial- 7 children on melatonin throughout trial Small sample size, difficult to generalise results	Children wore ACT devices simultaneously in two locations. (a) non-dominant wrist location ((b) non-dominant shoulder location for 7 nights.	Assessment ACT sleep assessment *"AW-64 Actiwatch device (Phillips Respironics, Bend, OR). Each device contains an accelerometer, which is able to detect motion > 0.01 G-force in all directions, and translates it into an electrical signal"* Parents instructed to activate interval button at lights out and upon waking and record times on a sleep diary.	Outcomes: ACT: total sleep time, sleep onset latency, sleep efficiency, and wake after sleep onset (sum of all wake episodes during the sleep period)	Correlation between wrist and shoulder recordings for Sleep onset latency – p.058 Total sleep time – p.0.07 Sleep efficiency – p 0.09 Wake after sleep onset – p 0.05 Showing no statistical difference between 2 sites and therefore agreement No confidence limits reported. Study concluded shoulder placement offered a feasible alternative site for actigraphy in ASD children
2	Adkins, Molloy et al (2012) Randomised controlled trial. Aim – to determine whether a sleep education pamphlet alone could help with a child's insomnia. SHE intervention study	Sample - 36 children with ASD, (18 received SHE) ages 2 to 10 yrs, 67% male, with prolonged SL confirmed by ACT. US study.	Assessment: All children screened by Paediatrician to exclude co-morbid conditions affecting sleep, but no sleep history reported. CSHQ and ACT used to assess Sleep latency at start of study. Parents trained & examined in ACT use.(wrist and shoulder placement used) Randomised to receive pamphlet or no intervention. Intervention group	SHE components: Sleep timing Bedtime routines Behavioural management Environment Physiological factors	Outcome measures: CSHQ Sleep dairies, ACT collected for 21 days over course of trial for both groups. Intervention group also completed a feedback questionnaire. Outcome measures repeated for 2 weeks post intervention. Outcomes: ACT total sleep time, sleep onset latency, sleep efficiency, and wake after sleep onset (sum of all wake episodes during the sleep period)	Effectiveness: Sleep efficiency was the only sleep parameter showing statistically significant change with treatment-P value 0.04. No confidence intervals stated. Assessment: ACT wrist and shoulder placement found to be equally reliable. Parent experience: *"They (*parents) *indicated that what might have been more useful would have been to have more specific ideas of how to take the information and put it into practice"* Page 143

				CSHQ, TCSHQ	
3	Allison et al. (1993) Quasi experimental single case design study Aim- To evaluate the effectiveness of non-aversive behavioural intervention to treat sleep problems in a child. MCSHE intervention study	Sample- 1 girl with Down's Syndrome aged 8 years. US study. Sleep problems- tantrums at bedtime and co-sleeping with mother	Assessment Baseline data recorded over 15 days, but method of assessment not reported. Child received two phases of treatment Phase one- 26 days- SHE Phase two- 30 days- Graduated extinction and stimulus control	SHE components: Bedtime routines Communication adaptations Plus graduated extinction and stimulus control	Outcomes: Sleeping alone in bed	Effectiveness At Baseline the child spent 6% of the time alone in bed (SD 14.7) after first treatment phase (SHE) this rose to 26% (SD 38.2). After the second treatment phase this rose to 78.2% (SD 7.4). No P values or Confidence intervals stated.
4	Autism Society (2012) Bespoke SHE tool for children with ASD.		Assessment Emphasises the importance of ruling out other sleep disorders including sleep related breathing disorder, medical issues and sensory needs, but no assessment formats recommended.	10 sleep tips SHE Components: Sleep timing Bedtime routines Environment Communication adaptations		
5	Bartlet and Beaumont (1998) Before and after study Report on a 1 year sleep project with DD children MCSHE intervention study	UK study (Southampton) Carried out at home and clinic setting Sample- 57 children 40 boys and 21 girls mean age 4.8 years.	Assessment SSMS. Sleep disturbance index (SDI) –Sleep diaries Medical investigations deemed relevant (EEGs) were carried out. Intervention begins with up to 2 contacts (clinic or home visit lasting 1-2 hours each) then up to 5 telephone calls (lasting between 5 -60 mins each) 3- 6 month follow up.	SHE components SHE components- Bedtime routines Behavioural management Intervention also includes- Extinction Medication used concurrently in 8 children to treat sleep problems	Outcome measures & outcomes SDI scores Q.O.L measure GHQ 30 scores	Effectiveness Fall in SDI scores p= 0.000. Fall in GHQ 30 scores p=0.00 (Report resulted in the establishment of a once a month sleep clinic at paediatric dept. Staffed by Paediatrician, child psychologist, special needs nurse and health visitor. Access available to polysomnography also. Experience Facilitators- Professionals need to adopt a supportive and understanding approach, build relationships with families Barriers- Physical exhaustion of parents, marital conflict, separation anxiety
6	Beresford et al. (2012) Research report, chapter 14 Intervention E (p190)	Sample: 15 children with DD, including ASD (all 15 received MCSHE) 6 children HV group mean age 2.67 years(6 males) 7 children TSP group mean age 2.86 (5	Assessment Sleep assessment/history (unspecified) taken. Sleep dairies, CSHQ Detailed assessment by CDC to rule out physiological causes, sleep dairy	SHE components Environment Bedtime routines Sleep strategy also includes other intervention: Extinction techniques	Outcome measures GAS, up to 3 goals per family.10 point rating scale, max 10 fully achieved. CSHQ, sleep dairy Q.O.L. measures PSOC Sleep outcomes GAS CSHQ	Effectiveness Mean GAS pre- 12 week f/up-HV 2.15- 6.50 and TSP 2.33- 9.38. Mean TCSHQ pre-post. HV 59.50-52.17, TSP 53.33- 53.33. Mean TCSHQ pre-12 week f/up- HV 59.50- 49.0, TSP 53.33 – 49.25 Mean PSOC Pre-post Efficacy- HV 30.83-30.80, TSP 31.33-27. Mean PSOC Pre-12 week f/up Efficacy –HV

	"*Specialist Health Visitor Sleep Support Programme*" Pilot randomised controlled trial MCSHE intervention study	males) UK study, home setting and child development centre Aim – To evaluate individually delivered HV versus telephone support TSP as a mode of support for parents implementing sleep strategy.	Contact a family leaflet given in some cases. Families receive initial HV from health visitor then randomised into HV or TSP group, weekly basis, over 6 weeks. No waiting list control group.		TCSHQ, bedtime resistance, sleep anxiety and night awakenings Q.O.L outcomes Satisfaction, Efficacy	33.75-30.80, TSP 28.00-27.86 Mean PSOC pre-12 week f/up Satisfaction- HV 44.25-36.80, TSP 29.25-31.17 P values not reported. 95% confidence interval reported graphically but not clearly displayed. Study concluded parent goals and CSHQ scores improved following both interventions however small sample size prevents evaluation of relative effectiveness.
	Beresford et al. (2012) Research report, chapter 15 Intervention G (p209) "*Managing your child's behaviour to promote better sleep*" Un controlled pre-post study MCSHE intervention study	Sample: 22 children (with and without DD) (all 22 received MCSHE) Mean age 8.91 years, 11 males UK study setting not specified Aim – to test efficacy of group based parenting sleep intervention for treating sleep problems	Assessment CSHQ, sleep dairies No sleep history/ sleep assessment reported. 4 group sessions delivered over 5 week period – with supporting manual	SHE components Bedtime routines Environment Additional unspecified interventions	Outcome measures GAS, up to 3 goals per family.10 point rating scale, max 10 fully achieved. CSHQ Q.O.L measures PSOC Outcomes GAS CSHQ TCSHQ, Bedtime resistance sleep anxiety, night awakenings Q.O.L outcomes Efficacy, satisfaction.	Effectiveness GAS pre int. to 24 week f/up- Mean 2.10 to 5.35 – statistically significant change p value <0.001 PSOC Efficacy pre int. to 24 week f/up – Mean 26.48- 29.39- statistically significant change p value <0.001 95% confidence intervals also reported graphically but not clearly displayed. Study concluded intervention was effective in treating sleep problems.
	Beresford et al. (2012) Research report, chapter 16 Intervention H (p223) "*Sleep solutions Time to Sleep*" Un controlled pre-post study MCSHE intervention study	Sample- 25 children DD mean age 7. 16 males (all 25 received MCSHE) Uk study setting not specified Aim- to test efficacy of a 1 day sleep management workshop intervention for treating sleep problems	Assessment CSHQ No sleep history/ sleep assessment reported Single day workshop delivered.	SHE Components Environment Bedtime routines Plus simple cognitive behavioural therapy techniques.	Outcome measures GAS, up to 3 goals per family.10 point rating scale, max 10 fully achieved. CSHQ, Q.O.L measures PSOC Outcomes GAS, CSHQ, TCSHQ, Bedtime resistance sleep anxiety, night awakenings Q.O.L outcomes Efficacy, satisfaction	Effectiveness Mean GAS improved from 2.47 to 5.92 at 24 week f/up statistically significant change p value <0.001. 95% confidence intervals reported graphically, but not clearly displayed. Study concluded there was some evidence to support this as an effective intervention
	Beresford et al.(2012) Research report, chapters 5, 13, 17 and 18. Collection of qualitative focus	Explores views of parents and practitioners of 1:1, group and workshop based sleep interventions UK based study	Sample:35 Parents who had received a sleep intervention, 15 Practitioners who had delivered sleep interviews. Participants selected using purposive sampling frame	SHE Components Pertains to MCSHE interventions listed in chapters 14, 15 and 16.	.	Experiences Parents and practitioners views around the take-up of sleep interventions and outcomes were reported.

	groups and 1:1 interviews MCSHE intervention study		Data collected by face-to-face, telephone and focus group interviews and were audio-recorded with permission			
7	Bramble (1996) Observational cohort study. Structured questionnaire MCSHE intervention study	Explores parent's views of MCSHE . Study produced experience data although was not qualitative in design. Behaviour intervention carried out at home full report in Bramble (1997). 18 month evaluation by post or telephone. UK based study	Sample: 15 children 10 boys and 5 girls. Mean age 7.2 years. All had severe learning disabilities (SLD) and lifelong severe night settling and night waking problems Definition Night settling and night waking problems (frequency & duration unspecified)	SHE Components Sleep timing Bedtime routines Behaviour management Plus rapid extinction Theory *The theory is that complete withdrawal of the primary reinforcing element (i.e. the parents' attention) will lead to rapid extinction of the child's demands while cueing the child into a regular sleep pattern by employing stimulus control techniques and promoting good sleep hygiene"* p. 358	Outcome measures Evaluation questionnaire designed for the study Outcomes Form of prior help for sleep problems Quality of prior help Changes in parent rating of child's sleep problem severity scores Acceptability of approach Overall levels of treatment satisfaction What advice components worked How soon changes seen	Experiences & effectiveness Two thirds of parents had received prior help, forms listed. Quality of prior help Mean score of 1.4 (SD 2.5 range 0-7). Sleep problem score severity Mean score SS 2.3 (SDI 1.9 , range 0 to 5) at the end of treatment Acceptability of approach- 12 families found the treatment just right and 3 families considered it rather tough but were willing to continue. Overall levels of treatment satisfaction- end of treatment score, mean 8.6 SD 1.6. Mean scores of helpfulness of advice components listed. How soon changes seen reported as after 3.6 nights (SD 1.9 range 1 to 7 nights) No P values or confidence intervals reported. The paper concluded the treatment was successful and acceptable to parents.
8	Bramble (1997) Before and after study MCSHE intervention study	Sample: 15 children with severe learning disabilities and chronic sleep disorders. (15 received MCSHE) 3.5-12 years. 2:1 male female ratio. UK study, carried out in clinic or home setting.1:1 session with author therapist. Aim: test the efficacy of behavioural sleep intervention	Assessment Visual analogue scale – 0-100, 100 indicating most severe sleep problems, reported by parents and author, designed for the study. Sleep diaries.Data recorded at pre-treatment phase, 2 week post treatment, 4, then 18 month follow up A single face to face advice session (home or clinic) with follow up advice phone-calls.	SHE Components Sleep timing Bedtime routines Also includes : Extinction	Outcome measures Sleep dairies , visual analogue scale Q.O.L measures- Maternal sleep scale, Malaise inventory, Behaviour problem index Outcomes Parent ratings of severity of sleep problems. Principle investigators ratings of sleep problems.	Effectiveness Parent ratings of severity of sleep problems pre to 18 month f/up 80.5 – 32. P <0.001 Principle investigators ratings of sleep problems pre to 4 month f/up 8.4 - 1 1 P<0.01. Study concluded the intervention was effective in reducing sleep problems. Confidence limits (although % not specified) reported graphically, with significant spacing between pre – treatment and 4 month follow up periods, indicating good precision.
9	Capability Scotland (2012) Bespoke SHE tool for children with DD		Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem.	SHE Components Sleep timing Bedtime routines Behavioural management Environment		

			Recommends sleep diaries.	Physiological Communication adaptations		
10	Catherall and Williams Jones (2011) Bespoke SHE tool for children with DD		Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem. Recommends sleep diaries.	SHE Components Sleep timing Bedtime routines Also includes sleep restriction, scheduled awakening , graduated extinction		
11	Cerebra (2011) Bespoke SHE tool for children with DD		Assessment Cerebra sleep service questionnaire- sleep interview	SHE Components Sleep timing Bedtime routines Behavioural management Environment Physiological Communication adaptations		
12	Christodulu and Durand (2004) Multiple baseline across participants experimental design MCSHE intervention study	Sample- 4 children with developmental disabilities aged 2-5 years(4 received MCSHE).US study Carried out in children's homes Aim : test efficacy of behavioural sleep interventions to reduce sleep difficulties in children with DD	Assessment ASPS, diaries and behaviour logs.All children assessed by paediatrician to exclude associated medical conditions known to affect sleep.ACT used to sample data integrity. X4 participants received positive bedtime routines which included SHE and x3 also received sleep restriction.	SHE Components Bedtime routines Sleep timing Behavioural management Also includes extinction. Sleep Restriction was the 2nd intervention applied	Outcome measures Sleep diaries Outcomes Total sleep time, bedtime resistance, night awakenings. Q.O.L measures Parental sleep satisfaction questionnaire.	Effectiveness Study concluded the intervention was effective in reducing sleep problems in all 4 children.X1 child had positive bedtime routines (SHE) only applied (Joey): Mean Bedtime resistance score reduced from 6.5 to 2.5 per week. Mean night awakenings score reduced from 9.29 to 1.50 per week. P values and confidence limits not reported.
13	Chronic Care for Sleep (2012) Bespoke SHE tool for children with DD			15 sleep tips – SHE Components Sleep timing Bedtime routines Behaviour management Environment Physiological Communication adaptations.		
14	Contact a Family (2007) Bespoke SHE tool for children with DD		Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem. Recommends sleep diaries	SHE Components Sleep timing Bedtime routines Behaviour management Environment Physiological Communication adaptations		
15	Cortesi et al.(2012)	Sample – 160 children aged 4-10	Assessment. CSHQ, ACT and sleep	SHE Components: Sleep timing	Outcome measures: CSHQ, sleep diaries and ACT	Effectiveness: Children in all three treatment groups were

	Randomised controlled 12week trial.

MCSHE intervention study | years with sleep-onset or maintenance insomnia.(of which 134 suitable for analysis) (68 received MCSHE)

Study carried out in Italy.
CBT carried out at out-patient university clinic.
Aim – to assess the relative and combined efficacy of CR melatonin and cognitive behaviour therapy (CBT) for treating sleep disorders in children with ASD. | diaries used to assess 7 nights of baseline sleep. Polysomnography performed to rule out associated medical conditions. Associated behaviours and psychiatric conditions excluded through completion of Child behaviour checklist. No sleep history/sleep assessment reported. Children randomly assigned to either Combination group (Comb)- controlled release (CR) melatonin and CBT x4 sessions (n=-35) Melatonin group (MLT) – CR melatonin(n=34) CBT group (CBT) – x4 CBT sessions (n=-33). Placebo group (Plac) – Placebo drug (n=32). For 12 weeks in a 1:1:1:1 ratio | Bedtime routines Behavioural management CBT also includes extinction Other intervention groups: Melatonin Melatonin and CBT combined Theory- "Healthy sleep practices also promote sleep and enhance sleep regulation by reducing environment stimulation and behavioural sleep conditioning, which reinforce the association of certain activities and environments with sleep, limit wake-promoting activities and may play a crucial role in sleep promotion."p707 | used after 12 weeks of treatment. Outcomes: CSHQ TCSHQ, bedtime resistance, sleep onset delay, sleep anxiety, night awakenings, sleep duration, daytime sleepiness, sleep disordered breathing, parasomnias ACT Total sleep time, sleep latency, wake after sleep onset, sleep efficiency, naptime, bedtime | more improved than those in the placebo group at the 12-week assessment (P > 0.01) according to actometry data and CSHQ scores.

Children whose sleep onset latency was reduced by 30 mins or reduced by 50% at 12 week assessment was 84.62% comb. 39.39% MLT, 10.34% CBT group.

The study concluded comb group was most effective in reducing insomnia symptoms, followed by MLT and the CBT group.

Confidence limits reported graphically, with some overlapping indicating reduced precision and less significant difference between intervention groups. |
| 16 | Disabled Living Foundation (2012)

Bespoke SHE tool for children with DD. | | | 11 sleep tips SHE components Bedtime routines Behavioural management Environment Communication adaptations | | |
| 17 | Down's Syndrome Association (2008)

Bespoke SHE tool for children with Down's Syndrome | | Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem.

Special section on sleep related breathing disorders | A sleep advice leaflet for parents detailing SHE and graduated extinction. SHE components Sleep timing Bedtime routines Behavioural management Environment | | |
| 18 | Durand (2011)

Book chapter detailing a bespoke SHE tool for children | | Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem. | SHE components Sleep timing Bedtime routines Environment Physiological | | |

	with DD.		Use of sleep diaries.			
19	Durand (2013a) Book chapter detailing a bespoke SHE tool for children with DD.		Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem. Use of sleep diaries.	SHE components Sleep timing Bedtime routines Environment Physiological Communication adaptations		
20	Durand (2013b) Book chapter detailing a bespoke SHE tool for children with DD.		Assessment ASPS Emphasises importance of assessment and eliminating physical causes of sleep problem. Use of sleep diaries	SHE components Sleep timing Bedtime routines Environment Physiological Communication adaptations		
21	Early Support (2010) Bespoke SHE tool for children with DD.		Assessment Emphasises importance of assessment and eliminating physical causes of sleep problem. Use of sleep diaries.	40 page document for parents giving sleep advice. SHE components Sleep timing Bedtime routines Behaviour management Environment Physiological Communication adaptations		
22	Goodlin-Jones et al.(2008) Sleep assessment study Studies the use of the CSHQ in a sample of TD and ID 2-5 ½ year old children	US study.carried out in children's homes. Diverse demographic sample of 194, 2- 5 1/2 year old children (121 in toddler cohort and 73 in preschool cohort) 74% males, then collapsed into a single group and divided into problem sleep (*n* _ 79) and non-problem sleep groups (*n* _ 124) according to parent report..	ACT and sleep diaries recorded for 7 days and CSHQ taken during this time. .	CSHQ – sleep assessment 52 item questionnaire related to sleep behaviour in children originally developed for 4-10 yr olds. Uses Likert-type scale to assess frequency in last week. .	Non sleep problem and sleep problem group results compared Results compared with 33 items grouped to form 8 subscales/ outcomes: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night awakenings, Parasomnias, Daytime Sleepiness and Sleep Disordered Breathing.	Total CSHQ scores for non sleep problem group – mean 45.98, problem group mean 54.87. p value 0.000 showing a statistical difference between groups and CSHQ is effective in detecting sleep problems in 2-5 ½ year old children. No confidence limits reported. .
23	Handsel Project (2012) Bespoke SHE tool for children with DD		Assessment Handsel Project sleep interview schedule and sleep diary	Checklist of SHE tips SHE components Sleep timing Bedtime routines Behavioural management Environment		

				Physiological Communication adaptations		
24	Hewitt (1985) Case series study MCSHE intervention study	UK study Aim to evaluate effectiveness of behavioural sleep interventions	Sample- 10 children with learning disabilities and sleep problems Assessment Unspecified interview and sleep diary. Following assessment parents were given tailored behavioural advice	SHE components Bedtime routines Also included graduated extinction	Outcomes Not clearly reported; different for each case.	Results Treatment was immediately effective in 8 children. No P values or confidence intervals reported.
25	Johnson et al. (1995) Bespoke SHE tool for children with DD.	US paper	Assessment Reports a bespoke sleep history tool	SHE components Bedtime routines Behavioural management Environment Sleep timing Physiological		
26	Johnson et al.(2012) Sleep assessment study Study comparing CSHQ (39 item) with MSPSQ (shortened version) (Sleep interview and outcome measure) in children with ASD.	US study, setting not specified. Dominance of males in sample 86.3%	124 Parents of ASD children 2-16 years completed both CSHQ and M SPSQ.	Sleep assessment. CSHQ - . Abbreviated 39 item version used. MSPSQ shortened version- 58 item.(Sleep interview and outcome measure)	Outcomes Total MSPSQ scores and total CSHQ scores plus CSHQ sub scales.Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night awakenings, Parasomnias, Daytime Sleepiness and Sleep Disordered Breathing.	The correlation for the total scores of the two measures was .70 (p 0 .01). Indicating the 2 measures were highly correlated. 95% confidence limits reported on total scores: CSHQ 47.57-60.6 MSPSQ 64.52-69.72 No overlapping indicating good precision.
27	Khan and Underhill (2006) Sleep assessment study Study detailing 2 part sleep outcome measures –CPQ commissioned by Scope, to obtain sleep information and associated medical factors in children aged 2-10 yrs with Cerebral palsy (CP).	CP Group- 28 10 female, 18 male, mean age 6yrs, age range 2-10 yrs. Able bodied Control Group- 30 16 female, 14 male, mean age 6yrs, age range 2-10 yrs. UK study No setting stated	Outcome measure CPQ completed with children and parents at Time 1 and 1 month later Time 2. Parents also asked to keep sleep dairy for 1 week and repeat 1 month later Age matched, able bodied control group. .	2 part sleep outcome measure, CPQ: Sleep section 3 parts: Bedtime routine Night-time behaviour Breathing quality during sleep Medical section: Identified medical conditions of child	Difference in responses to questionnaire at Time 1 and Time 2 were compared. Outcomes: Sleep latency Night awakenings Night awakenings with pain Position changes through night	CP children scored significantly higher than control group in bedtime routine section (U = 248.5, two-tailed p<.01) & night-time behaviour section (U=42, p<.001), inferring increased sleep problems. No confidence limits reported. Shows CPQ produces statistically significant differences between groups and is a reliable tool in detecting sleep problems in children with CP.
28	Leeds Community Healthcare (2010) Bespoke SHE tool			Sleep advice leaflet for parents SHE components Bedtime routines		

	for children with special needs			Environment Sleep timing Physiological		
29	Lester (2012) Bespoke SHE tool for children with ASD.			10 sleep tips, includes stimulus control SHE Components: Behavioural management Environment Physiological Communication adaptations		
30	Maas et al.(2011) Sleep assessment study Study examining psychometric properties of Part 4 of the (MSPSQ) with individuals with intellectual disabilities (ID)-outcome measure	Netherlands study. Setting - questionnaires sent to parents or professional care givers by mail. -Good sample size of 345 individuals with ID-Sleep clinic group -Control group taken from range of ID care settings -Age range of sample – 15 mths-66 yrs	A range of recruitment procedures were followed for each group. MSPSQ completed by parents or professional care-givers -Exploratory & confirmatory factor analysis performed to define factor structure of MSPSQ.	.Sleep assessment: Part 4 of the MSPSQ outcome measure assesses the frequency of occurrence of 45 sleep behaviours (refer appendix) (3 items (15,22,29) were deleted because of insufficient reliability)	Outcomes Total MSPSQ score Range 42-294 based on 42 items. -Assessed degree to which MSPSQ identified differences between control group individuals with ID (no prior sleep complaint identified) and individuals with ID referred to a sleep clinic.	Total MSPSQ score for control group 76.81 mean and sleep clinic group 91.24 - p level 0.001, indicating the MSPSQ shows statistically significant difference between groups, and MSPSQ is effective in detecting sleep problems in individuals with ID -Concluded the MSPSQ appears to be a valid and reliable tool for assessing sleep problems in individuals with ID -Heterogeneity of sample limits validity of results No confidence limits reported
31	Malow et al. (2009) Sleep assessment study Study testing the reliability and validity of (FISH) (12 item) outcome measure with 2 groups of children.	157 children between 3- 10 yrs. ASD group – N 93 13% taking psychotropic medications. 90% male. Typically developing (TD) control group – N 64 5% taking psychotropic medications. 59% male. US study. No setting stated.	Parents completed FISH (12 item) and (CSHQ) (33 item) Items selected for FISH tool based on *"daytime behaviours known to influence sleep that would also be amenable to change with treatment"*p20	12 item FISH sleep outcome measure (detailing sleep habits only), CSHQ.	Total FISH scores for each group were correlated with the following CSHQ outcomes: Bedtime resistance, Sleep onset delay Sleep duration Sleep anxiety Night- awakenings Parasomnias Sleep disordered breathing Daytime sleepiness	Total CSHQ score for ASD and TD control group p value 0.000 Total FISH score for ASD and TD group p value 0.000 Indicating both sleep assessments show statistically significant differences between groups No confidence limits reported Total FISH and CSHQ scores for ASD group p value 0.002, for TD group 0.004. Showing statistically significant correlation between the 2 measures.
32	Malow et al.(2013) Randomised controlled trial MCSHE intervention study	Sample: 80 children 2-10 years with sleep onset delay (80 received MCSHE) US study carried out at 3 clinic sites. Aim: Comparing efficacy of sleep	Assessment Outcome measures-ACT, CSHQ (33 item), sleep diaries, FISH. Rule out medical co-morbidities.	SHE Components Environment Sleep timing Bedtime routines Physiological Communication adaptations	Outcome measures ACT, CSHQ and FISH Q.O.L outcome measures- PSOC,Child behaviour checklist, repetitive behaviour scale, Parent proxy report of the paediatric quality of life inventory.. Parent satisfaction survey	Effectiveness Study concluded Improvements in sleep achieved regardless of mode of delivery. Combined treatment groups TCSHQ mean total pre-post 55.5- 49.3. P value< 0.001 . No confidence limits reported Parent experience

				Outcomes CSHQ,TCSHQ, sleep onset delay, night awakenings, sleep duration, bedtime resistance, sleep anxiety, parasomnias, sleep disordered breathing, daytime sleepiness. ACT Total sleep time, sleep latency, wake after sleep onset, sleep efficiency., PSOC, Efficacy, satisfaction	Group and individual delivery modes for sleep education equally adequate.
	education delivered individually and in group format to parents of ASD children with ASD.	Children randomised into individual - 1, 1hour session or group delivery - 2, 2 hour sessions arms.(no waiting list controls) Both groups had 2 follow up phone calls.	Also includes: Sleep theory Extinction techniques Bedtime pass		
33	Middlesborough Moving Forward (2011) Bespoke SHE tool			Sleep leaflet for parents includes graduated extinction. SHE Components Environment Sleep timing Bedtime routines Physiological Communication adaptation	
34	Montgomery et al. (2004) Randomised controlled trial . MCSHE intervention study	Sample- 66 LD children 2-8 years (42 boys and 24 girls) with settling or night waking problems. (66 received MCSHE) UK study carried out in home setting. Aim-to compare efficacy of x2 delivery methods of behavioural sleep intervention.- Face to face delivery versus booklet delivery versus waiting list control group.	Assessment elimination of medical co-morbidities, SHR sleep interview, 2 week sleep diary. Composite sleep disturbance score. Children randomised to - Brief treatment – giving parents a booklet.Conventional treatment – Booklet plus face to face instruction- x1 90 minute session. Crossover control group- no intervention for 6 weeks then randomised to a treatment group. Measures repeated at 6 weeks and 6 month follow up.	SHE Components Sleep timing Bedtime routines Behavioural management Physiological Also included Extinction techniques	Outcome measures Composite sleep disturbance scores used and parental evaluation responses.	Effectiveness Pre-post treatment Mean Composite sleep disturbance scores for brief treatment 6.18-2.55 conventional treatment 6.55- 1.89 W/list control 6.00- 5.75 P value reported for group (brief and conventional treatment) differences in Composite sleep disturbance scores when compared with waiting list controls and was found to be statistically significant p<0.001. But could not detect a significant difference between the 2 methods. No confidence limits reported
35	National Autistic Society (2008) Bespoke SHE tool for children with Autistic Spectrum Disorder		Assessment Recommends sleep diaries	11 page information sheet SHE Components Bedtime routines Environment Physiological Communication adaptations		
36	O'Connell and	Australian study	X3 parent education	SHE Components	Outcome measures	57% (N13) families reported positive

	Vanaan (2008) Pilot study MCSHE intervention study	Sample: 23 children with DD including ASD aged 10 months- 7 year 1 month. Report on the findings of the Sleep wise approach, a community based educational approach implemented over 1 year Sleep workshops with follow up home visits and telephone support	sleep workshops, delivered by 2 allied health workers. Supported by manual for professionals "*Sleepwise-positive sleep practices for children with DD*" Assessment GAS.Families received home visits and phone – calls in between sessions and throughout intervention for an average of 8 weeks before goals achieved.	Sleep timing Bedtime routines Behavioural management Communication adaptations Intervention also included Extinction Scheduled awakening Sleep restriction	GAS	outcomes (achievement of long term goals?) at 6-11month follow up. 26% (N6) families achieved short term goals. Families took an average of 8 weeks to realise short term sleep goals. No p values or confidence intervals reported. Experience She needs to be delivered with continued professional instruction and support, repeating advice over several months
37	Peak Interventions (2012) Bespoke SHE tool for children with autism			SHE Components Bedtime routines Behavioural management Environment Communication adaptations		
38	Petta et al. (2012) Conference proceedings Bespoke SHE tool for children with DD		Assessment Lists the CSHQ, FISH, sleep diaries, sleep interview (unspecified)	SHE Components Sleep timing Bedtime routines Behaviour management Physiological Communication adaptations Environment		
39	Piazza et al. (1997) Randomised controlled trial . SHE intervention study	Sample- 14 children with DD average age 7.8 years (4-14 years) (7 received SHE) US study. Carried out in in-patient unit. Aim: To compare the efficacy of a Faded bedtime with response cost with bedtime scheduling in children with mental retardation and sleep problems	Assessment Children were included if they slept for 90% or less than was expected for their chronological age. Sleep assessment/history not reported. 24 hour momentary time sampling procedure designed for the study, used to obtain baseline level of child's sleep.7 children randomised to each group No waiting list control group	SHE Components Sleep timing (Bedtime scheduling).Bedtime routines Other intervention group: Faded bedtime with response cost Theory *"By setting a regular sleep and wake time, the bedtime scheduling procedure might be hypothesized to maximise the likelihood that the individual's circadian rhythms would become entrained to the 24 hour cycle"* p418.	Outcomes: Hours of disturbed sleep	Effectiveness Mean Hours of disturbed sleep baseline- post treatment for; Bedtime scheduling- 1.37-1.10 hours mean change 0.27. Faded bedtime with response cost- 1.44-0.53 hours mean change 0.91 hours. P values reported for combined treatment – statistically significant at 0.026. Sample size is small. No confidence limits reported. Study concluded Faded bedtime with response cost was superior to bedtime scheduling in reducing mean hours of disturbed sleep.
40	Raising Children Australia (2012)			3 step sleep tips SHE components Sleep timing		

	Bespoke SHE tool for children with DD			Bedtime routines Behavioural management Communication adaptations		
41	Redwood (2012) Bespoke SHE tool for children with DD			SHE components Sleep timing Bedtime routines Environment Physiological Communication adaptations		
42	Reed at al (2009) Un-controlled Pre-post test study , single group design MCSHE intervention study	Sample:-20 children 3-10 years – 16 boys and 4 girls (20 received MCSHE). US based study, no setting specified. Aim:to test efficacy of a parent sleep education class in treating insomnia in children with ASD.	Group parent education workshops 3x2hour sessions, targeted and based on responses to CSHQ and FISH Assessment CSHQ, ACT, FISH (22 item), sleep diaries. Exclude medical co-morbidities	SHE components Bedtime routines Physiological Sleep timing Behaviour management Communication adaptations Also extinction, bedtime pass, sleep theory	Outcome measures CSHQ, FISH (22 item), ACT, sleep diaries. Q.O.L measures Parental concerns questionnaire, parenting stress index Outcomes CSHQ, TCSHQ, bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night awakenings, daytime sleepiness, sleep disordered breathing, parasomonias FISH All 22 outcomes included	Effectiveness TCSHQ Pre-post – 56.63 to 49.74 P 0.004. P<0.05 in x6 FISH outcomes. Incomplete data set reported for ACT scores Study concluded the intervention was effective in improving sleep problems. No confidence limits specified. .
43	Research Autsim (2007) Bespoke SHE tool for children with ASD			9 page SHE leaflet for parents includes graduated extinction SHE components Bedtime routines Sleep timing Behaviour management Physiological Communication adaptations Environment		
44	Scope (2012) Bespoke SHE tool for children with DD		Assessment Emphasises need to rule out medical causes and advises on sleep diary use	SHE components Bedtime routines Sleep timing Behaviour management Physiological Communication adaptations Environment		
45	Sense (2012) Bespoke SHE tool for children with DD		Assessment Advises on sleep diary use	SHE components Bedtime routines Sleep timing Behaviour management Physiological Communication adaptations Environment		
46	Sitnick et al.(2008)	US study carried out	Behavioural sleep	Sleep assessment	Outcomes	All variables significantly correlated p<0.01

	Sleep assessment study Study comparing ACT with video-somnography in preschool-aged ASD, DD and TD children, and focused on the accuracy of detection of nigh-time awakenings.	in children's own homes Sample size 58 (50% boys) ASD group (N =22) DD group (N=11) TD group (N25) Mean age 47 mths. Broad socio-demographic characteristics	problem definition- Prolonged sleep onset and night wakings. Children wore ACT devices for 7 nights and had home video recordings for 2 consecutive nights ACT monitors placed on non-dominant ankle, but 2 participants wore it on their wrist.	A Mini-Mitter® Actiwatch Actigraph (AW64, Mini-Mitter, Inc., Bend, Ore). AG6749P time-lapse video recording system (Osaka, Japan) with a Sanyo VDC 9212 low-level illumination camera (Osaka, Japan).Parents also asked to complete a 24 hour sleep diary for the 7 days	ACT Sleep-onset time, Sleep Latency, Total sleep time, Sleep end time, number of awakenings ,wake after sleep onset 3 diagnostic groups and placement groups collapsed as no significant differences in results.	and nocturnal awakenings p<0.05, however further investigations revealed this correlation was relative rather than absolute. In fact " only one third of the observed video awakenings were detected by the actigraph" No confidence limits reported. Concluded ACT has poor agreement for detecting night-wakings, compared with videosomnography
47	Sleep Practitioners (2012) Bespoke SHE tool for children with DD			SHE Components Bedtime routines Sleep timing Behavioural management Environment Physiological Communication adaptations		
48	Stores and Stores (2004) Randomised controlled trial MCSHE intervention study	Sample: 46 children with DS, 18 children TD (U group) (46 received MCSHE) Aged 7mths-4years, 48% boys. UK study Sleep assessment carried out in children's homes and group instruction in clinic setting. Aim: To assess the efficacy of group instruction for mothers of children with Down Syndrome(DS) in preventing or minimising sleep problems	Assessment: Participants characterised by having no other neurological, psychological or developmental problems. MSPSQ (full version), CSPS, ACT. DS children randomised into instruction group or control group (who did not receive the instruction until the end of the study).Results compared at 1 and 6 months after intervention. Then same intervention repeated to control group Baseline measures of sleep in DS group also compared with baseline sleep measurement taken from U group.Instruction session 90 mins, groups tailored to age range and mothers given "Encouraging good sleep habits in young children	SHE Components Bedtime routines Behavioural management Group instruction also includes : Sleep theory Specific advice for specific sleep problems	Outcome measures: MSPSQ (full version), Composite sleep problem score, ACT, sleep related breathing problem score Outcomes: Composite sleep problem scores Sleep related breathing problem score ACT Total sleep time, wake after sleep onset, mean duration of wake episodes, number of waking episodes, duration of longest waking episode, mean activity score, sleep efficiency, total minutes scored as sleep.	Effectiveness Instruction group showed statistically significant lower Composite sleep problem score score than control group at 6 month follow up.p<0.05, indicating some effectiveness of intervention. ACT results did not show any significant results following intervention.3x2 mixed ANOVAs -> p.0.05. Authors concluded the intervention showed some effectiveness in treating behavioural sleep problems. However, DS group was heterogeneous and children presented a mix of physical sleep disorders which would be unresponsive to behavioural interventions including SHE. No confidence limits reported Parent experience Parents reported consistency, reinforcing behaviours, instilling good sleep habits and bedtime routines was useful advice, also "The importance of giving confidence in their ability to achieve good sleep habits in their child"p67

			with down syndrome" illustrated booklet.		
49	<u>Stores and Wiggs (2001)</u> Book chapter Bespoke SHE tool for children with DD		<u>Assessment</u> Sleep history review of 24 hour sleep wake pattern and (SHR) sleep questionnaire and sleep diaries.	SHE components Sleep timing Bedtime routines Behavioural management Physiological Environment	
50	Teel (2012) Bespoke SHE tool for children with DD.		9 tips for a good night's sleep. <u>SHE components</u> Sleep timing Bedtime routines Physiological factors Behavioural management Environment		
51	<u>Weiskop et al.(2001)</u> Case study MCSHE intervention study	Australian study Evaluated the effectiveness of behavioural sleep intervention on a 5 year old boy with Autism.	The programme consisted of an interview, three weekly training sessions and a review session. <u>Assessment</u> Sleep interview (adapted from a broad children's problems assessment) (Murphy, Hudson, King & Remenyi, 1985), actual tool used unobtainable. Sleep diary, GAS,	Intervention included extinction and graduated extinction SHE components Bedtime routines Behavioural management Physiological Communication adaptations	<u>Outcome measures</u> GAS, sleep dairies Outcomes Sleep latency Number of nights child falls asleep in own bed Number of nights child stays asleep in own bed. Sleep latency Programme evaluation questionnaire	During baseline the child's sleep latency averaged 22.5 minutes (SD =11.83) By the 3 monthfollow-up, his sleep latency decreased (M = 11.64, SD = 4.89) By the 12 month follow-up, his sleep latency had increased again (M = 34.3, SD = 19.79). Showing some limitation in the intervention.100% achievement of GAS reported, but no P-values or confidence intervals. The positive changes in the child's sleep did not occur until extinction was implemented. Both parents approved of the techniques taught in the programme: on a scale of 1 to 5 where 1 was 'not at all' and 5 was 'very strongly', both parents rated their approval as 5.
52	<u>Weiskop et al.(2005)</u> Multiple baseline across participants design. MCSHE intervention study	Australian study Study conducted in clinic and home setting Sample:13 children , 6 with ASD 7 with fragile x . Age range 3-9years(13 received MCSHE) Aim: to evaluate a parent training programme to reduce sleep problems in children with Fragile X or Autism	Participants were grouped in 3 cells and 7 week intervention introduced at different time periods, during which other cells retained baseline conditions. Follow up at 3 and 12 months.X3 weekly parent training sessions delivered using modelling, role play , hand-outs and checklists.Combination of home visits and clinic sessions, plus telephone	SHE Components Sleep timing Bedtime routines Behavioural management Communication adaptations Parent training programme also includes : Partner support strategies Extinction	<u>Outcome measures</u> GAS Programme evaluation questionnaire Sleep diary <u>Outcomes</u> No of pre- sleep disturbances per week, no of nights fell asleep alone, sleep latency, night wakings, co slept nights, average duration of night-time sleep	<u>Effectiveness</u> Across all participants the mean GAS was 76.3% following intervention. Improvement was sustained to 12 month f/up. However, no p values or confidence intervals reported. Improvements did not occur until extinction was introduced. <u>Parent experience</u> Open ended questions responses to programme evaluation- Mothers liked the outcomes, support, phone calls and method of delivery.7 mothers disliked record keeping and 3 mothers thought it was very time consuming.

			support. <u>Assessment</u> Sleep interview (adapted from a broad children's problems assessment) (Murphy et al.,1985), actual tool used unobtainable, sleep dairy (with telephone support)			

Appendix 4 – Characteristics of Sleep Histories and Outcome Measures

	Sleep history tool/outcome measure	Dimensions description	Target group	Outcomes	Comments on theory
1.ASPS- Albany Sleep Problems Scale Durand (2013b) Christodulu and Durand (2004)	Sleep history tool	46 item questionnaire, 32 items 5 point Likert scale.12 yes/no and 2 open questions.	Children with special needs	None specified	Assessment designed to assist in identification of sleep problem type.
2. SHR Sleep History Review of 24hr sleep wake pattern Stores and Wiggs (2001)	Sleep history tool	5 parts– Evening, going to bed, night-time, waking, daytime. 30 open questions	Children with intellectual impairment	None specified	None specified
3. Cerebra Cerebra Sleep Service questionnaire Cerebra (2011)	Sleep history tool	44 item questionnaire 5 parts– medical, about your child, home environment, sleep problems, additional information	Children with brain related conditions	None specified	None specified
4.HP Handsel Project sleep interview schedule. Handsel Project (2012)	Sleep history tool	42 item questionnaire 7 parts– background, medical, environment, evenings, bedtimes, night-times, waking, daytime	Children with disabilities	None specified	None specified
5. SSMS Southampton Sleep Management Scale. Bartlet and Beaumont (1998)	Sleep history tool	73 item questionnaire Mix of qualitative and quantitative	Handicapped or chronically ill children	None specified	None specified
6. MSPSQ (Shortened version) Modified Simmonds & Parraga sleep questionnaire Johnson et al. (2012)	Sleep history tool and outcome measure	Shortened version 51 item, including 36 items amenable to Likert scoring. 15 additional qualitative questions based on sleep history to	Original SPSQ developed for TD children, and adapted for DD by Wiggs and Stores (1996a)- MSPSQ (Shortened version)	Total MSPSQ score based on 36 quantitative items range 36-180– Score over 71.24 indicates poor sleeper.	Assessment extracts quantitative and qualitative information to support sleep disorder diagnosis.

		help direct treatment	tested with children with ASD 2-16 years—Johnson et al. (2012)		
7.MSPSQ **(Full version)** Modified Simmonds & Parraga sleep questionnaire Stores and Stores (2004)	Sleep history tool and Outcome measure	Full 5 part version, 105 items. Part 4—45 item quantitative element as described in Maas et al.(2011). Remaining 4 parts reflect descriptive element.	Original SPSQ developed for TD children, and adapted for DD by Wiggs and Stores (1996a)- MSPSQ (Full version) used with children with Down Syndrome 6mths-5 years Stores and Stores (2004)	Total MSPSQ score based on 45 quantitative items. High total score reflects high frequency of sleep problems, but does not reflect severity	Items based on symptoms of sleep disorders classified in the AASM (2005)
8. MSPSQ **(Part 4)** Modified Simmonds & Parraga sleep questionnaire Maas et al.(2011)	Outcome measure	Part 4- 45 item , 7 point Likert scale assessing the frequency of sleep behaviours.	Original SPSQ developed for TD children, and adapted for DD by Wiggs & Stores (1996)- MSPSQ(Part 4) Tested with individuals with ID 15 months-66 years –Maas et al (2011)	Total MSPSQ score based on 42 quantitative items range 42-294 (3 items deleted in Maas et al (2011) study due to insufficient reliability). High total score reflects high frequency of sleep problems, but does not reflect severity.	Items based on symptoms of sleep disorders classified in the AASM (2005)
9. CSHQ(39 item) Child Sleep Habit Questionnaire Johnson et al. (2012)	Outcome measure	39 item version– 31 quantitative items 3 point Likert scale plus yes/no response to "Is this a problem?" plus 8 unspecified open-ended questions. Scores grouped into 8 subscales.	Originally developed for TD children.4-10 years. Tested with ASD children 2-16 years—Johnson et al. (2012)	Total sleep CSHQ score -Range 35-73, over 41 indicates poor sleeper. Plus 8 subscale scores; Bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night awakenings, parasomnias, daytime sleepiness, sleep disordered breathing.	Items based on symptoms of sleep disorders classified in the AASM (2005)
10.CSHQ (52 item) Child Sleep Habit Questionnaire Goodlin-Jones et al.(2008)	Outcome measure	52 item version– 3 point Likert scale. Scores grouped into 8 subscales	Originally developed for TD children.4-10 years and tested with 2-5 1/2 year old children with ID– Goodlin-Jones et al.	Total sleep CSHQ Score—range unspecified. Over 41 indicates poor sleeper	Items based on symptoms of sleep disorders classified in the AASM (2005)

				Plus 8 subscale scores; Bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night awakenings, parasomnias, daytime sleepiness, sleep disordered breathing. Insomnia subscale also used in some studies.	
Beresford et al.(2012) Chapters 14,15, 16 Adkins, Molloy et al. (2012)			(2008)		
11. CSHQ (33 item) Child Sleep Habit Questionnaire Cortesi et al.(2012) Malow et al.(2013) Reed et al.(2009)	Outcome measure	33 item version– 3 point Likert scale. Scores grouped into 8 subscales	Originally developed for TD children.4-10 years and used with children ASD aged 4-10 years– Cortesi et al (2012)	Total sleep CSHQ score Plus 8 subscale scores; Bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night wakings, parasomnias, daytime sleepiness, sleep disordered breathing.	Items based on symptoms of sleep disorders classified in the AASM (2005)
12. FISH Family Inventory of Sleep habits. (12 item) Malow et al.(2009). Malow et al.(2013)	Outcome measure	12 item. 5 point likert scale of sleep habits. Includes bedtime routines, sleep environment and parental interactions	Children with ASD age 3-10 years	Total FISH score— higher score indicates better sleep hygiene. Range 12-60	Poor sleep hygiene known to contribute to insomnia and children with ASD susceptible. Items generated to identify daytime behaviours known to affect sleep; responsive to intervention.
13 FISH Family Inventory of Sleep habits. (22 item) Reed et al.(2009)	Outcome measure	22 item 5 point likert scale of sleep habits. Includes bedtime routines, sleep environment and parental interactions	Children with ASD age 3-10 years	Total FISH score— higher score indicates better sleep hygiene. Range 12-60	Poor sleep hygiene known to contribute to insomnia and children with ASD susceptible. Items generated to identify daytime behaviours known to affect sleep; responsive to intervention.
14. CPQ Cerebral Palsy	Outcome measure	2 parts-	Children with CP age 2-	Sleep latency, Night	No information on what

questionnaire Khan and Underhill (2006)		*Sleep section*: Bedtime routines, Night time behaviour, Breathing quality during sleep. *Medical section*: Identified medical conditions. Items making up these sections not reported.	10 years	time behaviours, Breathing quality. Total section scores grouped into low, medium or high categories.	or how items selected, methodology or theoretical background.

Appendix 5 – SHE component content analysis excerpt.

SHE Components	Teel (2012)	Adkins, Goldman et al. (2012)	Adkins, Molloy et al. (2012)	Allison et al. (1993)	Lester (2012)	Autism Society (2012)	Bartlet and Beaumont (1998)	Beresford et al. (2012)	Bramble (1996)	Bramble (1997)	Capability Scotland (2012)	Catherall and Williams Jones (2011)	Cerebra (2011)	Christodulu and Durand (2004)	Chronic Care for Sleep (2012)	Contact a Family (2007)	Cortesi et al.(2012)	Disabled Living Foundation (2012)	Down's Syndrome Association (2008)	Durand (2011)	Durand (2013a)	Durand (2013b)	Early Support (2010)	Goodlin–Jones et al (2008)	Handsel Project (2012)	Hewitt (1985)	Johnson et al.(1995)	Johnson et al.(2012)	Khan an Underhill (2006)	Leeds Community Healthcare (2010)	Maas et al.(2011)	Malow et al.(2009)	Malow et al.(2013)	MiddlesbrougMoving forward (2011)	Montgomery at al.(2004)	National Autistic Society (2008)	O'Connell and Vanaan (2008)	Peak Interventions (2012)	Petta et al (2012)	Piazza et al (1997)	Raising Children Australia (2012)	Redwood (2012)	Reed et al (2009)	Research Autism (2007)	Scope (2012)	Sense (2012)	Sitnick et al (2008)	Sleep Practitioners (2012)	Stores and Stores (2004)	weiskop et al (2001)	Weiskop et al (2005)	Wiggs and Stores (2001)
1.Sleep timing																																																					
1:1. Consistent bedtimes and wake times (35)	x		x			x			x	x	x	x	x	x	x	x	x	x	x	x	x	x		x		x	x			x		x	x	x	x				x	x		x	x	x	x	x		x		x	x	x
1:2. Age appropriate bedtimes(6)															x				x															x								x			x	x						
1:3. Age appropriate daytime napping(12)			x													x	x		x							x	x							x	x			x						x	x							x
1:4. Avoid late afternoon age appropriate napping (9)											x		x		x		x	x						x																					x	x	x					
1.5 Going to bed only when tired (1)																																																				x

Appendix 6 Ethical approvals

Appendix 6.1.- School of healthcare sciences 5.3.14 (exploratory study)

Appendix 6.2. Wales research ethics committee 5. 23.4.14 (exploratory study)

Appendix 6.3. Research and development internal review panel. 12.5.14 (exploratory study)

Appendix 6.4. School of healthcare sciences 10.3.15 (co-design study)

Appendix 6.5. Sponsor permission letter 9.7.15 (co-design study)

Appendix 6.6. BCUHB response letter 12.6.15 (co-design study)

Healthcare and Medical Sciences Academic Ethics Committee

5th March 2014

Julie Sutton
School of Healthcare Sciences
Bangor University
Fron Heulog
Ffriddoedd Road
Bangor
LL57 2EF

Dear Ms Sutton

Re: 2014-01-06 / Sleep Hygiene Education and Children with Developmental Disabilities

Thank you for your application to the AEC which was subject to an expedited review as requested. All of the necessary documentation was provided and appropriately completed.

I am therefore able to give approval for your study on behalf of the AEC, and this letter constitutes evidence of that approval should it be necessary for any applications to other RECs.

Please note that approval from this AEC does not convey automatic authority to proceed with your study. You are formally advised that it is essential to confirm with the relevant authorities whether you are required to submit your proposal to any other Ethics Committee(s), such as Local NHS Research Ethics Committee, and NHS Research Governance Departments, prior to commencing your study.

Should you need to make any substantial amendments to your study protocol during the lifetime of the research, you are required to submit notice of these to the AEC for further approval, including major amendments requested by an external REC or R&D Committee.

If you have any queries, please do not hesitate to contact myself or Dr Joyce Wilkinson, Vice Chair, for clarification.

Yours sincerely

Dr Siôn Williams
Chair, HCMS AEC

PRIFYSGOL BANGOR   BANGOR UNIVERSITY
FRON HEULOG   FRON HEULOG
FFORDD FFRIDDOEDD   FFRIDDOEDD ROAD
BANGOR, GWYNEDD   BANGOR, GWYNEDD
LL57 2EF, UK   LL57 2EF, UK

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FFACS: 01248 383175   FAX: 01248 383175

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Registered charity number: 1141565

HCMS AEC approval letter V2 July 2013

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Gwasanaeth Moeseg Ymchwil | **RES** | Research Ethics Service

Pwyllgor Moeseg Ymchwil Cymru 5
Wales Research Ethics Committee 5
Bangor

Clinical Academic Office
Ysbyty Gwynedd Hospital
Betsi Cadwaladr University Health Board
Bangor, Gwynedd
LL57 2PW

Telephone/ Facsimile: 01248 - 384.877
Email: Rossela.Roberts@wales.nhs.uk
Website : www.nres.nhs.uk

Mrs Julie Sutton
Centre for Health Related Research
Bangor University,
Fron Heulog, Ffriddoedd Rd,
Bangor
LL57 2EF   j.sutton@bangor.ac.uk

23 April 2014

Dear Mrs Sutton,

Study title: Developing the evidence base for sleep hygiene education (SHE)
                     in treating behavioural sleep problems in primary school aged
                     (3-10 years) children with developmental disabilities (DD)
REC reference: 14/WA/0138
IRAS project ID: 153545

The Research Ethics Committee reviewed the above application at the meeting held on
17 April 2014. Thank you for attending to discuss the application.

We plan to publish your research summary wording for the above study on the HRA website,
together with your contact details, unless you expressly withhold permission to do so.
Publication will be no earlier than three months from the date of this favourable opinion
letter. Should you wish to provide a substitute contact point, require further information, or
wish to withhold permission to publish, please contact the REC Manager Dr Rossela
Roberts, rossela.roberts@wales.nhs.uk.

Ethical opinion

**Ethical issues raised by the Committee in private discussion, together with responses
given by you when invited to join the meeting**

The following issues were discussed:

Recruitment arrangements and access to health information: fair participant selection
The Committee was satisfied that the selection of eligible participants has taken into account
the their relationship with the children and sufficient details are provided in the protocol
regarding the inclusion and exclusion criteria.
A clarification was requested regarding the recruitment method.
Mrs Sutton clarified that the research team aim to recruit professionals (from the NHS and
third sector) experienced in the delivery of the Sleep Hygiene Education intervention and
parents who are using Sleep Hygiene Education in children with developmental disabilities.
The Committee raised no further issues in relation to the recruitment arrangements.

GIG | Bwrdd Iechyd Addysgu Powys
NHS WALES | Powys Teaching Health Board

Cynhelir Cydweithrediad Gwyddor Iechyd Academaidd y Sefydliad Cenedlaethol ar
gyfer Ymchwil Gofal Cymdeithasol ac Iechyd gan Fwrdd Addysgu Iechyd Powys

The National Institute for Social Care and Health Research Academic Health Science
Collaboration is hosted by Powys Teaching Health Board
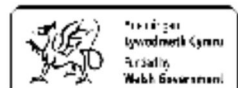
Llywodraeth Cymru
Welsh Government

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Panel Arolygu Mewnol Y&D
R&D Internal Review Panel

Betsi Cadwaladr University Health Board
Ysbyty Gwynedd
Clinical Academic Office
Bangor, Gwynedd
LL57 2PW

Bwrdd Iechyd Prifysgol
Betsi Cadwaladr
University Health Board

Mrs Julie Sutton
Centre for Health Related Research
Bangor University
Fron Heulog
Bangor
LL57 2EF   j.sutton@wales.nhs.uk

Chairman/Cadeirydd – Dr Nefyn Williams PhD, FRCGP
Email: rossela.roberts@wales.nhs.uk
wendy.scrase2@wales.nhs.uk
sion.lewis@wales.nhs.uk
Tel/Fax: 01248 384 877

12 May 2014

Dear Mrs Sutton,

Re: Confirmation that R&D governance checks are complete / R&D approval granted

Study Title	Developing the evidence base for sleep hygiene education (SHE) in treating behavioural sleep problems in primary school aged (3-10 years) children with developmental disabilities
IRAS reference	153545
REC reference	14/WA/0138

Thank you for submitting your R&D application and supporting documents.
The above study was eligible for Proportionate Review and was reviewed by the R&D Manager and Chairman of the Internal Review Panel.

The Committee is satisfied with the scientific validity of the project, the risk assessment, the review of the NHS cost and resource implications and all other research management issues pertaining to the revised application.

The Proportionate Review Committee is pleased to confirm that all governance checks are now complete and to grant approval to proceed at Betsi Cadwaladr University Health Board sites as described in the application.

The documents reviewed and approved are listed below:

Document:	Version	Date
R&D Form	-	02/04/2014
SSI Checklist	-	-
Non-NHS SSI Form	-	03/04/2014
Protocol	1	06/12/2013
Information Sheet (INVS) Parents	1	31/03/2014
Information Sheet (FGs) Parents	1	31/03/2014
Parent Consent Form	1	06/12/2013
Parent Contact Form	1	06/12/2013
Professionals Consent Form	1	06/12/2013
Professionals Contact Form	1	06/12/2013
Parent Interview Discussion Areas	1	06/12/2013
Preparation Sheet for Parent Focus Groups	-	-
Focus Group Discussion Areas	1	06/12/2013
Feasibility Questions	1	06/12/2013
Sleep Hygiene Education Pathway	1	06/12/2013
Insurance Certificate	-	-
Healthcare & Medical Sciences Academic Ethics Committee	-	05/03/2014
CV of PI (J Sutton)	-	02/01/2014

All research conducted at the Betsi Cadwaladr University Health Board sites must comply with the Research Governance Framework for Health and Social Care in Wales (2009).

An electronic link to this document is provided on the BCUHB R&D WebPages.
Alternatively, you may obtain a paper copy of this document via the R&D Office.

Attached you will find a set of approval conditions outlining your responsibilities during the course of this research. Failure to comply with the approval conditions will result in the withdrawal of the approval to conduct this research in the Betsi Cadwaladr University Health Board.

If your study is adopted onto the NISCHR Clinical Research Portfolio (CRP), it will be a condition of this NHS research permission, that the Chief Investigator will be required to regularly upload recruitment data onto the portfolio database.

To apply for adoption onto the NISCHR CRP, please go to: http://www.wales.nhs.uk/sites3/page.cfm?orgid=580&pid=31979.

Once adopted, NISCHR CRP studies may be eligible for additional support through

the NISCHR Clinical Research Centre. Further information can be found

at:http://www.wales.nhs.uk/sites3/page.cfm?orgid=580&pid=28571 and/or from your

NHS R&D office colleagues.

To upload recruitment data, please follow this link: http://www.crncc.nihr.ac.uk/about_us/processes/portfolio/p_recruitment.

Uploading recruitment data will enable NISCHR to monitor research activity within NHS organizations, leading to NHS R&D allocations which are activity driven.

Uploading of recruitment data will be monitored by your colleagues in the R&D office.

If you need any support in uploading this data, please contact wendy.scrase2@wales.nhs.uk or sion.lewis@wales.nhs.uk

If you would like further information on any other points covered by this letter please do not hesitate to contact me.

On behalf of the Committee, may I take this opportunity to wish you every success with your research.

Yours sincerely,

Dr Nefyn Williams PhD, FRCGP
Associate Director of R&D
Chairman Internal Review Panel

Healthcare and Medical Sciences Academic Ethics Committee

Our ref: SW/SM

10 March 2015

Julie Sutton
c/o School Healthcare Science
Fron Heulog
Bangor
Gwynedd

Dear Julie

Re: Proposal 2014-12-03 Sleep Hygiene Education and Children with Developmental Disabilities

Thank you for your application to the AEC which was subject to an expedited review as requested. All of the necessary documentation was provided and appropriately completed.

I am therefore able to give approval for your study on behalf of the AEC, and this letter constitutes evidence of that approval should it be necessary for any applications to other RECs.

Please note that approval from this AEC does not convey automatic authority to proceed with your study. You are formally advised that it is essential to confirm with the relevant authorities whether you are required to submit your proposal to any other Ethics Committee(s), such as Local NHS Research Ethics Committee, and NHS Research Governance Departments, prior to commencing your study.

Should you need to make any substantial amendments to your study protocol during the lifetime of the research, you are required to submit notice of these to the AEC for further approval, including major amendments requested by an external REC or R&D Commit

If you have any queries, please do not hesitate to contact me for clarification.

Yours sincerely

Dr Sion Williams
Chair, HCMS AEC

c.c. Dr Jaci Huws, Supervisor

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PRIFYSGOL
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UNIVERSITY

9th July 2015

Julie Sutton
School of Healthcare Sciences
Bangor University
Fron Heulog
Ffriddoedd Road
Bangor
LL57 2EF

Dear Julie

Re: Developing Sleep Hygiene Education (SHE) as a Complex Intervention for Behavioural Sleep Problems in Children with Developmental Disabilities (DD).

Further to your e-mail of 8th May 2015 confirming that the HRA have decided that, as a service evaluation, you do not need REC approval to continue with the study. I write to confirm that, as the project is not recruiting parents through the NHS and is being carried out in non-NHS premises, there is no need to seek R&D approval prior to commencing the study.

Please do not hesitate to contact me should you require anything further.

Yours sincerely

Jo Rycroft-Malone
Head of the School of Healthcare Sciences

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Betsi Cadwaladr
University Health Board

**Canolfan Blant Sir Ddinbych Hyfrydle
Lawnt DINBYCH Sir Ddinbych LL16 4ST**

**Denbighshire Children's Centre Hyfrydle
Lawnt DENBIGH Denbighshire LL16 4ST**

Julie Sutton RNLD
PhD Student & RCBC Fellow at;
Bangor University
School of Healthcare Sciences
Fron Heulog
Ffriddoedd Rd
Bangor LL57 2EF

☎ **07825244752**

Gofynnwch am / Ask for: Dr Chris Catherall
E-bost / Email: chris.catherall@wales.nhs.uk
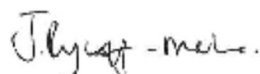**Dyddiad / Date:** 12th June 2015

Annwyl/Dear Julie

Re: **Developing Sleep Hygiene Education (SHE) as a Complex Intervention for
Behavioural Sleep Problems in Children with Developmental Disabilities (DD).**

I was pleased to meet with you yesterday to discuss the reasons why we were unable
to support your application.

I briefly outlined the work stream's aims and where we were up to in terms of service
developments; this discussion focused specifically on the childhood disability
population but gave an account of the broader aims of providing a pan North Wales
'sleep service' to children irrespective of their developmental needs.

I then outlined the primary reasons why we were unable to support your application, as
follows:

1. We were of the opinion that the proposal may overlap significantly with core
   service provision that we have/and are continuing to establish across North
   Wales. As a consequence this may complicate and create potential confusion
   for families; particularly when you consider the investments made in developing
   a formal intervention strategy for this issue.

2. Through commissioning, the child learning disability service has provided 'Sleep
   Scotland' training to a large cohort of BCUHB staff. The training and the
   associated course materials have a strong emphasis on SHE. Indeed, the sleep
   history assessment is a tool that focuses on key sleep hygiene factors that
   inform intervention. With reference to your HRA V(4) ethics summary we are
   concerned that developing 'a complex sleep hygiene intervention' is not
   absolutely necessary given the above developments.

3. The recently conducted Foundation of Nursing project (Catherall, Davies and
   Brewerton, 2015), mirrors some of your research aims. Here this study
   consulted with parents (parent stories) to understand sleep related problems. It

Cyfeiriad Gohebiaeth ar gyfer y Cadeirydd a'r Prif Weithredwr / Correspondence address for Chairman and Chief Executive:
Swyddfa'r Gweithredwyr / Executives' Office,
Ysbyty Gwynedd, Penrhosgarnedd
Bangor, Gwynedd LL57 2PW   **Gwefan:** www.pbc.cymru.nhs.uk / **Web:** www.bcu.wales.nhs.uk

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employed a Claims, Concerns and Issues exercise with relevant nursing staff, which in tandem with the Sleep Scotland training informed and determined service direction. We are of the opinion that elements of your research aims duplicate work already undertaken within BCUHB.

It is important to reiterate here that the decision not to support your application does not relate to the projects methodology but was due to concerns raised by the service management team, as outlined above.

I would like to wish you all the very best with your research and subsequent write up.

Yn gywir /Yours sincerely

Dr Chris Catherall
Clinical Nurse Specialist – Sleep/Behaviour
Continuing Care Team

c.c.   Trevor Smith, Clinical Audit and Effectiveness Manager
       Yvonne Harding, ACOS, C&YP CPG

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Appendix 7 participant information sheets and invitation letters.

Appendix 7.1. Parent information sheet (exploratory study)

Appendix 7.2. Professional information sheet (exploratory study)

Appendix 7.3. Parent information sheet (co-design study)

Appendix 7.4. Professional information sheet (co-design study)

Appendix 7.5. Parent invitation letter (co-design study)

Appendix 7.6. Professional invitation letter (co-design study)

The Sleep Easy Project

We are looking at ways to improve the advice given

to families of children with developmental

disabilities who have sleep problems.

Information Sheet for Parents/Carers

Why have I been contacted?

You have been contacted because you have received sleep advice for a child/young person with sleep problems in your family.

Who are we?

We are a team of researchers interested in health issues affecting children, based at Bangor University. We are Dr Jacqueline Huws, Dr Christopher Burton, and Julie Sutton. We are funded by a Wales research organisation supporting research into child health.

How can you help us?

We are keen to hear your views and thoughts on developing a sleep advice package for children with developmental disabilities.

You can help by telling us about;

- Your experience of sleep advice and your child/young person
- Important things to include in a sleep advice package

Why does sleep advice for children with developmental disabilities need to be improved?

Childhood sleep problems affect up to 80% of families with children with disabilities and parents have said they don't always get the right support.

They want effective support to help improve their child's sleep.

What does taking part involve?

We would like you to take part in a one to one interview by meeting with Julie Sutton who is a member of the research team. Her photo is on the back of this leaflet.

The interviews will be conducted in English. If you prefer, Welsh medium interviews will also be available.

We will listen to your views and experiences of sleep advice and record what you say.

The recording will then be typed.
Unless you say so – your name will not be written down in reports and your identity will be kept anonymous.

Anything you say to the researcher will not be shared with people from outside the research team. However if safety concerns for a child or vulnerable young person are disclosed or apparent to the researcher such as signs of abuse, this will be taken seriously and a referral made to the local safeguarding children team.

What will happen to the information I provide?

We will use the information to design a sleep advice package tailored for children with developmental disabilities to help them sleep.

What will happen to the results of the overall Sleep Easy Project?

A final study report will be produced in 2015.

Findings will then be shared through publication

Do I have to take part?

No, it is entirely up to you if you want to take part. If you decide

to take part, you are still free to withdraw at any time

If you would like to take part in the project or would just like more

information, there is a contact form and stamp addressed envelope

enclosed with this leaflet. Please respond as soon as possible if you

would like to take part. Thank You.

Julie Sutton, PHD student

Centre for Health Related Research

School of Healthcare Sciences

Bangor University

Fron Heulog, Ffriddeodd Rd

Bangor, Gwynedd, LL57 2EF

Tel: 01248 383425/07983 245892 Email – j.sutton@bangor.ac.uk

If you have any concerns or queries which cannot be

resolved by the research team please contact **Professor Jo**

Rycroft-Malone Head of School of Healthcare Sciences,

Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd, LL57

2EF. **E-mail: j.rycroft-malone@bangor.ac.uk Tel:**

01248 383119

The Sleep Easy Project: Sleep Hygiene Education and Children with Developmental disabilities.

We are looking at how we can develop a manualised sleep hygiene education tool for children with developmental disabilities, experiencing behavioural sleep problems.

We would like to speak to professionals for their input in developing this.

Who are we?

We are a team of researchers interested in health issues affecting children, based at Bangor University. We are Dr Jacqeline Huws, Dr Christopher Burton and Julie Sutton. We are funded by a Wales research organisation supporting research into child health.

Why have I been contacted?

You have been contacted as your work involves supporting families of children with developmental disabilities with sleep problems.

What do we want to hear about?

We are keen to hear your views and thoughts on developing a sleep hygiene education package for children with developmental disabilities.

You can help by telling us about;

- Your experience of working with sleep problems and children with developmental disabilities
- How you think sleep hygiene education should be delivered and families supported to implement it
- Important things to include in a sleep hygiene education package for children with developmental disabilities

Why does sleep hygiene education for children with developmental disabilities need to be improved?

Childhood sleep problems affect up to 80% of these families and parents have said they don't always get the right support.
Sleep hygiene education is a first-line treatment for behavioural sleep problems, however there are no evidenced based sleep hygiene education tools available developed specifically for children with developmental disabilities.

What does taking part involve?

You can take part by meeting with Julie Sutton who is a member of the research team.

She will lead two small focus groups lasting no more than 90 minutes, at a mutually convenient time with practitioners supporting families with sleep problems. The focus groups will be conducted in English. If you prefer, Welsh medium one to one interviews will also be available. The focus group will be digitally recorded and video filmed and transcribed.
Unless you say so –your name will not be written down in reports and others will not be able to match your name with your words.
Anything you say to the researcher will not be shared with people from outside the research team. However if safety concerns for a child or vulnerable young person are disclosed or apparent to the researcher such as signs of abuse, this will be taken seriously and a referral made to the local safeguarding children team.

Participation is entirely voluntary and you are free to withdraw from a focus group or interview at any time without giving a reason.

What will happen to the information I provide?

We will use the information to design a sleep advice package tailored for children with developmental disabilities to help them sleep.

What will happen to the results of the overall Sleep Easy Project?

A final study report will be produced in 2015.

Findings will then be shared through publication

- in professional and academic journals
- seminar and conference presentations

If you would like to take part in the project or would just like more information, there is a contact form and stamp addressed envelope enclosed with this leaflet. Please return to Julie Sutton as soon as possible. Thank you.

Julie Sutton, PHD student
Centre for Health Related Research
School of Healthcare Sciences
Bangor University
Fron Heulog, Ffriddeodd Rd
Bangor, Gwynedd, LL57 2EF
Tel: 01248 383425/07983 245892 Email – j.sutton@bangor.ac.uk

Tel: 01248 383425, 07983 245 892
Email: j.sutton@bangor.ac.uk

If you have any concerns or queries which cannot be resolved by the research team please contact **Professor Jo Rycroft-Malone** Head of School of Healthcare Sciences, Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd, LL57 2EF. **E-mail: j.rycroft-malone@bangor.ac.uk Tel:** 01248 383119

PARENT INFORMATION SHEET - THE SLEEP EASY PROJECT.

You are being invited to take part in a Bangor University project. Please take time to read the following information about why this work is being carried out, before deciding if you want to participate. Should you like to discuss anything further, please contact the study researcher: Julie Sutton whose details are provided at the end of this information sheet.

Why is this project being carried out?

The project aims to provide the opportunity for approximately 8 parents and sleep professionals (Health Visitors, Community Nurses, Paediatricians and sleep counsellors) to work together to improve local sleep services for children with developmental disabilities.

Who is carrying out and funding the project?

This work is being conducted by a team of researchers at Bangor University. We are Dr Jacqueline Huws, Dr Chris Burton and Julie Sutton. We are funded by a Wales research organisation supporting research into child health.

Why have I been asked to get involved?

In this project we are focusing on parents of children with developmental disabilities who have received sleep advice from professionals to help with their child's sleep problem. As a parent who has been in contact with local sleep services you are ideally placed to inform us how to improve parents' experiences of professional sleep support.

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Do I have to take part?

It is your decision if you would like to participate in this project. Please read this information sheet before deciding. If you decide to take part, you are still free to withdraw at any time, without giving a reason. Should you wish to discontinue, any information you have already given will only be used in the study with your consent.

What does taking part involve?

There are three events in this project which you will be invited to attend (see below).

Parent feedback event
Meet other parents

Joint parent-staff event
Parents meet with sleep professionals

Co-design event
Parents and professionals work together to decide on service improvements

Parent feedback event.

You will be invited to take part in a meeting with other parents of children with developmental disabilities who have accessed local sleep services. You are welcome to bring a relative or friend along with you if you prefer. There will be an opportunity to listen to an audio podcast that has been compiled from earlier 'Sleep Easy Project' parent interviews which you may have participated in. At this meeting you will be asked to share how you think sleep services could be improved for parents of children with developmental disabilities. This event will take around three hours to ensure parents have ample time to discuss issues with each other and share views.

Joint parent-staff event.

The next meeting will involve both parents and local sleep professionals (health visitors, community nurses, paediatricians and sleep counsellors). This will begin with the audio podcast and parents and professionals will then have the opportunity to share their experiences. The purpose is for parents and professionals to start working alongside each other to develop improved experiences for parents accessing sleep services. This event will take around three hours.

Co-design event.

The final meeting will include parents and professionals again and will involve them working in partnership to design actions for sleep service improvements which are realistic and achievable. This event will take around three hours.

All events will be facilitated by the researcher Julie Sutton and another research officer from Bangor University or Wales research organisation. Lunch and refreshments will be provided and travelling expenses to and from each meeting will be paid for. The date and time of each event will be organised according to the availability of participants and you will be given as much notice as possible.

What are the potential disadvantages of participating?

Taking part in the project will mean you have to think about your experiences of professional sleep advice and support, which may raise some concerns for you. If you feel you would like to discuss these further outside of the meeting with the researcher, there will be an opportunity to do so.

What are the potential advantages of participating?

We intend that the findings of the 'Sleep Easy project' will improve local sleep services for children with developmental disabilities. It is acknowledged that this may not directly benefit you, but your contribution will help shape services in the future.

What will happen to the information I provide?

The events will be digitally recorded and video filmed. The information will be typed and analysed for use in a final report which will be available in 2016. Findings of the study will also be shared through publications in professional and academic journals and in conference presentations. Your name will not be written down in reports and your identity will be kept anonymous. If you disclose any information to the research team which indicates safety concerns for a child or vulnerable young person such as signs of abuse, this will be taken seriously and a referral made to the local safeguarding children team.

What if there are any problems?

You are not likely to experience any harm as a result of taking part in this study however if you have any cause for concern please contact Professor Jo Ryecroft-Malone Head of School of Healthcare Sciences whose contact details are at the end of this sheet.

Who has reviewed the study?

Ethical approval for this project has been granted by Bangor University, School of Healthcare Sciences Ethics Committee.

If you have any questions Julie Sutton can be contacted as follows:

Julie Sutton, PHD student

Centre for Health Related Research

School of Healthcare Sciences

Bangor University

Fron Heulog, Ffriddeodd Rd

Bangor, Gwynedd, LL57 2EF

Tel: 01248 383425/07983 245892 Email – j.sutton@bangor.ac.uk

If you have any concerns or queries which cannot be resolved by the research team please contact **Professor Jo Rycroft-Malone** Head of School of Healthcare Sciences, Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd, LL57 2EF. **E-mail: j.rycroft-malone@bangor.ac.uk Tel:** 01248 383119

PROFESSIONAL INFORMATION SHEET- THE SLEEP EASY PROJECT.

You are being invited to take part in a Bangor University research project. Please take time to read the following information about why this work is being carried out, before deciding if you want to participate. Should you like to discuss anything further, please contact the study researcher: Julie Sutton whose details are provided at the end of this information sheet.

Why is this project being carried out?

The project aims to enhance the experiences of parents of children with developmental disabilities accessing professional sleep support and advice. It also seeks to improve the experiences of professionals providing this service. We are using an adapted version of Experience-Based Co-Design EBCD which has been used extensively in health services in the UK and Australia. The method provides the opportunity for parents and professionals to work together to design improvements in sleep service provision for families of children with developmental disabilities.

Who is carrying out and funding the project?

This study is being conducted by a team of researchers at Bangor University. We are Dr Jacqueline Huws, Dr Chris Burton and Julie Sutton. We are funded by a Wales research organisation supporting research into child health.

Why have I been asked to get involved?

In this work we are focusing on parents of children with developmental disabilities who have received sleep support from professionals to help with their child's sleep

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problem. As a professional who provides sleep advice you are ideally placed to inform us how to improve experiences of care for professionals and parents who use sleep services.

Do I have to take part?

It is your decision if you would like to participate in this project. Please read this information sheet before deciding. If you decide to take part, you are still free to withdraw at any time, without giving a reason. Should you wish to discontinue, any information you have already given will only be used in the study with your consent.

What does taking part involve?

There are two events in this project which you will be invited to attend (see below).

> **Joint parent-staff event**
> *Parents meet with sleep professionals*

> **Co-design event**
> *Parents and professionals work together to decide on service improvements*

Joint parent-staff event.

The joint-parent-staff event follows a parent feedback event which parents of children with developmental disabilities have attended to share their experiences of sleep services. The joint parent-staff event brings together parents and sleep professionals and aims to facilitate partnership working to design improvements to sleep service delivery. It commences with the opportunity to listen to an audio podcast compiled from parent interviews carried out earlier on in the 'Sleep Easy project'. Views captured from earlier sleep professionals' focus groups (which you may have participated in) will also be shared with the group and parents and

professionals will then be asked to discuss priorities for service improvement. This event will take around three hours.

Co-design event.

The final meeting will include parents and professionals again and will involve them working together to design actions for sleep service improvements which are realistic and achievable. This event will take around three hours.

All events will be facilitated by the researcher Julie Sutton and another research officer from Bangor University or Wales research organisation. Lunch and refreshments will be provided. The date and time of each event will be organised according to the availability of participants and you will be given as much notice as possible. We are aware participation in these events will take up some of your work time and we appreciate your support. We also hope you understand the necessity of your input to improving experiences of professionals and parents in the future.

What are the potential advantages of participating?

We intend that the findings of the 'Sleep Easy project' will improve local sleep services for children with developmental disabilities. It is acknowledged that this may not directly benefit you, but your contribution will help shape services in the future.

What will happen to the information I provide?

The events will be digitally recorded and video filmed. The information will be transcribed and analysed for use in a final report which will be available in 2016. Findings of the project will also be shared through publications in professional and academic journals and in conference presentations. Your name will not be written

down in reports and your identity will be kept anonymous. If you disclose any information to the research team which indicates safety concerns for a child or vulnerable young person such as signs of abuse, this will be taken seriously and a referral made to the local safeguarding children team.

What if there are any problems?

You are not likely to experience any harm as a result of taking part in this study however if you have any cause for concern please contact Professor Jo Ryecroft-Malone Head of School of Healthcare Sciences whose contact details are at the end of this sheet.

Who has reviewed the study?

Ethical approval for this project has been granted by Bangor University, School of Healthcare Sciences Ethics Committee.

If you have any questions Julie Sutton can be contacted as follows:

Julie Sutton PhD Student
Centred for Health Related Research
School of Healthcare Sciences, Bangor University
Friddeodd Rd, LL57 2EF.
Tel: 01248 383425 / 07983245892. Email, j.sutton@bangor.ac.uk

If you have any concerns or queries which cannot be resolved by the research team please contact **Professor Jo Rycroft-Malone** Head of School of Healthcare Sciences, Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd, LL57 2EF. **E-mail: j.rycroft-malone@bangor.ac.uk Tel:** 01248 383119

Dear PARENT'S NAME

I am writing to invite you and one relative, friend or carer to come along and take part in three workshops that aim to involve parents in improving local sleep services for children with developmental disabilities. These follow on from a set of parent interviews which were conducted recently as part of the 'Sleep Easy project', which you may have participated in.

The first is a **parent feedback event** where you will be able to meet with other parents who have been interviewed about their experiences of professional sleep advice and support. Together you will be invited to listen to an audio podcast that has been compiled from the parent interviews and asked to share your views.

The **joint parent-staff event** follows on from the parent feedback event and aims to share parents' experiences with sleep professionals (health visitors, community nurses, paediatricians). You will have the opportunity to, jointly with local sleep professionals identify shared priorities for improving the sleep services provided to children with developmental disabilities.

The third event; a **co-design workshop** will involve both parents and professionals again but will be more focused towards developing ideas into concrete actions for sleep service improvements. Further information on these events and the 'Sleep Easy project' can be found in the attached parent information sheet.

At each event we will be serving refreshments, so please let me know if you or the person who is attending with you has any specific dietary requirements or any other needs that need to be met to enable you to attend. We will be happy to reimburse you for any costs associated with attending.

Events will take place at community venues in the local area such as The Interchange in Colwyn Bay or Llandudno Library, in the months of May, June and July.

I will be in contact with you shortly to answer any questions you may have and find out if you would be happy to participate. The final dates for all groups will be set according to the availability of attending parents. I look forward to working with you to help improve local sleep services for families of children with developmental disabilities.

Kind regards

Julie Sutton, PhD Researcher, Bangor University

Dear PROFESSIONAL'S NAME

Invitation to take part in a Bangor University Project

Please find enclosed a 'participant information sheet' relating to a Bangor University study the 'Sleep Easy project'. The study aims to improve the experience of families accessing specialist sleep services for children with developmental disabilities. Your team has been selected to take part in some intervention development work which has been approved by the ethics committee at Bangor University, School of Healthcare Sciences.

I would like to invite you to attend two events which aim to develop improvements in sleep services in partnership with parents of children with developmental disabilities. The information sheet provides further details and makes clear your involvement is voluntary and responses confidential.

At each event we will be serving refreshments, so please let me know if you have any specific dietary requirements or any other needs that need to be met to enable you to attend.

Events will take place at community venues such as The Interchange in Colwyn Bay or Llandudno library in the months of June and July.

I will be in contact with you shortly to answer any questions you may have and find out if you would be happy to participate. The final dates for all groups will be set according to the availability of attending parents and professionals. I look forward to working with you.

Kind regards

Julie Sutton, PhD Researcher, Bangor University.

Appendix 8 Contact Forms

The Sleep Easy Project

CONTACT FORM (Parents)

Please complete the form below within 2 weeks and return with signed consent form in the pre-paid envelope to the research team (address below). Thank you.

☐ I am interested in taking part in the 'Sleep Easy' Project

Your name: _____

Preferred Language: _____

Relationship to child with disabilities: _____

Son/daughter's age: _____

Your address: _____

_____

_____

_____

Your telephone number: _____

Your email address: _____

When are good times to contact you? _____

Signed _____

Date _____

Julie Sutton PHD student
Centre for Health Related Research
Bangor University, Fron Heulog, Bangor
LL57 2EF
Tel: 01248 383425/ 07983 245892
Email: j.sutton@bangor.ac.uk

The Sleep Easy Project: Sleep Hygiene Education

CONTACT FORM (Professionals)
Please complete the form below within 2 weeks and return with signed consent form using pre-paid envelope to the research team (address below). Thank you.

☐   I am interested in taking part in the 'Sleep Easy' Project

Your name: _____
Preferred Language: _____

Your work address: _____
                          _____
                          _____
                          _____

Your work telephone number: _____
Your work email address: _____

When are good times to contact you?   _____

Signed   _____
Date   _____

Julie Sutton PHD Student
Centre for Health Related Research
Bangor University
Fron Heulog
Bangor LL57 2EF Tel: 01248 383425/ 07983 245892
Email: j.sutton@bangor.ac.uk

Appendix 9 Consent Forms.

PARENT/CARER CONSENT FORM

Name of study: The Sleep Easy Project

Researcher: Julie Sutton (Bangor University)

Contact details: Centre for Health-Related Research, School of Healthcare Sciences, Bangor University, Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd LL57 2EF.

Tel. 01248 383425 / 07983 245 892 **Email: j.sutton@bangor.ac.uk**

Please initial the boxes

1. I confirm that I have read and understood the information booklet provided and have had the opportunity to ask questions. ☐

2. I understand that taking part in the project is voluntary and that I am free to withdraw at any time, without giving reason. ☐

3. I agree to take part in the project and give my consent. ☐

	Yes	No
4. I agree to being interviewed	☐	☐
5. I agree to being digitally recorded (audio)	☐	☐
6. I agree to being video recorded (if interviewed in a focus group)	☐	☐
7. I agree to the use of anonymised quotes in the research and other publications	☐	☐
8. I agree to allow the information I give to be used for educational purposes in the future	☐	☐

Participant's name   Date   Signature

_____   _____   _____

Name of Researcher   Date   Signature

All details including this consent form information will be held securely on a computer at the School which is protected by a password. All paper copies will be filed in a locked cabinet.

Your unique identification number will be on the top right hand side of this consent form,

please quote on all correspondence. Two forms need completing. Please return one to the

researcher. You keep the other copy. Thank you.

PROFESSIONALS CONSENT FORM

Name of study: The Sleep Easy Project – Sleep hygiene education

Researcher: Julie Sutton (Bangor University)

Contact details: Centre for Health-Related Research, School of Healthcare Sciences, Bangor University, Fron Heulog, Ffriddoedd Road, Bangor, Gwynedd LL57 2EF.

Tel. 01248 383425 / 07983 245 892 **Email: j.sutton@bangor.ac.uk**

Please initial the boxes

1. I confirm that I have read and understood the information booklet provided and have had the opportunity to ask questions.   ☐

2. I understand that taking part in the project is voluntary and that I am free to withdraw at any time, without giving reason.   ☐

3. I agree to take part in the project and give my consent.   ☐

	Yes	No
4. I agree to being interviewed	☐	☐
5. I agree to being digitally recorded (audio)	☐	☐
6. I agree to being video recorded (if interviewed in a focus group)	☐	☐
7. I agree to the use of anonymised quotes in the research and other publications	☐	☐
8. I agree to allow the information I give to be used for educational purposes in the future	☐	☐

Participant's name   Date   Signature

_____   _____   _____
Name of Researcher   Date   Signature

All details including this consent form information will be held securely on a computer at the School which is protected by a password. All paper copies will be filed in a locked cabinet.

Your unique identification number will be on the top right hand side of this consent form,

please quote on all correspondence. Two forms need completing. Please return one to the

researcher. You keep the other copy. Thank you.

Appendix 10 Co-Design Study Podcast Clip.

The CD attachment contains a 5 minute clip of the audio parent podcast, which can be played using Windows Media Player.