NETSCC, HTA

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Autism Spectrum Social Stories in Schools Trial

ASSSIST

Trial Protocol

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Summary

Design: A three phase study following the MRC framework for complex interventions. Pre clinical theory will consist of a systematic review to inform Phase I and II. Phase I will consist of qualitative analysis of a user group to inform an expert writing panel who will develop a manualised Social Stories toolkit. Phase II will assess and evaluate this toolkit in a feasibility study.

Setting: Qualitative interviews and focus groups will take place in CAMHS and primary care settings. The feasibility study will take place in 4 secondary and 10 primary local mainstream schools.

Target Population: Children (aged 4-15) in mainstream school settings with a diagnosis of ASD according to ICD-10 research diagnostic criteria (WHO, 1993), ADI-R (Lord et al, 1994) and ADOS (Lord et al, 2000), who exhibit challenging behaviour in the school setting. This will be assessed using instruments informed by the systematic review (examples include the Strengths and Difficulties Questionnaire (SDQ) and the Developmental Behaviour Checklist (DBC)). We will also set a clear goal with the child, family and teacher and rate it using Likert scales before and after the study. Schools will be approached through an existing network of clinical and educational practitioners and researchers led by co applicant Grigg.

Intervention: A manualised toolkit of appropriate Social Stories for use with school aged children exhibiting challenging behaviour within the mainstream school setting. The toolkit and exact methodology of the feasibility study will be determined in earlier phases of the study. The comparator treatment will be an attention control.
Both arms will receive treatment as usual, for example, behavioural approaches, parenting support, systemic approaches and social skills training.

Outcomes: Primary Outcomes: utility and acceptability to parents/teachers/children of using the manualised Social Stories intervention in a mainstream school setting, as assessed by qualitative interviews and questionnaires. Secondary Outcomes: Acceptability of the research methods (e.g. randomisation procedure) to parents /carers /teachers/ children. Standard deviations of the measures used (e.g. SDQ, DBC, Likert Scales) in the two groups (Social Stories and attention control) to inform power calculation.

Economic Evaluation: The economic evaluation will take the form of within-trial cost-effectiveness analysis that will determine the incremental cost per unit of effectiveness measure for Social Stories compared with an attention control in children with autism.

Sample Size: 10 parents, 5 children/young people, 5 teachers and 5 non research clinicians will be involved in the qualitative interview phase. The manualised intervention will be delivered to 25 children (and 25 children in the control group) in our feasibility study, where we will demonstrate recruitment, attrition and follow up rates, with preliminary estimates of effect size to inform sample size calculations for a full trial.

1.0 Study Identifiers

1.1 Full title of trail
Autism Spectrum Social Stories in Schools Trial

1.2 Acronym
'ASSSIST' - Autism Spectrum Social Stories in Schools Trial

1.3 ISRCTN

1.4 HTA Reference
09/169/07

2.0 Study Background

A Social Story is an intervention designed by Carol Gray (1) for use with children who have autism. Originally, Social Stories were developed for children who had higher functioning autism and Asperger syndrome, but increasingly Social Stories have been used for children with autism. Social Stories are widely available for use in school and other environments without any cost to parents or teachers and with no licensing fees.

Social Stories are short stories which describe a social situation or social skill to help children and young people to understand the situation more easily and hence learn about socially expected behaviors and norms. Importantly, they require consideration and respect for the perspective of the person with autism. They are defined by a number of characteristics and criteria which Carol Gray believes are the hall mark of their success. 'A Social Story describes a situation, skill or concept according to ten defining criteria. These criteria guide story development to ensure an overall patient and supportive quality, and a format, "voice" and relevant content that is descriptive, meaningful, and physically, socially, and emotionally safe for the audience. The criteria define what a Social Story is, and the process that researches, writes and illustrates it" (2).

Some of the characteristics include the fact that they should be written in the first or third person, they should be literally accurate, there should be no negative reference to the young person and at least half of the story should focus on what the person is doing well.

The story describes positive social and situational coping. By regularly reading or hearing a story in which they feature the child learns to adopt a specific skill or set of skills. According to Gray's guidance, they are written in a specific way using a variety of defined sentence types. These include; descriptions of objective, often observable, statements of fact, which collectively describe the context and/or the relevant aspects of a situation, person, activity, skill or concept (descriptive sentences). ; sentences that gently guide the behavior by identifying suggested responses (coaching sentences, previously known as directive sentences); sentences that refer to or describe a person's internal state including thoughts, feelings, beliefs knowledge and opinions, (perspective sentences) and sentences that enhance the meaning of surrounding and describe a positive principle, value or shared opinion (affirmative sentences) (3,1). Gray has recommended a formula for the ratio of sentences which is that the number of sentences that describe (descriptive+ perspective+ affirmative) divided by the number of coaching sentences should be equal to or greater than 2. At least 50% of the Social Story should applaud the skills and traits of the young person.

The common misperception is that the goal of the Social Story is to change problematic behaviour. This was never the case. However, it is often the behaviour which draws attention to the young person's difficulty with a particular concept or skill. The focus of the Social Story is to understand the underlying causes of their frustration or misinformation. The people who write the Stories (Authors) try to share information that supports more effective responses. 'The theory is that the improvement in behaviour that is credited to a Social Story is the result of improved understanding of events and expectations.' (2).

For the purposes of this bid we are referring to Social Stories as those developed by, and adhering to the guidelines of Carol Gray. A wider literature exists using broadly similar interventions but without adhering to these principles and we will refer to these as social stories.

Current evidence suggests that Social Stories can be effective when tackling problem behaviours when they set out to explicitly teach social skills (6). It has been argued that exploring the meaning of behaviour from a child's perspective enables a better understanding and therefore more appropriately designed social stories. Until recently research exploring efficacy and outcome has been confined to case reports and case series. Case reports in children with autism have suggested improvements in social interactions (7-8), choice making in an educational setting (8), voice volume in class (9) and mealtime skills (10). Successfully written individual Social Stories around peer interaction are often targeted specifically at one or two behaviours, for example, saying a peer's name and looking at their face when talking (11).

Successful cases have also been reported in reducing tantrums in a 5 year old boy with autism (12) and a 12 year old boy with autism (13). Disruptive behaviours have been decreased in one small study with three children with autism (14) and it has also been reported to reduce behaviours associated with frustration (15).

Often the Social Story is used alongside other interventions. Social Stories have been shown in case series to be useful in three 7-11 year old children with autism when used adjunctively with behavioural social skills training strategies (16), and when presented as' comic strip conversations' (17) in a 14 year old with Asperger syndrome (18). One case series of four (6-9 year olds) reports on Social Stories being used in musical form (19). They have also been used in conjunction with visual symbols and computer-based instruction (20).

Social Stories have also been found to be effective in children who demonstrated challenging behaviour in a school setting but were not diagnosed with Autism Spectrum Disorder (ASD). Two studies by Toplis and Hadwin (21) and Whitehead (22) found that successful intervention outcomes in children with challenging behaviour could be related to perspective taking difficulties relative to age, a specific feature of autism. PPI Co- applicant Whitehead has specifically explored the use of Social Stories in a mainstream context.

In terms of the effective delivery of Social Stories small studies appear to suggest that it is possible to train tier one professionals, for example teachers, in the use of Social Stories and for them to have benefit for children on the autism spectrum (23). Research has examined their use in special education (24) and mainstream education settings (25), as well as their application within the home (26).

The strength of Social Stories for autism relates to some of the specific needs of children with autism, such as the presentation of material in a visual format, alongside simple

language, concrete and clear descriptors of social routines and behaviours, gradual learning of alternative perspectives, repetition of learning and positive learning (27-30)

Two systematic reviews have been conducted on Social Stories, both of which have found that empirical data suggests that the effects of Social Stories appear to be highly variable (5-6). The first, conducted in 2006, included published and unpublished studies prior to 2003. . The review found that there were numerous individual outcomes in the publications (5). These included suggestions that where, when, with whom and how Social Stories are used needs further research. Alsosome small case series report that the positive outcomes seen in case series continue after the Social Story is withdrawn (31), whilst others have not found this (32). Reynout & Carter (5) concluded that in some situations Social Stories can be very effective but only under certain conditions. Their review was criticised in a second, more recent review by Kokina & Kern (6) for not measuring a number of variables. They found broadly similar results, namely that Social Stories have variable effectiveness. Kokina and Kern suggested that the success of Social Story intervention may be dependant upon a number of variables (6). In particular the studies collated in the review suggested that the general education setting produced substantially larger effect sizes on student's behaviour than those implemented at home or in self contained settings e.g. separate schools or self contained classrooms. Additionally they found that brief interventions between 1-10 sessions were more effective than 10 + sessions and that studies which used a number of Social Stories per child were more effective than those that used one per child. They also identified Social Stories that used illustrations as more effective than those which used written text alone, with additional variables dependant on behaviour and the situations targeted.

Kokina and Kern (6) concluded that 'additional methodologically robust interventions are needed.' They suggested that further studies should include data on the differential results of children with different diagnoses, generalization and maintenance of the skills learnt the social acceptability of the intervention and treatment fidelity. The review also called for studies which separated out the effects of Social Stories from other methods.

Research methodologies vary greatly, as discussed by Rust and Smith (33) who highlight the very variable approaches to researching Social Stories, and the paucity of good quality research in the area. Additionally implementation and faithfulness to the Social Stories model is likely to vary in clinical practice. The vast majority of case reports have been carried out in the USA, and the guidelines about language have been written for the American audience, so there is a need to make sure that interventions are culturally tailored to their target audience. These concerns have been echoed in the few reviews that exist on the effectiveness of Social Stories. For example a review undertaken by Sansosti and colleagues (34) criticized Social Story research for its lack of experimental controls and problems with the integrity of implementation as being methodological weaknesses. These considerations make our study very timely.

Social Stories can be difficult to design well, but their relative ease of implementation has made them a popular intervention for parents/carers and teachers to use; for example Social Stories were included in the development of an Autism Toolbox for Scottish Schools (35). While there is a general consensus that Social Stories have much potential to improve behaviour, the current evidence base is weak and research with strong methodologies is required. There are still important gaps in our knowledge, including which variables make Social Stories effective, the acceptability, longevity and generalisability of the intervention, and who it is effective for. In particular the intervention has largely been

used in special education and its use in mainstream education, where it has a large potential, needs exploring.

2.1 Research objectives

Social Stories for autism represents a complex intervention. This technology is not yet sufficiently evolved such that it would be premature to design and conduct a full scale randomised controlled trial at this stage. The aim of our study is to build upon previous research to develop a manualised Social Stories intervention for use with Autism Spectrum Disorder (ASD) children in mainstream schools that has the effect of reducing challenging behaviour. We will conduct a feasibility study which will inform the design of a full Randomised Controlled Trial (RCT) that includes a justification and description of appropriate costs, outcomes and parameters to include cost effectiveness. In line with the Medical Research Council (MRC) complex interventions framework, we will first meet the following aims to ensure our trial is successful and informative to the HTA and the NHS:

1. To carry out a systematic review examining the use of Social Stories and other social stories in autism spectrum disorders, with particular reference to an outcome of reducing challenging behaviour in mainstream school aged children (co- applicant Kokina was first author on the 2010 systematic review).

2. To conduct a qualitative analysis, with user interviews and a user focus group, to gather information relating to the optimum design and use of Social Stories in children and young people with autism spectrum disorders.

3. To form an expert writing panel and to develop a manualised toolkit (including a training package) of appropriate Social Stories for use in mainstream school children and young people.

4. To conduct a feasibility study in the form of a randomised controlled trial comparing the manualised Social Stories intervention with an attention control (demonstrating recruitment, delivery of the intervention and successful follow up).

5. To establish the acceptability and utility of the manualised Social Stories intervention to parents and carers.

6. To identify parameters, outcomes and cost effectiveness from the feasibility study in order to inform a future full scale RCT.

Subject to the achievement of the previous objectives and further findings, we will proceed seamlessly to a fully powered RCT to evaluate the clinical and cost effectiveness of Social Stories.

3.0 Study Design

The study will be carried out in three phases, following the guidelines set out by the Medical Research Council in their ‘framework for development and evaluation of RCTs for complex interventions to improve health.’ Each of the three phases corresponds to one or more of the research aims.

3.1 Pre-clinical/ theoretical phase

This phase aims to obtain up to date information about the current available research on how Social Stories are written and used, and how this is faithful to current theories on what makes them effective. This process will inform the subsequent research aims, for example ensuring the best choice of interventions and predicting any major confounders or strategic design issues. This will be achieved by conducting a systematic literature review. The reviewers in this stage will include academics, clinicians and PPI representatives including individual parents/carers and the National Autistic Society.

1) To carry out a systematic review examining the use of Social Stories in autism spectrum disorders, with particular reference to an outcome of reducing challenging behaviour in mainstream school aged children.

The systematic review will identify as many potential uses of Social Stories that may impact positively on behaviour as possible. We will also identify studies which have failed to have 'positive' impact on behaviour. We will include all such studies but will pay particular attention to those used in a mainstream school setting and those potentially generalisable across populations, age ranges and cultures. The review has two broad aims: first, to provide a comprehensive description of how Social Stories have been used in education and clinical practice, and secondly to provide a quantitative estimate of the effectiveness of this intervention.

On the basis of the previous systematic review of effectiveness conducted in this area by applicant Kokina, we expect that the majority of the effectiveness studies reviewed will use a single-case experimental design. Therefore we have specifically designed this systematic review to synthesis single case data. Whilst our group has the expertise and experience in using standard data synthesis strategies for randomised trials, and could employ these if found to be necessary, we believe that given the previous work of co-applicant Kokina (6) these standard strategies will need to be modified given the nature of the available research in this area. The research team has specific expertise in the design and analysis of these approaches (Co-applicants Kokina and McMillan).

We will use the following definition of a single-case design throughout the review: a study that uses repeated collected of quantifiable (numerical) data on a single case, typically involving repeated measurement over a baseline period; experimental manipulation through some form of randomization procedure may or may not take place; such an approach may or may not include replicating the design over several cases to form a single case series (36). It is of note that this definition excludes descriptive case studies and case histories; it also excludes simple pre-post designs of a single case, in which measurement is taken once at pre-treatment and repeated once at post-treatment.

Whilst social stories are widely referred to in education and clinical practice their precise definition is malleable. On the other hand Social Stories™ have a clear definition and structure. Our intention is to look particularly closely at Social Stories™, because of the clarity and integrity of them as an intervention, and also because most of the research literature on social stories are actually about Social Stories™. However, our systematic review will also include studies which have used social stories in a broader sense, in order not to miss important literature. We will take care to distinguish between these and Social Stories.

3.1.1Search strategy

Data sources. We will search the following sources (from inception to current for databases):

1. Databases of published literature (e.g., PsycINFO, MEDLINE, EMBASE, the Cochrane Library, ERIC, The British Education Index, Applied Social Sciences Index and Abstracts, International Bibliography of the Social Sciences, Social Services Abstracts, SCI/SSCI, Soc Abs, Social Care Online, Campbell Library, HEED)
2. Databases of studies in progress, unpublished research and additional grey literature sources (e.g., Current Controlled Trials, World Health Organization International Clinical Trials Registry Platform, National Research Register, OAISTER, Index of Theses, ZETOC)
3. Websites (including American Psychiatrics Association, Mental Health Foundation, MIND, Royal College of Psychiatrists, National Collaborating Centre for Mental Health, National Institute of Mental Health, National Autistic Society) and others identified through Intute and advice from content experts and stakeholders
4. Reverse citations searches of key social stories publications
5. Examination of reference lists of studies meeting inclusion criteria
6. Contact with experts in the field, including national and international experts

Search terms. To identify studies for this review databases will be searched using a combination of terms related to Social Story interventions, children and autism spectrum disorders (ASD). The precise search terms and strategy will be developed by co-applicant Glanville who is an expert in this field. Co-applicant Glanville will develop the search terms in collaboration with content experts, PPI representatives and co-applicant Kokina, who has conducted a previous meta-analysis in this area.

3.1.2 Inclusion-exclusion criteria

The systematic review will use the following inclusion criteria:

Population. All children or young people between 4-15 years of age with a diagnosed autism spectrum disorder. Our foreknowledge of the literature suggests that there may be limited data on social stories interventions for this population. Given the provisional status of the current evidence base, we will not require that the diagnosis of ASD is established by a research gold-standard method. We will include all studies in which the sample is described as having ASD regardless of the method by which the diagnosis was made. The quality assessment of the studies will include a question regarding diagnostic method.

Intervention. Any social story used that may have impacted upon behaviour regardless of initial purpose. As described above, we intend to use a broad definition of social stories that is not limited to Social Stories ™.

Comparators. For between-group designs we will include any comparator, including no intervention, wait list, treatment as usual, attention control, or active psychological intervention. For single-case data, the comparison will be a no-treatment, usual treatment or alternative treatment phase as part of the within-subject design (e.g., baseline phase before introduction of intervention). In some cases, single-case experimental designs also use a between-subject control (e.g., counterbalanced AB design); in these cases, the comparator will also be, no-treatment, usual treatment or alternative treatment.

Outcomes. We will include any standardised measures of behavioural outcomes (e.g., Strengths and Difficulties Questionnaire) as well as non-standardised measures of objectively observable behaviour (e.g., daily counts of a target behaviour made by the

parent). Our reason for inclusion of non-standardised measures, is that single-case experimental designs rely on frequently repeated measurement as a main strategy through which internal validity is protected. Frequently repeated measurement necessitates the use of non-standardised approaches, because most standardised approaches are not validated for such frequent use. We will also look for secondary outcomes including pro social behaviours , behaviours specific to autism spectrum disorders and quality of life measurements.

Study design. For the first aim of the review (descriptive overview of how social stories have been used in clinical practice) we will include any type of study design, including case histories or brief descriptive accounts. For the second aim of the review (effectiveness of social stories) we will include any between-group design, including RCTs and controlled studies that do not use randomised assignments. In terms of single-case designs, we will include any study that uses repeated measurement as part of a recognised single-case design to rule out threats to internal validity; these include AB designs, reversal / withdrawal designs, changing criterion methods, multiple baseline, small N designs that use counterbalancing across participants, and alternating treatment designs (ATD). We will also include studies that use a hybrid of these basic approaches. We are aware that the capacity to rule out major threats to internal validity varies between the designs. We will evaluate this as part of the assessment of study quality. Our decision to take an inclusive approach to the design of effectiveness studies is based on our foreknowledge that the literature in this area is likely to be limited and of variable methodological quality.

3.1. 3 Data extraction

Data will be extracted independently by two researchers and will include characteristics of the intervention (e.g., type of social stories, duration), primary and secondary outcomes, and the methodological features of the studies. For between-group designs we will judge methodological quality and sources of bias using the Cochrane assessment tool (37). For single case methods, we will use the quality assessment tool developed by Tate et al. (37).

3.1.4 Data synthesis

In the first instance we will produce a narrative overview of both the uses of social stories in clinical practice and data on the effectiveness of those interventions. In terms of effectiveness, we suspect that there will be an insufficient number of between-group designs to conduct a meta-analysis of these data alone; there is however likely to be a larger number of single-case studies. There are a number of available effect size metrics for single-case data and methods of quantitatively synthesising these data, but there is no clear consensus about the most appropriate method for quantifying and combining the results of single-case studies (39-44). There is also debate about the appropriateness of combining data from between-group designs and single-case designs in a single meta-analysis (43). We will not, given this lack of consensus, restrict our analysis to one method. Instead, we propose to quantify and synthesise the single-case data using a small number of the more commonly used methods (e.g., percentage of non-overlapping data, R^2 change) and examine whether the results and conclusions drawn are robust to variations in these methods of calculation.

3.2 Phase One – Modeling and Refinement of Social Stories manual

Phase one relates to aims two and three of the research objectives and focuses on identifying the components of the intervention, and the underlying mechanisms by which they will influence outcomes to provide evidence that you can predict how they relate to and interact with each other:

2. *To conduct a qualitative analysis, with user interviews and a user focus group, to gather information relating to the optimum design and use of Social Stories in children and young people with autism spectrum disorders.*

3. *To form an expert writing panel and to develop a manualised toolkit (including a training package) of appropriate Social Stories for delivery in the school setting.*

This phase of development of the intervention (drawing on the literature work from the Pre-clinical theory phase) will use qualitative methods to take stock of current experience and elicit the views and experiences of various stakeholders, in order to ensure that our intervention can feasibly be delivered in this particular context and to ensure the intervention is acceptable to users and providers. The interviews will be focused and the analysis (using a Framework approach) will be specifically geared to address questions of the intervention design and would be conducted over a relatively short space of time.

The results of the systematic review, including the most promising Social Story themes, templates and uses, will be fed into two groups. These two main groups will work separately but will come together as mutually agreed (at least three times) to share ideas, findings, suggestions and developments. These two groups will be I – a user group and II – an expert panel.

3.2.1 User Group
The user group will comprise of:

- Ten Parents/carers
- Five teachers
- Five young people
- Five non-research clinicians

All of the user group will have previously used Social Story interventions with children or have used a Social Story intervention themselves.

Users will be drawn from at least two geographically distinct areas and will be given information about the study through local pediatricians, child psychiatrists, clinical psychologists and child mental health nurses. Should they choose families will be invited to meet a researcher for further information on the study. We will write to clinicians and teachers in the York area who may use social stories with information regarding phase 1 of the study and presentations about the research will be given to local autism support groups (such as the ASCEND group (45).

The role of the user group will be to ensure the best design and use of social stories, and the practicality and feasibility of the intervention to be delivered. We will use interviews and focus groups to gather a range of opinions and themes about:

- the helpful and unhelpful parts of the social story intervention,
- the characteristics of individuals best placed to deliver the intervention,
- determining which professional groups are best equipped to deliver and or support this package;
- mode of delivery including the role of parents or carers
- the style and content of the manual.

- the construction of the social stories (for example the ratio of directive to descriptive sentences in the story, the use of pictures etc),
- teaching in the social story theory, and dissemination,
- joining the steering group for collaboration on monitoring study progress,
- methods of dissemination of study findings for service users.

With informed consent a qualitative researcher will be present, who will transcribe the discussions and will thematically analyse key themes around potentially successful childhood outcomes.

Qualitative semi-structured interviews will be conducted using a topic guide developed to ascertain information on perceptions about feasibility and acceptability of the intervention. In addition we will gather a range of opinions and themes about the helpful and unhelpful parts of the social story intervention, the characteristics of who would be best placed to deliver the intervention, determining which professional groups are best equipped to deliver and or support this package; mode of delivery including the role of parents or carers, and the style of the manual. Where appropriate this may include information on the construction of the social stories (for example the ratio of directive to descriptive sentences in the story, the use of pictures etc), teaching in the social story theory, and dissemination.

Our group has considerable experience and expertise in this area and we would draw upon this when conducting the qualitative component of our programme of research. In addition we will carefully ask parents/carers and young people themselves which of many possible ways of conducting the qualitative feedback they would prefer and will be guided by this. In this way the user participation will be central in determining the best mechanisms for qualitative feedback.

Qualitative interviewers will have experience of working with children and young people with autism spectrum disorders. Techniques that may facilitate good feedback used in previous work will be employed. This may include using visual prompts, drawing, the Picture Exchange Communication System, using MSN and a range of other non-threatening techniques for discussion and feedback, depending on developmental stage of the child or young person.

3.2.2 Expert Group

The expert group will comprise of:

- Social Stories designer Carol Gray (co-applicant)
- Other co-applicants with expertise in Social Stories,
- Other co-opted experts in the field of autism and the use of Social Stories,
- A PPI parent (co – applicant author of a paper on the use of Social Stories in mainstream schools)
- National Autistic Society (NAS) representative
- At least 2 people who have expertise in developing and delivering training packages for parents in a child mental health setting.

The expert panel has been kept separate at the request of the PPI representatives in the research design as a mechanism for ensuring that their voices are heard in the user group and not drowned out by clinicians or academics. The important need to bring the groups together will be met by regularly mutually agreed meetings which will give equal opportunities for everyone to have their say.

The expert group will formulate a manualised approach informed by the systematic review, the expert opinions, user group feedback and the thematic analysis from the qualitative researcher. The objectives of the expert group are to:

- To balance and evaluate the existing Social Story model and write guidelines with new information arising from the systematic review and user and expert groups.
- To produce a manual on using Social Stories
- To produce a training package to train the service providers in use of the manual and provide familiarisation with the manual itself
- To focus on the design of an intervention that is fit for purpose in a mainstream school setting
- To advise about recruitment to the study

These two groups (an expert panel and a user group) working in tandem will therefore use an iterative process of feedback and redevelopment. This will lead to the generation of a list of Social Story uses that:

1. Have the strongest evidence base for efficacy and generalisablity
2. Have had positive user feedback and evaluation
3. Can be manualised
4. Are readily teachable

The focus groups will provide insight into the process of developing and using a Social Story as well as its content. This will aid us in the construction of a manualised toolkit of appropriate Social Stories for delivery in the mainstream school setting, adaptable for individual use, which we will validate in phase two. This information will be collated for the panel of experts who will formulate a manualised approach informed by the systematic review, the expert opinions, user group feedback and the thematic analysis from the qualitative researcher. This manualised approach will then be represented to the focus groups for further discussions using a SMART (Specific, Measurable, Achievable, Realistic, and Time limited) approach, for example; are the Social Stories from the manual likely to be faithful to the Social Story model? The group will include at least two people who have expertise in developing and delivering training packages for parents in a child mental health setting. In this way the manual will be complemented by a training package.

A key part of this process will be the need to balance and evaluate the existing Social Story model and rules with new information arising from the systematic review and user and expert groups. We will have full intellectual freedom to challenge and adapt any aspects of the model. In particular we are focusing on the design of an intervention that is fit for purpose in a mainstream school setting, where previously it has been extensively used in special school, home and clinic settings.

3.2.3 Manual Development and Pilot

The manual will be written by the expert group informed by the systematic review, the expert opinions and user group feedback. The user groups have already been clear about the need for;

- simplified, compact guidance on producing Social Stories,
- a clear, starting point from which to develop Social Stories once a challenging behaviour had been identified
- guidance on the use of strategies for making sense of this behaviour and how to determine the focus of the Social Story, which may be different to working on the challenging behaviour as such

- collaboration in writing Social Stories between parents and teachers,

Once initial decisions have been made regarding the refined intervention, five user/family and service provider 'dyads' as identified through the user group will be asked to 'walk through' the proposed package of care. Both users/family members and practitioners delivering the care will be interviewed (using a topic guide) to elicit their experiences and to raise any issues of concern. Practitioners applying the manual will also be asked to keep a reflective diary of the intervention process to note any issues or observations concerning the service provision. In particular they will be asked about facilitators and barriers to its delivery within existing service models; their perceptions of whether the intervention does or doesn't work well, including their understanding of why; and any experiences users have receiving the intervention which highlight where it works well and less well, including perceptions of reasons for discontinuation with the intervention by any participant. These will be analysed as qualitative documents. Any final amendments would then be made to the manualised Social Stories intervention.

The manual will be revised at all stages in the process. Throughout the intervention period, practitioners applying the manual will also be asked to keep a reflective diary of the intervention process to note any issues or observations concerning the service provision. Any final amendments would then be made to the manualised Social Stories intervention

3.2.4 Research outputs

By the end of this developmental phase (PHASE I), we will have produced a manualised intervention (including a training package to train the service providers in use of the manual and the manual itself) which will have been 'piloted' by a sample of users and their families who would be eligible to participate in the trial. Our intervention will then be amenable to evaluation in a pilot trial feasibility study.

This work will then be finalized into a manual for use in phase II. The manual content will be determined by phase one of the study but may be likely to include:

- A template element
- Guidance on Social Story development and construction
- Elements that can be tailored to any child
- Specific guidelines on Social Story elements that reduce challenging behaviour.

3.3.1 Phase Two - Exploratory Trial

Phase two involves describing the constant and variable components of the Social Stories intervention and the development of a feasible protocol for comparing the intervention to an appropriate alternative. This phase relates to aims 4, 5 and 6 of the research objectives as follows;

4. To conduct a feasibility study in the form of a randomised controlled trial comparing the manualised Social Stories intervention with an attention control (demonstrating recruitment, delivery of the intervention and successful follow up).

5. To establish the acceptability and utility of the manualised Social Stories intervention to parents and carers

6. To identify parameters, outcomes and cost effectiveness from the feasibility study in order to inform a future full scale RCT.

The validation stage of the manualised toolkit developed in phase 1 will take the form of a feasibility randomized controlled trial comparing the use of the toolkit versus an attention control l. The exact procedures will be informed by the preclinical theory phase and phase I of the study.

3.3.2 Identifying Participants

As with all local research studies we will let the local community know that they are taking place. We will give presentations about the research to local autism support groups (such as the ASCEND group; (45)). We will also provide presentations and leaflets to local autism support groups and through clinicians to parents of children with ASD attending mainstream schools. In the North Yorkshire and York PCT we have approximately 800 children with diagnoses on the autism spectrum. Given previous studies in autism in the locality and good relations with parents and parent support groups, we feel confident that will achieve our recruitment targets.

Letters will be sent to the parents of all children on the autism spectrum attending participating mainstream schools to let them know about the study and to provide a point of contact should they be interested in participating. There is in York and surrounding area a York Autism Spectrum Disorders Forum which is a multi agency multi disciplinary forum for diagnosing and discussing provision of supporting interventions for all local children on the autism spectrum. We will cross check the education list against this list to ensure that we have missed no children in participating mainstream schools with ASD.

We will use other methods of recruitment where appropriate, as advised by the service user parents on the steering group.

3.3.3 Recruitment

Interested families will be invited to meet a researcher for further information if their child has a diagnosed ASD and behavioural problems in a mainstream school. We will recruit 50 children attending mainstream schools, with autism spectrum disorders (Autism, Asperger syndrome and Atypical autism), using ICD-10 research diagnostic criteria [RDC] (46), supported by ADI-R (47) and/or ADOS (48). Other developmental disorders will not be included.

Schools will be approached to take part in the study through an existing network of clinical and educational practitioners and researchers. Co-applicant Grigg has previously facilitated many research studies across the full range of schools as part of his role within the Local Authority. There are 72 primary schools in the York Local Authority and 10 secondary schools. Two of these secondary schools are designated as ASD supportive schools, although all the other schools have some young people with ASD within them. We have already had preliminary discussions with a number of secondary and primary schools that are supportive of this bid and we are confident that we can recruit 30 children within primary schools and 20 young people within secondary schools without difficulty. We anticipate that this will be in 4 secondary schools and 10 primary schools, although we will test the feasibility of this and be adaptable to recruit more or less by rolling out school involvement and leaving enough time in the study for flexibility. We will stratify school randomization to take into account numbers of ASD children, levels of ASD support, socio economic indices and Ofsted value added scores. We will ask the local multi agency autism strategy group to compare the two groups of schools generated for equivalence. Any concerns will be fed back to the steering group and the matching process will be considered as part of the feasibility work.

3.3.4 Randomization

We will adopt a cluster randomization approach. This is to minimize the likelihood of participants in the different intervention groups being affected by changes in teacher or school behaviour. It also recognizes that social story expertise within a school is likely to be enhanced by sharing of skills and networking of ideas, and this design therefore more accurately reflects what would happen if social stories were used within a school in practice. We will use the qualitative interviewing within the feasibility design to explore this further, as described below.

To reduce detection bias we will first recruit the mainstream schools to be involved in the study. Then subsequently we will consent children and their families within these selected schools for involvement in the study. Only then will we randomize the schools into schools where the intervention takes place and those where it does not but where we are monitoring outcomes in the same way as in the intervention school. Participants will be randomised at whole school level, thus all children with ASD within any given school will be randomised to the same arm of the trial. This will minimize contamination as the likelihood of participants in the different intervention groups being affected by changes in teacher or school behaviour will be reduced. In order to limit detection bias, participants will be recruited prior to school randomisation. The children in the intervention school will receive the Social Story intervention. The control school children will receive an equivalent amount of time reading a story of similar length and linguistic complexity a range of which will be chosen by the expert panel as an attention control. In both schools children will receive all other treatment or support as usual and we will monitor carefully what this is.

3.3.5 Trial Intervention

Schools will be randomised to one of two arms; the intervention group and the control group. Prior to the intervention teachers and parents of children in the intervention group will undertake training on Social Stories. We will run Social Stories training workshops informed by the earlier parts of the study. The Social Stories will be delivered in a way that is informed by the pre clinical theory and phase 1 of this study using the most promising mode of delivery. This will include who designs the Social Story with parent and child, how this is achieved, and how and when the Social Story is delivered with the child. For example this could include a teacher, support assistant, special educational needs co-ordinator and /or a parent and they will receive support as part of the study from the clinical researchers. The precise from of this support will be designed using findings from the pre clinical theory and phase 1 of this study.

We will check the construction of the Social Stories paying particular attention to construction guidelines as laid down by the original Social Stories model. We will also explore any other parameters, including construction, content and delivery that have been found to be related to outcome as part of the systematic review and user and expert panels. As part of this, for example, we will measure goals of the intervention, intervention agents, intervention setting, timings of the intervention, length of the intervention and number of stories per child. We will also assess characteristics such as the number and distribution of pictures and photographs, and the length and complexity of sentences, types of behaviours described and any allied interventions or support.

Whatever the precise methodology we will keep a record of all the design interactions and precise number, timings and locations of delivery. We will also record attrition, non

compliance and any distress. We will have a clear methodology of how to deal with any difficulties including child distress.

3.3.6 Control Group

Children in the control schools group will receive an attention control and treatment as usual. At the end of the study, a second free workshop will be offered to the parents of children participating in the control group on the use of Social Stories. All participants in our study will continue to be able to access care and treatment as usual throughout the trial regardless of which arm of the study they are randomised to. To clarify, no participant in either the intervention or control arm will be denied NHS treatment by virtue of participating in the trial.

3.3.7 Withdrawal

Withdrawal can occur at any point during the study at the request of the participant. If a participant indicates they wish to withdraw from the study, withdrawal will be clarified as to whether the withdrawal is from the intervention, from follow-up or all aspects of the study. Where withdrawal is only from the intervention then follow-up data will continue to be collected. Data will be retained for all participants up to the date of withdrawal, unless they specifically request for their details to be removed.

3.3.8 Duration of intervention period

The intervention for participants will be delivered over 4- 6 months.

3.3.9 Inclusion and Exclusion Criteria

In the feasibility RCT stage of the study, children will only be recruited between the ages of 4-15 years, if they have a definite diagnosis of an ASD and have behavioural problems in school (as reported by parents and teachers). We will prescreen children for challenging behaviour, using clinical interviews and measures informed by the systematic review, which may include the Strengths and Difficulties Questionnaire and the Developmental Behaviour Checklist We have deliberately kept the age range broad in order to explore the acceptability of this intervention across age ranges and different settings (e.g. primary and secondary schools) and inform a fully powered trial.

This is a pragmatic trial which recognises the frequency and complexity of co-morbidities in children with autism spectrum disorders (ASD). We will therefore not exclude on the basis of comorbidity since the intervention would eventually be offered to this group if adopted more widely in the NHS. This will ensure that the research remains relevant to everyday clinical practice. This means for example that we will include children and young people with a learning disability, epilepsy, attentional problems, neurological disorders or syndromes (e.g. CHARGE syndrome, Fragile X syndrome etc). We will exclude any child from the study if they have used a Social Story within the last six months. We will exclude children and young people if they are likely to be moving school during the trial period, or if they develop behaviour or illness that warrants admission to a psychiatric unit (e.g. psychosis, serious self harm). These exclusion criteria were informed by discussions with parents who had previously used Social Stories in a focus group prior to submitting the full application. Children will only be randomised into the trial if they exhibit challenging behaviours using the validated cut offs from the Strengths and Difficulties Questionnaire or the Developmental Behaviour Checklist

3.3.10 Proposed sample size

We will use the information gathered in the pre-clinical theory stage of the study to determine sample sizes needed for the feasibility study / phase 2 of the study. The sample

size will be determined in order to be adequate to estimate the critical parameters for a full RCT. We will be estimating recruitment rate. With 50 participants in total we will be able to estimate the recruitment rate to within 10% based on 85% recruitment, using a 95% confidence interval (Dixon & Massey, 1983). We will demonstrate recruitment and follow up rates, with preliminary estimates of effect size to inform sample size calculations in a fully powered RCT.

3.3.11 Blinding

Blinding of the participants will not be feasible, nor is blinding of all members of the study team who are actively involved in the administration of the study. However, members of the study team responsible for the statistical analysis will be kept blind to group allocation.

3.3.12 Follow-up

Follow-up will be for a minimum of six months post-randomisation. Where a participant has been lost to follow-up their data will be included in the main analysis up to where they have been lost to follow-up. Where a participant is lost to follow-up, efforts will be made to contact the participant.

3.3.13 Qualitative Study

There has been little previous qualitative work exploring issues of effectiveness and acceptability of Social Stories amongst this population. An in depth appreciation of these issues will be essential for any future implementation of the manualised intervention if it proves to be clinically and cost effective. This process evaluation we will examine using semi-structured interviews, which behaviours are most problematic to children, their families and teaching staff, expectations of the technique and which of the treatment outcomes are of greatest importance to children, their families and teachers. In addition, data will be collected on the pilot trial process itself. We will ask questions relating to the acceptability of the treatment, the randomisation procedure and the methods of data collection. We will also explore qualitatively the experiences of the teachers in the context of the cluster randomisation design. We will be particularly interested in comparing those schools where several children have used the intervention compared to schools where smaller numbers of children are involved. We are interested in the systemic and individual effects. All interviews will be conducted using a topic guide to ensure consistency across participants, however, the format will be flexible in order to allow participants to generate naturalistic data on what they constitute as important and/or successful in terms of treatment outcomes.

It is anticipated a sample of 15 children and an associated family member (10 from the intervention arm and 5 from the usual care arm) would provide sufficient data to answer these questions. Trial participants will be purposively sampled on characteristics such as age, gender, extent of behavioural problems. All participants would be interviewed after the writing of the Social Story face-to-face at a location convenient for the participants with the family members; this would enhance rapport and facilitate data collection.

In addition, a sample of 5 professionals and 5 parents/carers delivering the manualised intervention will be interviewed. The interview will be semi-structured, asking them to record: their experiences associated with delivering the intervention, in particular facilitators and barriers relating to its delivery within existing service models; their perceptions of whether the intervention does or doesn't work well, including their understanding of why; and any experiences they have with users receiving the intervention which highlight where it works well and less well, including perceptions of reasons for discontinuation with the intervention by any participant.

3.3.14 Quantitative Collection and Analysis

The following questionnaires will be completed at three times; before, six weeks into the trial and six months after the trial. Teachers, parents and children will complete the Strengths and Difficulties Questionnaires, Developmental Behaviour Checklist, and the generalisability questionnaire about the children. Parents will complete the General Health Questionnaire about themselves. The target behaviour will be agreed at the beginning of treatment and the outcome questionnaire completed at 6 weeks and six months into the trial.

This will enable us to examine the change in behaviour scores over the course of the intervention and monitor whether the skills learnt are maintained over a longer period.

Change in parental stress will be measured using the General Health Questionnaire at the beginning, midpoint and end of the trial. We will also assess acceptability to parents and teachers using a 12 item Intervention Rating Profile at the end of the study. We will examine the effects of diagnosis and age of the participants when carrying out the analysis. Qualitative interviewing will take place to obtain feedback about acceptability and utility to parents and carers.

We are carrying out a feasibility study to assess parameters such as:

- willingness of children and young people and their parents to participate
- issues around consent and assent in children and young people with ASD
- acceptability of the intervention with children, young people and families
- acceptability of not receiving the intervention
- qualitative aspects around the process of delivery of the intervention including who delivers it and how it is formulated, when and how it is delivered and for how long
- acceptability of training for teachers and other staff, and the feasibility of arranging an delivering training in a busy school environment (e.g. release for training)
- follow up and response rates
- attrition and non compliance rates
- characteristics and utility of the outcome measures

At this point we have not chosen a main outcome measure as it is one of the purposes of the feasibility study to explore which outcome measures are most likely to be useful, which may include:

1. Developmental Behaviour Checklist (DBC; (49)), which includes ASD subscales and a disruptive subscale (parent and teacher)
2. Strengths and Difficulties Questionnaire (SDQ; (50)),which includes conduct, hyperactivity and prosocial subscales (parent, child, teacher)
3. Likert Scales for target behaviours designed and agreed by the focus groups/ expert panel (parent, child, teacher, and clinician). In this way we will develop a goal based outcomes scale and test its usefulness against the other instruments as part of our feasibility study.
4. Generalisability will be assessed using questionnaires designed for each individual child that ask about the frequency of challenging behaviours for both the Social Story target behaviour and for other associated or related behaviours (e.g. if more positive behaviours in the class result in reduced destruction in the class are there also reductions seen in other settings, such as the dining hall, home etc). These will be parent and teacher based questionnaires used before and after the intervention.

5. General Health Questionnaire (parents)
6. Intervention Rating Profile (51), which has been widely used to assess parents views of interventions used with their children. A separate questionnaire will be developed to be administered by parents to their child to assess the views and experiences of the child of using Social Stories.

Descriptive statistics will be calculated for recruitment rates, follow-up rates and attrition. Descriptive statistics and 95% confidence intervals will be calculated for the outcome measures. We will use the data to develop estimates for a fully powered RCT that will include estimates of change in outcome measures over time taking into account attrition and follow up rates.

Although the feasibility study is not powered to detect differences in outcome measures we will undertake inferential statistics (Chi-squared tests for categorical data, t- tests for continuous data, and Mann-Whitney tests for ordinal data). All analyses will be undertaken on SPSS version 17.

3.3.15 Qualitative Analysis

Data will be analysed as set out in phase I. As participants in this qualitative sample would also have responses to the quantitative data collected for the trial, this will allow for the possibility of taking a mixed-methods approach to data integration, in which the two forms of data can be used in a complementary way. For example, by using approaches such as 'triangulation protocol'[1] at a thematic level and 'mixed methods matrix'[2] at the level of individual cases, for data integration, greater insights will be gained from the process of data integration than would be obtained from using the two data types in isolation (52-53).

3.3.16 Economic analysis

The economic evaluation will take the form of within-trial cost-effectiveness analysis that will determine the incremental cost per unit of effectiveness measure for Social Stories compared with an attention control in children with autism. The choice of effectiveness measure will be informed by the choice of the primary outcome in the study. The evaluation will be carried out from the UK societal perspective, taking account of the use of health services, education services, and social and voluntary services over the study period. The cost of Social Stories intervention will be calculated using a bottom-up estimation of the time spent by professionals delivering the intervention, the cost of training and other resources used. Unit costs of health service use will be obtained from the UK national database of reference costs. The cost of social services will be calculated from the Unit Costs of Health and Social Care, produced by the Personal Social Services Research Unit (54), and the cost of other professional support will be estimated from relevant salary scales.

This pilot study will inform the choice of appropriate effectiveness measure for the economic analysis. Also it will identify the relevant resource use categories for the cost-effectiveness analysis, and evaluate the feasibility and challenges of measuring costs and outcomes in the target population.

3.3.17 Research outputs

1. A manualised approach to Social Story intervention design developed for use in a mainstream school setting. Coapplicant Gray has clarified that Social Stories are widely available for use in school and other environments without any cost to parents or teachers and with no licensing fees. Carol Gray has created no stipulations that may impede an

open minded enquiry into the efficacy and design of Social Stories. She will provide consultation in the further development of the Social Story intervention in this study and her overriding motivation is the provision of improved interventions and outcomes of children on the autism spectrum.

2. Feasibility findings for a full scale randomized controlled trial of a Social Story intervention versus treatment as usual. Outcome measures include acceptability to parents/teachers using qualitative interviews, and utility to parents, teachers and children using scores on the Strengths and Difficulties Questionnaires, the Developmental Behaviour Checklist and the General Health Questionnaire.

3. A proposal for a full clinical trial including justification of costs, outcomes, cost effectiveness and outcomes. From phase II, we will gather information regarding parameters and outcomes which would be relevant for a future full scale trial.

4.0 Ethical arrangements

4.1 Anticipated Risks and Benefits

We do not anticipate that trial participants will be subject to any risks during this study. Social Stories are widely used and focus on positive social and situational coping, therefore are unlikely to cause participants harm. A possible ethical issue with the study is that the control group will not receive the Social Story intervention during the trial. We have combated this by holding a second free workshop for those parents within the control group at the end of the study on the use of Social Stories.

It is possible that parents may become distressed when talking about their child's ASD at the focus group. Their clinicians involved in the training are very experienced in dealing with distressed parents and are well placed to either provide support themselves or to refer to other support as necessary. Similarly should a child become distressed the teachers, clinicians and researchers will have discussed prior to the intervention any necessary actions, distractions, activities or support that will be employed in the event of distress. Again, staff are experienced in these situations.

4.2 Informing potential trial participants of possible benefits and known risks

Participants will be sent information leaflets by post. Information sheets will be provided for children and young people aged 4-10 and 11-15 aged and their parents. They will choose whether to make contact with a researcher by telephone. Therefore potential participants will always have a minimum of 24 hours to decide whether or not to take part. We will contact the participants by letter or telephone if any relevant information related to ASD or Social Stories becomes available during the study. All information leaflets and consent forms will be co developed by the research team and the PPI group to ensure acceptability amongst participants.

4.3 Obtaining informed consent from participants

If children or parents are interested in taking part in the study after they have read the information sheets they will be able to phone a researcher to make an appointment. They will meet the researcher where they have the opportunity to ask any questions they have relating to the research and consent to the study if they are happy to do so. Written consent will be obtained from parents and consent or assent from children and young people will be obtained where possible.

5.0 Service User Involvement

As part of the development of this detailed project description and in accordance with the INVOLVE guidelines (57) we have discussed the review and specific questions with a focus group made up of parents of children with ASD and teachers who use Social Stories to provide input on the study design. The group was asked to comment on various aspect of the proposed design of the study and the findings from the focus group were reported back to the research steering group and channelled into the development of the proposal.

The concept of a manualised Social Stories toolkit was received positively by the parents who made up this focus group. They highlighted the need for simplified, compact guidance on producing Social Stories which adhered to Carol Gray's model. In particular a common theme which ran through the focus group was the need for a clear, starting point from which to develop Social Stories once a challenging behaviour had been identified. They also felt that more guidance was necessary on the use of strategies for making sense of this behaviour and how to determine the focus of the Social Story, which may be different to working on the challenging behaviour as such. There was agreement between a number of parents involved in the focus group that the beneficial outcomes of using Social Stories were not limited to the specific context of the Story but were generalised to everyday situations. The focus group also drew attention to the role of collaboration in writing Social Stories between parents and teachers,

Some members from the focus group have been invited to attend the research steering group, where they will be able to inform the design, recruitment and dissemination phases of the research. We will conduct a second focus group of parents of children with ASD in the local area in phase 1 of the trial. Parents will be involved in the development of consent forms and information leaflets, advising on methods of recruitment and developing and implementing a dissemination strategy.

Parents will be recruited to the focus group from a variety of sources, such as the local ASD forum list and the ASCEND (Autism Spectrum Conditions - Enhancing Nurture and Development Group), a popular course provided in the York and Selby area for parents and carers of children with ASD (45). After phase 2 parents and teachers will be invited to offer comments in the design and acceptability of the research, as a main outcome measure, in order to inform a future large scale RCT of Social Stories versus treatment as usual for mainstream children with ASD exhibiting challenging behaviour.

The members of the research team have successfully used public-patient engagement strategies in their previous work, including funded systematic reviews. For example, applicant Gilbody used public-patient involvement as part of a HTA-funded review of screening methods for post-natal depression.

6.0 Research Governance

6.1 Data Protection/Confidentiality

Data is to be managed by the Research Assistant and only members of the research team will have access to personal data during the study. All participant information will be stored in accordance with the Data Protection Act 1998. Participant personal identifiable information will be stored in a locked filing cabinet. All participant data will be anonymised by allocating each participant with an ID number. Anonymised participant data will be saved on a password-protected secure computer drive which only members of the research team will have access to. Participant personal identifiable data will be stored in a separate location to anonymised participant data. All data will be maintained by the Research Co-ordinator. All consent forms will be filed in a locked cabinet that only the

research team will have access to. Data inputed into the computer system will first be wiped of any identifying markers such as names/initials of each child to ensure confidentiality. Personal data will be stored for less than three months after the study and we will store research data generated by the study for 10 years. Electronic data will be stored in a secure drive that only the research team has access to.

6.2 Research governance and the conduct of the trial

The trial will be conducted to protect the human rights and dignity of the participant as reflected in the 1996 version of the Helsinki Declaration. Patients will not receive any financial inducement to participate. In order to protect the trial participants we will following will apply:

- the trial has been designed to minimise pain, discomfort and fear
- the trial has been designed to minimise any foreseeable risk in relation to the treatments involved
- the explicit wishes of the participants will be respected including the right to withdraw from the trial at any time,
- the interest of the patient will prevail over those of science and society,
- provision will be made for indemnity by the investigator and sponsor,
- contact details for further information will be provided.

We will seek ethical approval through the NHS Research Ethics Committee guidance. No new pharmaceutical compounds are used in this trial and Clinical Trials Authorisation is not required.

6.3 Monitoring and adverse events

This study is non-CTIMP (Clinical Trial of an Investigational Medicinal Product) and is therefore not subject to any additional restrictions. Decisions regarding prescription of medications will made by the participant in conjunction with their GP; participation in the study will have no bearing on this process. If a participant asks a member of the ASSSIST study team for an opinion on medication issues, they will be strongly encouraged to seek advice from their GP.

We will follow good clinical practice in monitoring for adverse events during all patient encounters with trial participants. Parents/guardians will be provided with an on-call number to ring if they have any concerns. All such calls will be reported to the Trial Steering Committee. Any calls that may represent any serious adverse event or serious untoward incidents will trigger an end to the trial. Any concerns raised will be dealt with either by these members of the research team or will be referred onto other appropriate clinicians or services as per usual practice.

This study will record details of any Serious Adverse Events (SAEs) that are required to be reported to the Research Ethics Committee (REC) under the terms of the Standard Operating Procedures for RECs [49]. An SAE is defined as a **'related'*** and **'unexpected'**** untoward occurrence
that:
(a) Results in death;
(b) Is life threatening;
(c) Requires hospitalisation or prolongation of existing hospitalisation;
(d) Results in persistent or significant disability or incapacity;
(e) Consists of a congenital anomaly or birth defect; or

(f) Is otherwise considered medically significant by the investigator.
* **'related'** is defined as: resulting from the administration of any research procedures.
**** 'unexpected'** is defined as: a type of event not listed in the protocol as an expected occurrence.

In the context of the current study, an occurrence of the type listed in (a) to (f) above will be reported as an SAE only if:
- It is suspected to be related to an aspect of the research procedures (e.g. completion of follow-up questionnaires, participation in qualitative sub-studies, telephone contact).
or
- It is an unexpected occurrence.

The Trial Manager will inform the Chief Investigator (CI) and 2 members of the Trial Management Group (TMG) who will jointly decide if the event should be reported to the main REC as an SAE. Related and unexpected SAEs will be reported to the main REC within 15 days of the CI becoming aware of the event. A SAE Form will be completed and a copy stored in the participant's records. The occurrence of adverse events during the trial will be monitored by an independent Data Monitoring Ethics Committee (DMEC) and the Trial Steering Committee (TSC). The DMEC/TSC will immediately see all SAEs thought to be treatment related and they will see SAEs not thought to be treatment related by the Trial Management Group at the next scheduled meeting.

The study will be stopped prematurely if:

1. Funding source stopped prematurely (and unable to replace it)

2. As stated, any reported Serious Adverse Events or Suspected Unexpected Serious Adverse Reaction or serious breach of the protocol will be urgently discussed with the data monitoring group and the Trial Steering Group and/or the Sponsor. Where the study was deemed to be a risk to any participant or needs to be altered by means of an amendment to the protocol (e.g. exclusion criteria) the study will be halted.

6.4 Trial Management

North Yorkshire and York will act a sponsor for this study.

Caroline Mozley
North and East Yorkshire alliance R&D Unit
Learning and Research Centre
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York
YO31 8HE

6.5 Indemnity
Normal NHS Indemnity procedures will apply.

6.6 Funding
Research funding has been secured from the National Institute of Health Research – Health Technology Assessment programme (reference: 09/169/07).

6.7 Trial Steering Committee (TSC)
A TSC will be set up and will include an independent chair and at least two other independent members, along with the lead investigator and the other study collaborators.

Two service users will join the steering group for collaboration on monitoring study progress. They will also be consulted on methods of dissemination of study findings for service users. They will meet at least annually (See Appendix 2 for details).

6.8 Data Monitoring and Ethics Committee (DMEC)
A DMEC committee will be set up and will comprise of an independent statistician and clinician (primary care physician and mental health professional). The role of the DMEC is to immediately see all SAEs thought to be treatment related and unexpected; they will also review outcome data. They will meet at least annually. (See Appendix 2 for details).

6.9 Recruiting centres
One centre (York) will be co-ordinating the recruitment of participants to the study

6.10 Day to day management of the trial
The chief investigator (Barry Wright) will be in charge of the overall management of the trial. The York-based trial manager (TBA) will be responsible for the co-ordination of the study between sites. A trial coordinator and trial secretary will carry out the day to day activities involved in running the trial. Delivery of training on social stories will be carried out by a team of clinicians. A research fellow will be responsible for the qualitative components of the study.

6.11 Responsibilities of the applicants
Barry Wright will act as the Chief Investigator with overall responsibility for the study and also act as the study mental health specialist. Barry Wright and members of the trial steering group will be responsible for study oversight, ensuring study milestones are met and adverse events are appropriately dealt with. **Simon Gilbody** and **Rachael Richardson** will provide advice and guidance on conducting the systematic reviews and trial management, and **Dean McMillan** will lead on the systematic reviews and provide support on the content and delivery of psychosocial interventions. Systematic searching and support will also be provided by **Julie Glanville**.

Victoria Algar will be the lead trial statistician. **Shezhad Ali** will lead the economic evaluation. **David Torgerson** will provide methodological advice about trial design and contribute to the economic evaluation. **Joy Adamson** will lead the qualitative components of the study. Study finances will be managed by the project manager and research co-ordinator in collaboration with the local research finance manager and overseen by the Trust Finance Officer.

Expertise on Social Stories will be provided by **Carol Gray** and **Anastasia Kokina,** with expertise on autism spectrum disorders provided by **Christine Williams**. Expertise in mainstream school education and conducting research within schools will be provided by **Steven Grigg**. **Richard Mills, Joanne Whitehead** and **Anne McLaren** will provide a service user perspective to inform the running of the study throughout the three stages. **Danielle Moore** and **Liz Littlewood** will support with day to day running of research

6.12 Dissemination of research findings
The research team has a strong track record of successful dissemination of work funded by the NIHR and other funding bodies, including the dissemination of systematic reviews. We will build on this track record for the current review. In recognition of the importance of successful dissemination, we have included in our research team applicant Richardson, who has specific expertise in the planning and implementation of dissemination strategies for systematic reviews.

We will begin to consider our dissemination strategy at an early stage of the project. As a basic step, we will publish the results of each of the phases of our study in high profile mainstream and specialist science journals, such as the British Journal of Psychiatry, the Journal of Child Psychology and Psychiatry, Clinical Child Psychology and Psychiatry and Cognitive Behaviour Therapy. Publications with high readership amongst clinical staff will be targeted such as the British Journal of Psychiatry; Journal of Child Psychology; Psychiatry; Clinical Child Psychology and Psychiatry; and the Journal of Autism and Developmental Disorders.

Presentations of study findings will be taken to relevant research conferences, local research symposiums and seminars for CAMHS professionals. In addition, the National Autistic Society and members of service user groups such as ASCEND will be consulted in the development of methods and dissemination which will be effective in reaching families of children with ASD. Additionally we will produce a short summary of the results that can be distributed to all trial participants, including patients and GPs, as well as relevant patient and other interest groups. Finally, we will aim to ensure coverage of our findings in the wider media by issuing a press release

We recognise that successful dissemination requires a pre-planned strategy that considers the groups who need to be aware of the results of the review and the methods with which to communicate with these groups. Our stakeholder group will be ideally placed to help us identify key factors that will be important in our dissemination strategy. These will include characteristics of the audience to be targeted, appropriate communication channels and the wider working environment of our audiences.

We are aware that this is a complex project likely to generate complex results and that planned dissemination will be crucial. We will seek separate funds to hold a research dissemination event for national and local clinicians and policy makers.

References

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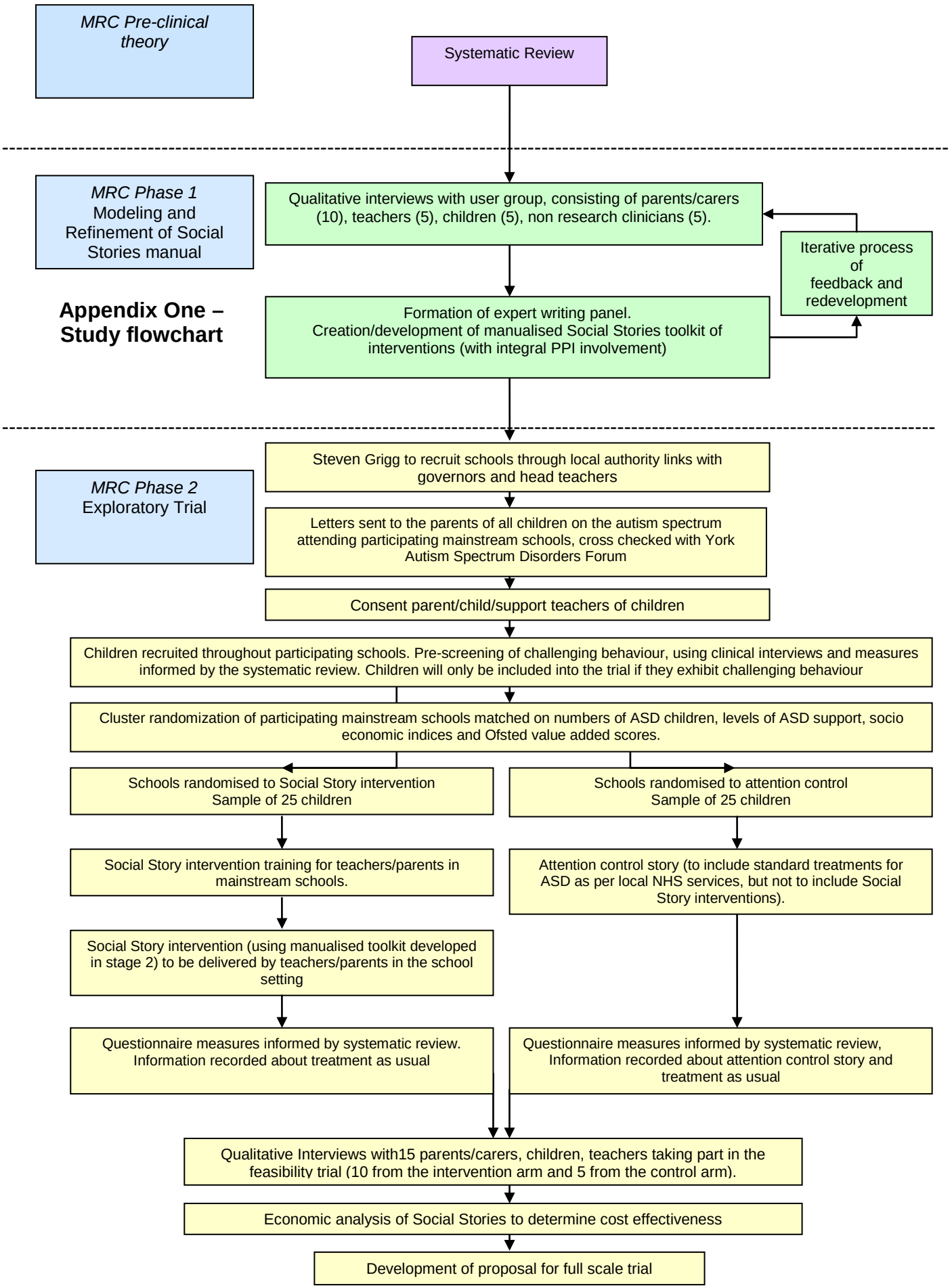
MRC Pre-clinical theory
Systematic Review
MRC Phase 1 Modeling and Refinement of Social Stories manual
Qualitative interviews with user group, consisting of parents/carers (10), teachers (5), children (5), non research clinicians (5).
Iterative process of feedback and redevelopment
Appendix One – Study flowchart
Formation of expert writing panel. Creation/development of manualised Social Stories toolkit of interventions (with integral PPI involvement)
MRC Phase 2 Exploratory Trial
Steven Grigg to recruit schools through local authority links with governors and head teachers
Letters sent to the parents of all children on the autism spectrum attending participating mainstream schools, cross checked with York Autism Spectrum Disorders Forum
Consent parent/child/support teachers of children
Children recruited throughout participating schools. Pre-screening of challenging behaviour, using clinical interviews and measures informed by the systematic review. Children will only be included into the trial if they exhibit challenging behaviour
Cluster randomization of participating mainstream schools matched on numbers of ASD children, levels of ASD support, socio economic indices and Ofsted value added scores.
Schools randomised to Social Story intervention Sample of 25 children
Schools randomised to attention control Sample of 25 children
Social Story intervention training for teachers/parents in mainstream schools.
Attention control story (to include standard treatments for ASD as per local NHS services, but not to include Social Story interventions).
Social Story intervention (using manualised toolkit developed in stage 2) to be delivered by teachers/parents in the school setting
Questionnaire measures informed by systematic review. Information recorded about treatment as usual
Questionnaire measures informed by systematic review, Information recorded about attention control story and treatment as usual
Qualitative Interviews with15 parents/carers, children, teachers taking part in the feasibility trial (10 from the intervention arm and 5 from the control arm).
Economic analysis of Social Stories to determine cost effectiveness
Development of proposal for full scale trial

Appendix Two – Trial Steering Group and DMEC Members

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Appendix Three – Study Timeline