Developing an Understanding of Family Engagement in Positive Behaviour Support

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June 2018

Research submitted in partial fulfillment of the requirements for the degree of Doctor in Clinical Psychology (DClinPsy), Royal Holloway, University of London

Acknowledgements

I would like to thank my supervisors Dr Kate Theodore, Dr Susan Cottam and Dr Alex Fowke for the invaluable support and guidance they have given me throughout this process. I would also like to thank Camden MOSIAC, the Hillingdon CAMHS LD team, and the Ealing Intensive Therapeutic and Short Breaks Service who helped with recruitment and all of the participants who gave up their valuable time to take part in this research.

I am immensely grateful for all of the support my family and friends have given me over the last few years, including the amazing people in my cohort. Finally a special thanks to my husband and to Piper both of whom I couldn’t have done this without.

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Executive Summary

The purpose of this thesis was to conduct a systematic review of the literature related to Positive Behaviour Support (PBS) delivered in family settings as well as to conduct an empirical study into the factors that family members find helpful and hindering in terms of their participation in PBS. The thesis is divided into three major chapters; 1) the systematic review, 2) the empirical article, and 3) an integration, impact and dissemination section which aimed to synthesise the findings of the two studies, provide critical reflection on how the studies were conducted and consider potential impacts and means of disseminating the results.

Background Information

People with intellectual disabilities and Autism Spectrum Disorder (ASD) are at an increased risk of displaying challenging behaviour. Challenging behaviour is a term used frequently in the literature to refer to behaviours which put the person's or other's safety or quality of life at risk and lead to responses that are aversive or restrictive. The definition recognises that it is the responses to the behaviour which determine whether it is challenging or not, rather than the behaviour itself, which serves a purpose for the person and arises from a mismatch between their needs and their environment. Challenging behaviour has a significant impact on not only the person but also those who support them.

PBS is currently considered to be best practice for managing challenging behaviour in people with intellectual or developmental disabilities. Rather than being a single intervention, PBS is a multicomponent framework that uses in-depth functional assessment to develop an understanding of why the behaviour occurs. This then forms the basis for the development and implementation of a comprehensive set of interventions that fit with the values of the client and their support network. One of the key principles of PBS is that all people in the person's support system are involved. For young people living at home, their family members play a key role in assessment and intervention due to their in-depth knowledge of the person and their ability to impact on behaviours.

Systematic Review

Aim

Much of the evidence base for PBS is based on studies conducted in American educational settings. Although there have been two systematic reviews of PBS used in community settings there has been no previous systematic review of the literature related specifically to implementing PBS in family-based settings. The aim of the current systematic review was to fill this gap in the literature and to consider the evidence for the effectiveness of PBS in managing challenging behaviour in family contexts.

Method

A systematic search of the PsychInfo and PubMed databases was conducted. Additional articles were also found through searches of reference lists and through referral from other professionals. Articles were screened against the inclusion and exclusion criteria, which were developed to include considerations of population, intervention, comparator and outcome. Broadly, studies were included if the recipient was a person with an intellectual disability or ASD who was receiving PBS with the aim of reducing challenging behaviour or increasing alternative appropriate behaviours and family members or other non-professional carers were involved in the intervention.

In addition to considering the outcomes, all included studies were also evaluated in terms of their methodological quality. This was done using a standardised methodological evaluation tool which was adapted to allow for evaluation of single-case design studies.

Results and Discussion

Eighteen studies were identified. Due to a limited number of studies reporting effect sizes a decision was made to evaluate outcomes based on the significance of the results. For single-case studies where results were graphical representations of behavioural observations a method for determining significance called percentage of non-overlapping data (PND) was used. All studies found some evidence for the effectiveness of PBS in family contexts, although the results were limited to improvements in only one type of behaviour in one service evaluation study. Other outcomes such as improvement in quality of life were also found.

The methodological quality of the included studies was generally poor, with thirteen studies being evaluated as having low methodological quality and only one as high. This was partly due to the prevalence of single-case study designs which tended to score lower on factors such as independent/blind raters, representativeness of participants and control of confounds. Although traditionally seen as a weaker form of evidence, there is a growing realisation that single-case study designs can be a useful form of evidence, particularly when factors such as these are addressed. Areas of strength for the single-case studies included in this review were the use of experimental designs, good inter-rater reliability for behavioural observations and the inclusion follow-up and generalisation phases.

Strengths and Limitations

A strength of the systematic literature review was that it clearly detailed the search strategy in line with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. It also included second reviewer checks for inclusion and exclusion criteria and ratings of methodological quality. Limitations of the review include the possibility of publication bias as only articles published in peer-reviewed journals were included. Additionally, given the variability in the way PBS is defined and described in the literature, it is possible that the search terms were not broad enough to ensure that all studies with interventions consistent with PBS were identified.

A further limitation is the generalisability of the results. A decision was made to exclude participants with primarily physical disabilities due to the likelihood that the nature of challenging behaviour may differ between these groups.

Empirical Article

Rationale

The systematic review showed that PBS delivered in family contexts can be effective. Given that one of the key principles of PBS is that it requires the involvement of everyone in the person's support system there is a good recognition in the literature of the need to consider how best to work well with and engage family members in intervention. Despite this, research in this area is limited and largely based on professional opinions. No study has asked family members specifically what their experiences of PBS were.

One of the findings of the systematic review was that the majority of the studies included required participants to be committed to engaging with the intervention, meaning that participants tended to be those who were more highly motivated and had sufficient time and/or family resources to participate. It is possible that this, combined with a lack of reporting of attrition rates, is partially responsible for the limited focus on understanding facilitators and barriers to engagement in PBS in the literature to date. The aims of the empirical study were therefore to address this gap and to explore family member experiences of PBS and what factors that they found helpful and hindering in terms of their participation.

Method

Six family members were interviewed regarding their experiences of PBS. Participants were recruited through two NHS services and were considered to have met the inclusion criteria if they were the family member of a young person with an intellectual or developmental disability who displayed challenging behaviour and had received PBS. Participants were excluded if they were under eighteen, unable to speak sufficient English to participate in the interview or were unable to consent.

Family members participated in a 60-90 minute semi-structured interview regarding their experiences of PBS. The interviews were then transcribed and a thematic analysis was conducted in order to identify and develop a thorough understanding of common patterns in their experiences.

Results

Thematic analysis resulted in five superordinate themes being identified; 1. *PBS is more than just strategies*; 2. *Considering the family context*; 3. *The therapist/family relationship*; 4. *Acknowledging challenges and the ongoing nature of the problem* and

5. *Supporting family member change*. Within these superordinate themes, thirteen subordinate themes were also identified. The majority of the findings were consistent with the literature related to PBS and parent interventions in general such as the importance of matching the intervention to the family member's priorities, strengths and resources; therapists who are knowledgeable and sensitive; developing an understanding of the function of the behaviour; and working with wider systems. Some novel results were also found such as family members not necessarily being confident that all strategies will work but finding it helpful when this is addressed and planned for from the beginning. Another important finding was the value some family members placed on having their own emotional needs addressed.

Strengths and Limitations

Qualitative research has been criticised for being more subjective than quantitative methods placing questions on transferability. Given that the aim of the study was to develop themes which could lead to practical recommendations, care was taken to detail the steps taken in analysis and to put in place a number of controls for methodological quality. This included having coding and results reviewed by an independent researcher as well as having the results reviewed by one of the participants.

A significant limitation of the empirical study relates to the size of the sample and the transferability of the results. All of the family members described their experiences of PBS as generally being positive and all reported some improvement in their child's behaviour. Despite this, some family members spoke of not being able to implement specific strategies and one of a regression in their child's behaviour, and all families were able to speak about some of the challenges they had faced.

Conclusions

Despite its limitations, the results of the systematic review suggest that PBS can be effective in managing challenging behaviours in family contexts supporting the need for further research into understanding what factors may play a role in its effectiveness, such as family member engagement, which has largely been neglected in the literature.

The empirical article aimed to gain a better understanding of family members' experiences and five superordinate themes were identified as being important to their engagement in PBS. Although these results should be considered tentative, they give valuable insight into the experiences of family members which can be considered when designing services and interventions to ensure that they meet family member needs.

Impact and Dissemination

The systematic review conducted as part of this thesis is the first to synthesise the research literature related to outcomes in family-based PBS, whilst the empirical study was the first to ask family members about their experiences of receiving PBS. Although the results of both studies should be considered preliminary they have important implications for both research and clinical practice.

Some suggested future avenues for future research include larger scale studies into the effectiveness of PBS in family contexts; improving the methodological quality of PBS studies, particularly single-case design studies; research into understanding the barriers experienced by families who do not find PBS helpful or drop out of intervention; and research to address some of the factors family members identified as being important for their engagement (e.g. the impact of addressing family emotional well-being on engagement and outcomes).

There are also implications for clinical practice. The systematic review lends support to guidelines which recommend the use of PBS for managing challenging behaviour displayed by people with disabilities. The empirical article gives clinicians valuable insight into the experiences of family members of young people with challenging behaviour who receive PBS and the themes identified include practical suggestions for working with and engaging family members in the PBS process. It is therefore hoped that the current findings will play a role in the development of services which better meet the needs of family members.

The systematic review and empirical article will be submitted to a peer-review journal for publication and also presented at a conference. The choice of journal and conference will be impacted on by the target audience and willingness to accept systematic reviews and qualitative studies.

Additional means of disseminating the results beyond academic circles include presenting the findings to the services who supported recruitment and providing a lay summary of results to participants. A plain language version will also be developed to send to services in the UK who provide PBS.

Chapter 1

A Systematic Review of Positive Behaviour Support Delivered in Family Contexts

Abstract

Challenging behaviour in people with intellectual disabilities and Autism has a significant impact on the quality of life of both the person and those that support them. Current best practice recommends the use of Positive Behaviour Support (PBS) in managing challenging behaviour and its effectiveness has been demonstrated in previous systematic reviews of the literature. One of the key principles of PBS is that all those involved in the person's care are involved in assessment as well as the development and implementation of strategies. A significant limitation of previous research is that it has largely been conducted in institutional settings or with professional carers. Since the move to deinstitutionalisation, people with intellectual disabilities are increasingly remaining in the family home and it is also important to demonstrate that PBS is effective in this context. This review, therefore, aimed to systematically review the literature related to the effectiveness of PBS in family contexts. A systematic search of the PsychInfo and PubMed databases was conducted to identify studies investigating the effectiveness of PBS in family contexts. Additional articles were located through searches of reference lists and through referral from other professionals. Retrieved articles were assessed against the inclusion and exclusion criteria and eighteen studies were retained for the final review. All eighteen studies found some evidence of effectiveness for PBS delivered in family contexts. The methodological quality of the included studies, however, was generally low. Fourteen of the included studies utilised a single case design and strengths and limitations of this are discussed. Improvements in quality of life were also reported in studies that assessed this. This systematic review demonstrates that PBS can be effective in managing challenging behaviour in family contexts. Limitations of this review are discussed and recommendations for future research made.

Introduction

People with intellectual disabilities and Autism Spectrum Disorder (ASD) are at an increased risk of using challenging behaviour, with prevalence estimates ranging from 5–15% for those in health, education or social services (National Institute for Health and Care Excellence [NICE], 2015). Challenging behaviour has been defined as behaviour which:

> is of such an intensity, frequency, or duration as to threaten the quality of life and/or the physical safety of the individual or others and it is likely to lead to responses that are restrictive, aversive or result in exclusion (Royal College of Psychiatrists, British Psychological Society, & Royal College of Speech and Language Therapists, 2007, p.88).

By definition, therefore, challenging behaviour has a significant impact not only on the quality of life of the individual but also those that support them.

Early approaches to behaviour intervention were primarily based on behavioural principles focussing on individual interventions and using reward and/or aversive response strategies (LaVigna & Donellan, 1986; Dunlap, Carr, Horner, Zarcone, & Schwartz, 2008). Results for these approaches were mixed and did not meet carers’ and families' needs (Carr et al., 1999). In interviews with parents, Turnbull and Ruef (1996) found that they wanted services which helped them to understand why the behaviour occurs, strategies which are practical and applicable in the home, a multi-area focus and to be included in the process. A later study by Griffith and Hastings (2014) identified interventions which focus on quality of life rather than just behaviour as a being a priority for parents.

The development of Positive Behaviour Support

Positive Behaviour Support (PBS) emerged in large part from Applied Behaviour Analysis (ABA) but also from the rise in systemic thinking and the person-centred and inclusion movements (Carr et al., 2002). ABA was developed by Donald Baer, Montrose Wolf and their colleagues in the early 1960s (Baer, Wolf, & Risley, 1968). It is based on Skinner's (1938) model of operant conditioning with assessment and interventions being based on principles of; reinforcement; contingency management; shaping, which involves gradually changing or teaching a new behaviour by providing stepwise reinforcement of small behaviour changes, which move closer in nature to the desired behaviour; and fading, which involves a gradual reduction of the reinforcement used to elicit a particular behaviour. Key to this is the concept of stimulus control, which involves the manipulation of behaviour by either the presence or absence of a specific triggering stimulus (Skinner, 1938; Wolf, Risley, & Hees, 1963). PBS shares a number of similarities with ABA including the fact that they are both empirically-based approaches using functional analysis. PBS, however, also draws on a range of other concepts such as the importance of system change, quality of life factors (as both contributor and outcome), social validity, social role valorisation or support to achieve valued social roles, the influence of culture, and consideration of the role of broader contexts (Dunlap et al., 2008).

Rather than being a single intervention, PBS is a comprehensive set of individualised interventions or strategies which are based on the values of the client and those in their support system (Gore et al., 2013; La Vigna & Willis, 2005). This consideration of a person's values directs the intervention in terms of identifying areas for change, leading to more individualised and meaningful goals. For example, if a person values acceptance and connection with others, this may lead to more specific goals such as joining a community group. This is important because, rather than simply reducing challenging behaviour, PBS is focused on improving quality of life (Carr et al., 1999; Gore et al., 2013; La Vigna & Willis, 2005). It does this through skill development and system and environment change with strategies designed to be implemented at multiple levels and throughout the entirety of the person's support network (Carr et al., 1999; Dunlap et al., 2008, Kinkaid et al., 2016). This, therefore, meets the parent needs identified in the Turnbull and Ruef (1996) and Griffith and Hasting (2013) studies mentioned previously.

How is PBS hypothesised to work?

The conceptual model upon which PBS is built sees challenging behaviour as a learnt behaviour, which serves a purpose for the person (Gore et al. 2013). The challenging behaviour is viewed as a person's attempts to get their needs met in the best way they can, using the skills and abilities they have, within the limitations and constraints of their environments (Carr et al., 2002; Gore et al., 2013). It is suggested that a person's behaviour is both influenced by, and influences their environment and the people in it (Franklin, 1980; Gore et al., 2013).

As PBS is a highly individualised, multicomponent intervention, which is not based on a single therapeutic approach, treatment, or philosophy, the specific mechanisms by which each intervention is hypothesised to work can vary from person to person (Gore et al., 2013). Broadly speaking, however, it centres around two primary elements, educational methods and system change (Carr et al., 2002). Educational methods involve the teaching of new skills which render the challenging behaviour unnecessary or increase a person's coping skills or self-control, for example, by teaching communication skills that facilitate a person being able to get their needs met.

The second component, system change, then aims to create opportunities for these positive behaviours to be displayed, for example, by ensuring that the person has access to appropriate communication aids in all settings and that the people interacting with them are familiar with and encourage their use (Carr et al., 2002).

Central to these methods is ensuring that the focus is on improving quality of life, which is not just seen as a desired outcome of PBS, but also as an intervention in itself (Carr et al., 1999; Gore et al., 2013). It is hypothesised that those who experience a higher quality of life are less likely to engage in challenging behaviour (Gore at al., 2013).

Main elements of PBS

La Vigna and Willis (2005) described a multi-element model of PBS which includes, first and foremost, a comprehensive functional assessment in order to develop an understanding of why the behaviour occurs. This then leads to the development of an individualised PBS plan containing multiple interventions or strategies which aim to make the challenging behaviour unnecessary. Examples of interventions include; (a) Ecological strategies, which reduce or remove mismatches in the person’s needs and their environment, such as providing visual aids; (b) Positive programming strategies which involve teaching new skills, such as how to communicate specific needs; (c) Focussed support strategies to prevent the behaviour, such as avoiding triggering locations or reinforcing alternative positive behaviours; (d) Non-aversive reactive strategies which aim to reduce the severity or duration of a behaviour.

In 2013 Gore and colleagues developed an updated multicomponent framework for PBS, drawing on previous research and literature. Their definition of the process of providing PBS includes four key components, (a) That all decisions are “data-driven,” meaning that they are based on past research or data collected about the person PBS is being delivered to; (b) Functional assessment is the basis for developing an individualised intervention; (c) Interventions should be multi-component including both proactive strategies (those that aim to change behaviour) and reactive strategies (those that manage behaviour); (d) Guidance and support is given to those implementing strategies along with on-going monitoring and evaluation. They also identified one of the key values of PBS as being that all key stakeholders participate in every stage of the process.

Evidence for PBS

There is an extensive evidence base suggesting that PBS is effective. One of the most influential studies is a large-scale literature synthesis by Carr et al. (1999). This included 109 PBS studies with a total of 230 participants published between 1985 and 1996. They found that 51.6% of PBS based interventions were effective, with effectiveness defined as a 90% reduction in the target problem behaviour and 68% were effective when this was defined as an 80% reduction in problem behaviour. Only 9% of studies showed minimal or no improvement. In a meta-analysis using the data collected from this synthesis Marquis et al. (2000) found a large overall effect size for the effectiveness of PBS in reducing challenging behaviour. Carr et al. (1999) argued that other factors besides percentage reduction in problem behaviour are also important for determining the success of PBS, such as quality of life. Unfortunately, this was only reported for six out of the 230 participants.

A systematic review by La Vigna and Willis (2012) investigating PBS for severe challenging behaviour also found it to be effective in terms of reducing the frequency or severity of challenging behaviour in all 12 identified studies, although, this was only qualitatively described. A significant weakness in the PBS literature is the fact that the majority of the evidence is based on single-case and small *n* studies, and there is still a need for larger-scale controlled research.

A further limitation of the literature is that the majority of studies have been conducted in institutional settings, meaning that carers are likely to be paid professionals. Many adults and children with intellectual disabilities and ASD are, however, being cared for in the family home. This is important as one of the key principles of PBS is that all those involved in the support and care of the person should be involved in the assessment, development of strategies and implementation. It would also appear that outcomes for PBS are dependent on how well this is done. Hieneman and Dunlap (2000) in a qualitative study investigating factors that affect outcomes in PBS found that six of the 12 factors related directly to caregivers. The Carr et al. (1999) review found that when people already in the person’s support system, such as parents and teachers, were the primary implementers of interventions, rather than behaviour intervention professionals, 61% of interventions were successful compared to 44.3%, with the Marquis et al. (2000) meta-analysis confirming that there was a significant difference in effect sizes. They also found that 55.2% of interventions were successful when they included strategies that required carers to make changes, compared to 41.8% when they did not (Carr et al., 1999), although the difference in effect size was not significant (Marquis et al., 2000).

Although the reviews by Carr et al. (1999) and La Vigna and Willis (2012) both included studies with family carers, these were very much in the minority. Given that the nature of the relationship between professional and family carers is likely to be very different this may affect the way in which interventions are developed and implemented and ultimately outcomes.

Rationale for Review

PBS is recommended as best practice by the Royal College of Psychiatrists et al. (2007) and is increasingly becoming the approach of choice when it comes to supporting people with intellectual disabilities or ASD who display challenging behaviour. One of the key principles of PBS is that all those in the person's support system should be involved in all stages of the intervention and outcomes have been found to be dependent on carer involvement. Although the evidence base supports the effectiveness of PBS the majority of this research has been conducted with professional carers and it is therefore not clear whether these results extend to family and non-professional carers.

To date, there has been no systematic review specifically looking at the effectiveness of PBS in family contexts. Although two previous systematic reviews (Carr et al., 1999; La Vigna & Willis, 2012) exploring the effectiveness of PBS in both community and institutional settings have been conducted, the majority of interventions were implemented by professional carers. In the Carr et al. (1999) review less than 35% of the included interventions were conducted in community settings, which in addition to family contexts, also included schools and group homes. In the La Vigna and Willis (2012) review, all but one study involved participants who lived in institutional or group home settings. Neither study explored the impact of implementing PBS with professional carers compared to family and non-professional carers as part of their results.

Objectives

The primary objective of this systematic review is to review the literature around the effectiveness of PBS when it is used in family contexts to manage challenging behaviour in people with intellectual disabilities or ASD. In this instance, effectiveness will be defined as whether PBS is able to be used to reduce challenging behaviour, or increase appropriate alternative behaviours.

Secondary objectives include exploration of the following questions:

1. Are any specific forms of PBS are more effective than others in in terms of reducing challenging behaviour?
2. Does PBS lead to improvements in quality of life?
3. Is PBS is an intervention which is acceptable to families?
4. What is the quality of the evidence base in this area and what are the implications for future research?

Method

Study Eligibility Criteria

Population

Participants were children and adults with an intellectual disability or an ASD. For the purpose of this review, the definition of intellectual disability and ASD are consistent with the criteria outlined in the DSM V. That is, intellectual disability is defined as limitations in both cognitive functioning with an IQ below 70, and adaptive behaviour, including daily living or social skills. ASD is defined as having social communication and social interaction deficits across multiple contexts. It was not necessary for the study to have formalised or detailed *how* the diagnosis of intellectual disability or ASD was established. A decision was made to exclude people with a primarily physical disability or acquired brain injury as it is likely that the nature and function of challenging behaviours may differ between these groups.

Participants had to display challenging or problematic behaviour that was described in the study as impacting negatively on themselves or their family. For this study, the definition of challenging behaviour was that agreed by the Royal College of Psychiatrists et al. (2007) given earlier.

In this review the definition of 'family members' was kept broad, extending beyond parents, and included anyone who provided everyday care to the person in a non-professional capacity (i.e. grandparents, foster parents) and excluding paid or professional support workers or carers.

Intervention

The intervention did not need to state specifically that it was PBS based in order to be included in this review but the intervention delivered needed to fit within the PBS framework. If the study did not state that it was PBS the intervention must have involved the following components as a minimum: a functional assessment, a multi-component behaviour plan, and system/family involvement. Studies were excluded when training on the PBS model only was delivered and there was no direct intervention.

Interventions combining PBS with other approaches (e.g. CBT) were included as long as the PBS component was substantial and clearly evident as PBS (i.e. included functional assessment, individualised multicomponent plan, and system involvement). Studies comparing different forms of PBS (e.g. PBS with or without CBT) were included as long as there were pre- and post-intervention measures.

Comparator: study selection

This review included experimental studies attempting to evaluate the effectiveness of PBS when used in family contexts. Due to limited studies utilising a randomised control trial (RCT) or control group design all intervention studies which included and compared baseline and post intervention data were included, including single-case design studies.

Outcomes

The primary outcome measures for this review were levels of challenging behaviour or incompatible adaptive behaviours. These could be measured using either standardised measures or scored behavioural observations of the frequency, severity or proportion of the target behaviour.

Other outcomes considered important and assessed where available included quality of life, quality of family interactions and social validity of the intervention.

Exclusion Criteria

Studies where the main focus was on an intervention other than PBS or which included primarily professional carers were excluded. This review is limited to published studies only. Additionally, due to resource constraints, only studies published in English were considered.

Search Strategy

Figure 1 illustrates the process used to select studies for this review. The PsychInfo and PubMed databases were searched on the 4/9/2017 and 11/9/2017 respectively. Title, keyword, and abstracts were searched, using the following terms;

1. “Positive Behaviour Support” or “Positive Behavior Support.”

An additional search of the same databases on the 9/10/2017 and 16/10/2017 using the following terms was also conducted;

1. “Family” OR “community” OR “parent” OR “home” AND
2. “Behaviour support” OR “behavior support” OR “behaviour intervention” OR “behavior intervention” OR “behaviour management” OR “behavior management” AND
3. "Challenging behavior" OR "challenging behaviour" OR "aggression" OR "aggressive behaviour" OR "violent behaviour" OR "aggressive behavior" OR "violent behaviour" OR “violence” OR "self-injury" OR “self-harm.”

Additional published studies were also identified by searching the reference lists of retrieved studies and through referrals by professionals working in the field.

Data extraction

The researcher screened all papers retrieved in the search by title and abstract for any studies that potentially might be relevant. Full articles were retrieved for 71 studies which were then screened for eligibility using the inclusion criteria. Nine full-text articles were screened for eligibility by a second reviewer, a clinical psychologist who was also the research supervisor, in order to ensure that the inclusion criteria were able to be applied consistently. There was 100% agreement between the two reviewers. Eighteen articles were considered eligible for this review.

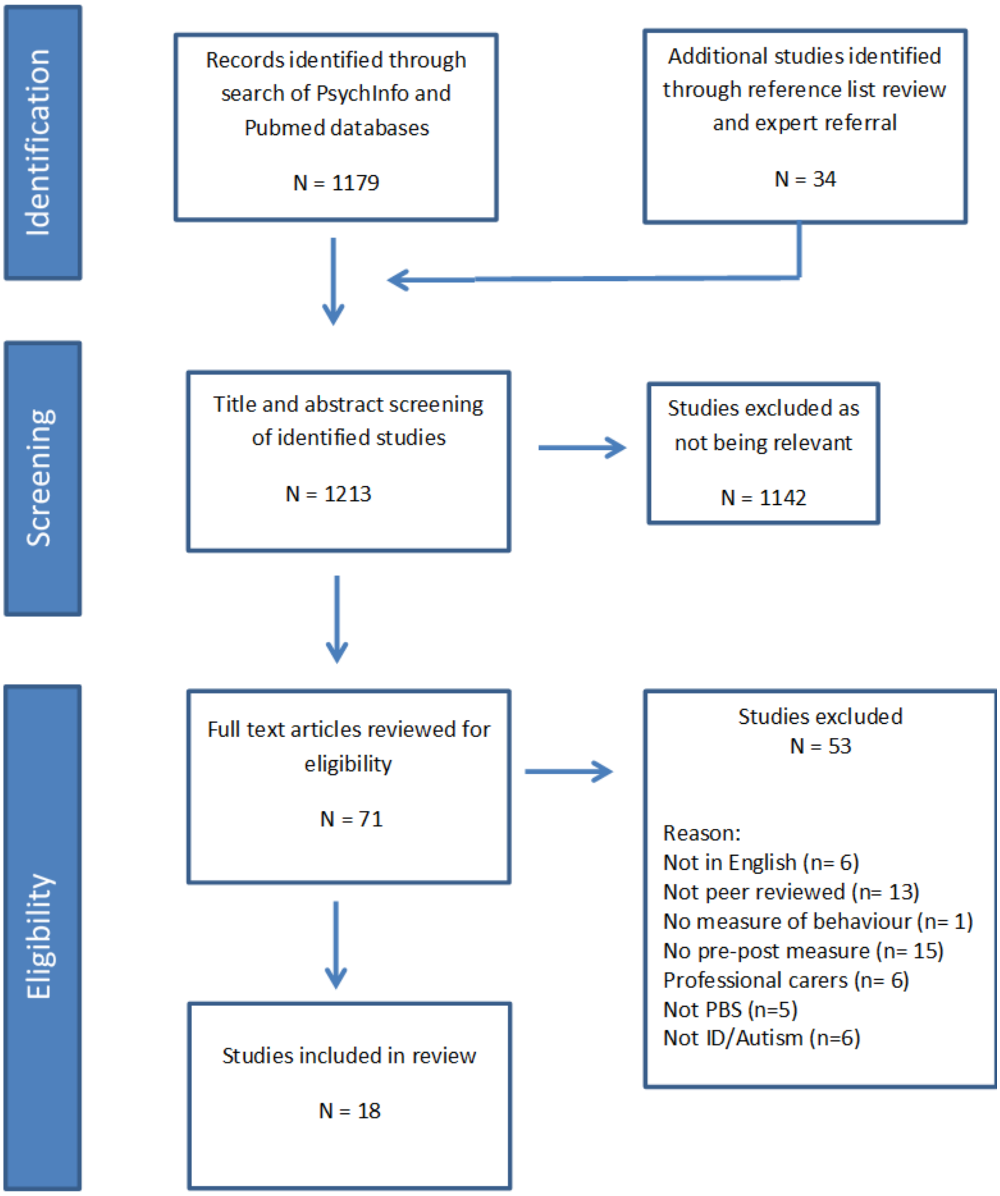

Figure 1. Systematic review search process flow-chart

The researcher then extracted relevant study information from each paper including study design, type of PBS intervention, population and sample size, recruitment procedures, attrition rates, blinding, outcome measures, results, consideration/controlling of confounder, follow-up, and generalisation, and intervention fidelity.

Appraisal of study methodological quality

An adapted version of the Quality Assessment Tool for Quantitative Studies (QA Tool; Effective Public Health Practice Project, 1998) was used to rate the methodological quality of the included studies (Appendix 1). The QA Tool was originally developed for use as a means of synthesising and rating information related to methodological quality in public health studies. It was intended to be used as a tool that would lead to high quality systematic reviews that would provide a strong evidence base for the public health sector (Thomas, Ciliska, Dobbins, & Micucci, 2004). Studies are rated as either ‘strong’, ‘moderate’ or ‘weak’ across six criteria: selection bias, study design, confounders, blinding, data collection methods and withdrawals and drop outs. An overall study rating of either strong, moderate or weak is then also given. In order to avoid different interpretations of these criteria, there is a QA Tool dictionary which provides direction as to how studies should be scored for each criteria. Thomas et al. (2004) found the QA Tool to be reliable and valid, and Deeks et al. (2003) found it to be one of six quality assessment tools suitable for use in systematic reviews.

Critical appraisal tools for research, including the QA tool used in this study, are not generally designed to consider the unique methodological challenges of single-case designs. Although traditionally seen as a weak form of evidence, Kazdin (2011) argues that when done correctly single case study designs can exert considerable control over sources of bias. Kazdin (2011) recommends the use of an experimental design, standardised observation rating, inter-rater agreement being derived through point to point agreement ratio as well as a measure of agreement on total, independence of raters and controlling for confounders as being necessary to reduce bias in single case study designs.

The QA tool was adapted to be able to better evaluate single-case design studies and incorporated recommendations found in Kazdin (2011) relating to study design and blinding. Specifically, single-case studies which utilised an experimental design (i.e. ABAB or multiple baseline) could be considered to have moderate quality. Blinding of raters was not necessary, although ratings done by an interventionist or the lead researcher scored as ‘weak’ on this item. Where behavioural observations were conducted raters had to have undergone training and achieved at least an 80% level of inter-rater reliability. For withdrawals and drop-outs, this was considered weak if there was no maintenance/follow-up period for single case studies.

The rating was done primarily by the researcher with a second reviewer co-rating four of the studies to control for bias. Inter-rater agreement was 83.33%. Areas of disagreement were discussed and an agreement reached. This tool was not used to further screen already included studies and no studies were excluded based on methodological quality, assuming that they met the inclusion criteria.

Data synthesis

Due to the level of heterogeneity in the studies relating to the type of PBS delivered, study design and outcomes measured a meta-analysis was not considered appropriate. Findings are therefore reported narratively.

Measures of treatment effect

For articles where statistical analysis was conducted and reported, treatment efficacy was determined by whether the results were statistically significant or statistically non-significant. This is due to the heterogeneity in the ways in which effect sizes were reported as well as the fact that only a limited number of studies reported effect size.

For single case studies, intervention effects were determined by calculating Percentage of Non-Overlapping Data (PND). There are several methods used for estimating effect size in single case design studies with PND, the percentage of data points exceeding the median (PEM) and Percentage of All Non-Overlapping Data (PAND), being the most common. PND is the percentage of data points in the intervention phase that are greater than (or lower than) the highest data point in the baseline phase. PEM is the number of data points in the intervention phase which are greater than the median data point in the baseline phase and PAND is the percentage of non-overlapping data points between the baseline and intervention phases. The reason for choosing PND in this instance is that it has been used more frequently in the literature and has also been found to be a more conservative and effective measure of effect size than PEM (Chen, Hyppa-Martin, Reichle, & Symons, 2016; Lenz, 2013). PAND is considered to be a more robust measure, although it is only appropriate when there are more than 20 data points, which was not the case for many of the studies included in this review (Lenz, 2013).

A PND greater than 0.70 is considered effective; 0.50-0.70 is considered to be marginal effectiveness and less than 0.50 is no observable effect (Scruggs & Mastropieri, 1998).

Results

The original literature searches produced 1,179 citations and a further 34 studies were identified through reference list reviews and from expert referrals. After title and abstract screening, 71 full-text articles were retrieved for further screening against the inclusion criteria. Eighteen studies were found to meet the above inclusion criteria. A flow diagram depicting the search process can be found in figure 1.

Characteristics of studies

A summary of the main study characteristics can be found in table 1. Fourteen of the included studies utilised a single case design. Other designs included one RCT which compared two different forms of PBS (Durand, Hieneman, Clarke, Wang, & Rinaldi, 2012), two service outcome evaluations (Inchley-Mort, Rantell, Wahlich, & Hassiotis, 2013; Reid, Scholl, & Gore, 2013) and a quasi-experimental longitudinal study (Lucyschyn et al., 2015).

The RCT was conducted by Durand et al. (2012) and investigated the effectiveness of Positive Family Intervention (PFI) compared to PBS alone. PFI is a manualised eight-week PBS-based program delivered to parents which also incorporates an adapted version of Seligman’s optimism training (1998). The protocol for sessions focussed on individual elements of the PBS process, such as gathering information, analysing it and developing a plan (functional analysis) as well as preventative strategies, consequences, and replacement behaviours, which would then be individualised in-session. Participants were 54 parents of children with a developmental disability who displayed challenging behaviour.

Table 1.

Summary of Main Study Characteristics.

Study	**Design**	**N**	**Intervention Type**	**Intervention Intensity**	**Intervention duration**	**Outcome Measures**	**Results**	**Follow Up**
Bailey & Blair (2015) USA	SCD - Non concurrent multiple baseline.	3	Prevent Teach Reinforce for families	FA plus 1 Training session + 1 per week for 2 weeks. Additional feedback sessions every week (10 – 15 min)	Varied	IBRST PTR Self-Evaluation: Social Validity form adapted from the TARF-R	Significant increase in AB for all participants (PND =0.89 - 1.00); significant decrease in PB for 2 participants (PND=1.00). Marginal decrease in PB for 1 (PND=0.69)	Maintenance but no follow-up
Durand et al.(2012) USA	RCT – Control group; PBS without optimism training.	54	Positive Family Intervention (PBS with optimism training)	90 min weekly sessions	8 weeks	SIB-R - GMI QRS-SF – Pessimism Scale. Behavioural Observations	Significant decrease for Pessimism, GMI and PB (p< .01) Significant interaction effect for GMI only (p< .01)	None
Sears et al. (2013) USA	SCD - Concurrent multiple baseline.	2	Prevent Teach Reinforce for families	FA plus 1 training session (additional as needed)	Varied	Behavioural Observations Social validity - TARF-R	Significant increase in AB for both (PND=0.71 - 0.83); Significant decrease in PB for both	2 weeks

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
							(PND=0.85-1.00	
Reid et al. (2013) UK	Service evaluation Pre – post.	11	Ealing Intensive Therapeutic and Short Break Service	Varied but intensive service	Varied (range 4 months to 2 years 11 months)	Developmental behaviour checklist Three concerns	DBC significantly decreased (p< .05, *r*= 0.44) Parents concerns significantly decreased (p< .01, r= 0.61)	None
Lucyshyn et al. (1997) USA	SCD - Multiple base line	1	Family based – comprehensive behaviour support	FA plus 1 – 3 sessions per week	66 sessions	Behavioural Observations Parent rated frequency of behaviour QOL - RLI Social Validity Contextual fit	Statistically significant decrease in PB for 2 out of 4 target routines, approaching significance for 1. (p< .01, p< .05, p=.051)	Maintenance plus follow-up at 3 and 9 months
Lucyshyn et	Longitudinal Quasi	12	Family Centred PBS targeting	FA plus average 1.2 sessions per	Varied (average =	Behavioural	PB significantly	Maintenance plus follow-up

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
al. (2015) Canada and USA	Experimental + SCD - multiple baseline		parent-child interactions	week)	76.6 sessions + 39 maintenance sessions)	Observations Social Validity measure Contextual fit measure	decreased (p< .01) AB significantly increased (p< .01)	(range from 3 – 24 months)
Vaughn et al. (2002) USA	SCD - Multiple baseline	1	Family centred functional assessment based intervention	FA + coaching in first 2 sessions	Not stated	Behavioural Observations	Significant decrease in PB for all 3 target routines (PND=0.75-1.00); Significant increase in AB for all 3 target routines (PND=0.75-1.00)	No follow-up
Dunlap & Fox (1999) USA	SCD - Multiple baseline	6	Individualized Support Project (ISP)	Unclear but FA plus demonstration and support over at least several days.	Varied	Behavioural observations BDI ABC Interviews	Significant decrease in PB for all 6 participants (PND=0.91-1.00)	No follow-up
Lucyschyn et al. (2007)	SCD – Longitudinal	1	Family Based Comprehensive	FA plus 1- 2 training sessions	23 weeks + 51 weeks	Behavioural Observations	Significant decrease in PB for all 4 target	Maintenance + follow up at 6,

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
USA	multiple baseline		Behaviour Support	per week.	maintenance support	QOL - RLI Social validity Contextual fit	routines (PND=0.71-1.00)	18, 36, 67, 86 months
Binnendyk & Lucyschyn (2008) Canada	SCD – Multiple probe	1	Family centred Positive Behaviour Support	FA plus intensive training 2-4 sessions per week, parent training 1-2 sessions per week	14 weeks intensive training, 8 weeks parent training.	Behavioural Observations Social Validity Contextual fit Family quality of life survey	Significant increase in total food acceptance and steps completed (PND=1.00); Marginal for self-initiated food acceptance (PND=0-60)	Follow-up at 1, 5 and 6 weeks as well as 26 months
Lee at al. (2007) USA	SCD - ABAB	1	Positive Behaviour Support	Unclear	Unclear	Self-monitoring of behaviours	Significant increase in AB before and after withdrawal (PND=0.86 and 1.00). No change in PB before and marginal after withdrawal (PND=0.14 and 0.50)	18 months

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
Blair et al. (2010) South Korea	SCD - Concurrent multiple baseline	1	Positive Behaviour Support – school and family collaboration	FA plus 10 hour initial training in PBS plus 1 coaching session and weekly/biweekly review meetings	Unclear	Behavioural observations: Social validity - adapted from TARF-R	Significant decrease in PB for all 3 participants (PND=1.00, 1.00, 1.00); Significant increase in AB for all 3 participants (PND=1.00, 1.00, 1.00)	3 weeks
Donellan et al. (1985) USA	SCD - ABC	16	Intensive Behaviour Intervention	Unclear however included both direct intervention and training mediators.	Varied 3 – 8 weeks	Behavioural observations: Consumer satisfaction - interview	Significant decrease in PB for 12 participants (PND=0.75-1.00); marginal effect for 1 (PND=.50); No decrease for 3 (PND=0.00-.40)	Follow-up however unclear when.
Inchley-Mort et al. (2014) UK	Service evaluation with matched control group	24	Outcome Evaluation of Complex Behaviour Service	varied	varied	ABC HoNOS-LD CANDID-s PASSAD	Significant reduction in following ABC domains: Irritability ($p < .05$) Stereotypy ($p < .05$)[1]	12 months

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
							Total Score (p = .05) No significant difference for other domains or HONOS	
Koegel et al. (1998) USA	SCD - Multiple Baseline	3	Functional assessment and parent-implemented intervention	Unclear and likely varied	Unclear and likely varied	Behavioural observations Social Validity.	Significant decrease in PB for 2 participants (PND=0.90 and 1.00); No impact for 3rd.	No follow-up
Barry & Singer (2001) USA	SCD - Non-concurrent multiple baseline	1	Functional assessment and clinician implemented PBS with family involvement	2-4 times per week	26 months	Behavioural observations	Significant decrease in PB for all 5 target routines (PND=1.00); Significant increase in AB for all 5 target routines(PND=0.91-1.00)	Follow-up at 1 month and between 3-4.5 months

Study	**Design**	**N**	**Intervention Type**	**Intervention Intensity**	**Intervention duration**	**Outcome Measures**	**Results**	**Follow Up**
Carr et al. (1999) USA	SCD – Longitudinal multiple baseline	2	Comprehensive Multi-situational Intervention	FA plus several days per week during intervention and monthly during maintenance	Intervention 2 – 3 years, maintenance 2 – 3 years.	Behavioural observations Social validity	Significant increase in AB for all 3 participants (PND=0.92-1.00); significant decrease in PB for all 3 participants (PND=1.00).	Maintenance only
							Significant effect for engagement for 1 participant (PND=1); Marginal effect for 1 (PND=0.55) and no effect for 1 (PND=0.08)	
Moes & Frea (2000) USA	SCD - ABC	1	Prescriptive vs Contextualised Behaviour intervention	1 X weekly	Prescriptive = 15 weeks Contextualised	Behavioural Observations Parent ratings of	Prescriptive: Significant decrease in PB (PND=0.93) and increase in 1 AB	Follow-up 3 months

Study	Design	N	Intervention Type	Intervention Intensity	Intervention duration	Outcome Measures	Results	Follow Up
					= 7	sustainability	(PND=1.00). No impact for 2nd AB. Contextualised: Significant decrease in PB (PND=1.00) and increase in both AB (PND=0.71, 1.00).	

1. Only Stereotypy maintained at follow-up.

2. Key: FA = Functional Assessment; PB = Problem behaviour; AB= Appropriate Behaviour; Individualized Behavior Rating Scale Tool (IBRST); Treatment Acceptability Rating Form–Revised (TARF-R; Reimers & Wacker, 1992); Scales of Independent Behavior-Revised, General Maladaptive Index (SIB-R GMI; Bruininks, Woodcock, weatherman & Hill, 1996); Questionnaire on Resources and Stress (QRS; Holroyd., 1982); Resident Lifestyle Inventory (RLI; Newton et al, 1987); Battelle Developmental Inventory (BDI; Newborg, Stock, & Wnek, 1984); Autism Behavior Checklist (ABC; Krug, Arick, & Almond, 1980); Health of the Nation Outcome Scales for People with Learning Disabilities (HoNOS–LD; Roy, Matthews, Clifford, Fowler & Martin, 2002); The Camberwell Assessment of Need for Adults with Developmental and Intellectual Disabilities Short Form (CANDID-S; Xenitidis, Slade, Thornicroft & Bouras); Psychiatric Assessment Schedule for Adults with Developmental Disability (PASS-ADD; Moss et al., 2002).

Two studies were outcome evaluations of services based on the PBS model (Inchley-Mort et al., 2014; Reid et al., 2013). The former paper compared 24 people with intellectual disabilities who received services from the Complex Behaviour Service, an enhanced behaviour support service in London, to a group of 22 matched controls. The specific intervention provided to each participant varied in content, length and intensity, although all interventions were based on the PBS model. The second evaluation study by Reid et al. (2013) evaluated outcomes for the Ealing Intensive Short Breaks service, based on the PBS model and incorporating system support, therapeutic interventions and respite services. Participants were 11 young people with intellectual disabilities who were at risk of residential placement. Again the specific intervention each participant received varied based on their needs, as did the intensity and duration.

Lucyschyn et al. (2015) conducted a longitudinal study using a quasi-experimental, pre-post design, to investigate family centred PBS in a group of twelve parent-child dyads. In addition to impact on behaviour they also looked at the impact of PBS on parent-child interactions.

Variations in PBS

The definition of PBS described previously allows for significant variation in how the intervention is delivered in terms of content, duration and intensity. Whilst this is reflected in some of the research, the vast majority of studies described include examples of comprehensive functional assessments followed by significant support for families to implement the strategies and follow-up sessions. There is a question as to how likely or cost effective this is in a real world setting and also if it is possible to assess this in larger-scale studies. The two evaluation studies (Inchley-Mort et al., 2014; Reid et al., 2013) included in this review are an important first step and appear to have been based on providing an intensive service, although specific details about each intervention are not available.

Alternate forms of PBS included in this review include PFI, which has already been described, and Prevent Teach Reinforce (PTR; Bailey & Blair, 2015; Sears, Blair, Iovannone, & Crosland, 2012). PTR is a manualised PBS based intervention which has previously been used in school contexts. PTR is a five-step model including teaming, goal setting, assessment, intervention, and evaluation; within the intervention stage there are also three components: prevent, teach, and reinforce. The first four steps were delivered over two extended sessions with the families.

Although not necessarily an alternate form of PBS, Moes and Frea (2000) provided PBS in both a prescriptive and contextualised format. The aim was to reduce challenging behaviour and increase compliance and task engagement of a three-year-old boy for home routines. In the prescriptive format, they matched intervention strategies to the function of the behaviour but did not contextualise it to the family situation. In the contextualised version they took into account family member needs and preferences.

Other differences in the way PBS was delivered included having an extended assessment stage (Carr et al., 1999) and investigating a school and home collaboration where both teachers and parents were taught the principles of PBS and supported to work as a team (Blair, Lee, Cho, & Dunlap, 2010). Dunlap and Fox (1999) looked at outcomes for six children involved in the Individualised Support Project (ISP). Although given a different name, ISP still follows the principles of PBS including collaborative functional assessment and the development of strategies which build skills and competence and are implemented in all areas of the child's life.

Participants

Overall there were 141 participants. The overwhelming majority were aged under eighteen (n=114) and male (n=119). 15 of the 18 studies had exclusively child participants, with child being defined as aged under 18; two studies included a mix of adult and child participants (Carr et al. 1999; Donellan, LaVigna, Zambito, & Thvedt, 1985), and the Inchley-Mort et al. study (2013) included only adult participants. The majority of studies were conducted in the United States or Canada (n=15), the two outcome evaluations (Inchley-Mort et al. 2014; Reid et al., 2013) were conducted in the United Kingdom and one single-case study was conducted in South Korea (Blair et al., 2010).

For the majority of the single-case design studies there was no information relating to where participants were referred from or about drop-outs or withdrawals. For studies which reported the source of referrals, this was usually from local services and doctors/ pediatricians. Three of the four non-single case design studies did report attrition/retention rates, with the exception of Reid et al. (2013). The highest rates of attrition were for the Durand et al. (2012) RCT, which defined a drop out as missing three consecutive sessions. The PFI group had a 33.33% attrition rate, whilst the PBS alone group had a 37.04% attrition rate. Lucyschyn et al. (2015) reported a retention rate of 83%. Inchley-Mort et al. (2014) did not directly report on attrition rates, however, there were five less participants in the post data than in the pre suggesting an 80% retention rate. Sears et al. (2013) reported that one of two families were unable to complete follow-up data due to a vacation. Donellen et al. (1985) reported that one of sixteen participants withdrew after nine weeks of follow-up. Lucyschyn et al (2007) in a single case study did report on a lack of follow-ups related to some outcomes which was due to scheduling difficulties and the family no longer valuing the routine. Koegel, Stiebel and Koegel (1998) also reported that one of here participants did not participate in follow-up due to moving away.

Procedural integrity

Five studies included a measure of implementation fidelity, either related to the clinicians or family member implementation (Bailey & Blair, 2015; Blair et al., 2010; Carr et al., 1999, Durand et al., 2012; Sears et al., 2012). In general levels of fidelity were high with all reported measures being greater than 80%. In regards to professional interventionists specifically, this provides a measure of intervention fidelity which adds to methodological quality. Possibly more useful, however, is the information related to how consistently families were able to implement interventions. It is worth noting that in two studies additional training was provided to parents when implementation fidelity dropped (Bailey & Blair, 2015; Sears et al., 2012). Although this would be considered good practice, there is a question as to the level of external validity as clinicians in real-world settings are unlikely to have access to ongoing video-recordings of families implementing strategies.

Methodological quality of studies

Using the adapted QA Tool only one study could be considered strong in terms of its methodological quality and that is the RCT conducted by Durand et al. (2012). Four studies could be considered moderate with the remaining studies scoring as weak (Dunlap & Fox., 1999; Inchley-Mort et al., 2013; Koegel et al., 1998; Lucyschyn et al., 2015). Full details of the scoring for methodological quality of the included studies can be found in table 2

Table 2.

Quality Assessment Tool Results.

QA Tool Assessment	***Bailey & Blair (2015)***	***Durand et al. (2012)***	***Sears et al. (2013)***	***Reid et al. (2013)***	***Lucyshyn et al. (1997)***	***Lucyshyn et al. (2015)***
Selection bias	Moderate	Moderate	Moderate	Moderate	Weak	Weak
Design	Moderate	Strong	Moderate	Weak	Moderate	Moderate
Confounders	Weak	Moderate	Moderate	Weak	Weak	Moderate
Blinding	Weak	Strong	Weak	Weak	Moderate	Moderate
Data collection	Moderate	Strong	Moderate	Moderate	Moderate	Moderate
Drop outs	Moderate	Moderate	Weak	Moderate	N/A	Strong
Overall	Weak	Strong	Weak	Weak	Weak	Moderate

QA Tool Assessment	***Vaughn et al. (2002)***	***Dunlap & Fox (1999)***	***Lucyschyn et al. (2007)***	***Bynnendyk & Lucyschyn (2008)***	***Lee at al. (2007)***	***Blair et al. (2011)***
Selection bias	Weak	Moderate	Weak	Weak	Weak	Moderate
Design	Moderate	Moderate	Moderate	Weak	Moderate	Moderate
Confounders	Weak	Weak	Weak	Weak	Weak	Weak
Blinding	Weak	Moderate	Moderate	Moderate	Weak	Weak
Data collection	Weak	Moderate	Moderate	Moderate	Weak	Moderate

Drop outs	N/A	Moderate	N/A	N/A	N/A	Moderate
Overall	Weak	Moderate	Weak	Weak	Weak	Weak

QA Tool Assessment	***Donellan et al. (1985)***	***Inchley-Mort et al. (2014)***	***Koegel et al. (1998)***	***Barry & Singer (2001)***	***Carr et al. (1999)***	***Moes & Frea (2000)***
Selection bias	Moderate	Moderate	Moderate	Moderate	Moderate	Moderate
Design	Weak	Moderate	Moderate	Moderate	Moderate	Moderate
Confounders	Weak	Moderate	Weak	Weak	Weak	Weak
Blinding	Weak	Weak	Moderate	Unclear	Moderate	Unclear
Data collection	Moderate	Strong	Moderate	Weak	Weak	Weak
Drop outs	Strong	Moderate	Moderate	Moderate	Moderate	Moderate
Overall	Weak	Moderate	Moderate	Weak	Weak	Weak

It is worth noting that although the Durand et al. (2012) RCT scored as strong methodologically, this was specifically related to considering PFI vs PBS. In the context of this review, the effectiveness of PBS in general is being considered and not specifically one form of PBS vs another. Of the two service evaluation studies, the Inchley-Mort (2014) study was found to be of moderate quality due to the use of a matched-control group and attempts to control for confounders in the analysis. The Reid et al. (2013) study, however, did not have these same controls and was scored as having a weak methodological quality. A strength of these two studies, not taken into account by the use of the assessment tool, is that they have increased external validity as they are both examples of implementing PBS based interventions in real-world settings.

Only two single-case design studies scored as moderate, those by Dunlap and Fox (1999) and Koegel et al. (1998). Areas of strength in the single case study designs reviewed here included study design and data collection. All but three of the single case studies utilised an experimental design such as a withdrawal (ABAB) or a multiple baseline design. These designs limit the chances that changes are due to a co-occurring external factor. Ideally, a concurrent multiple baseline is recommended, although this is not always possible due to nature of the behaviour either putting the person or others at risk or behaviours emerging later or subsequent participants being referred later. This was a factor raised in several of the studies included in this review (Donellan et al., 1985; Bailey & Blair, 2015). Furthermore, all but one study provided details as to how behavioural observations were standardised and rated and provided measures of inter-rater reliability. The exception was Lee, Poston and Poston (2007) which used a self-rating measure.

Three areas that the single case studies included in this review tended to score poorly on were participant representativeness, blinding and confounders. Participant representativeness has already been discussed and will therefore not be repeated. None of the single case study designs specifically mentioned controlling for confounders beyond the choice of study design. Blinding is traditionally considered an important aspect of ‘higher level’ and RCT research and whilst the nature single case research often precludes the participant from being blind it is possible for raters to be blind. Only one study in the current review reported using blind raters (Koegel et al., 1998). In practice, this is often challenging due to limited resources and research suggests that it does not have a significant impact on the ratings derived (Kent & Foster, 1977). In five of the studies included in this review, the person providing the intervention was also the primary rater (Bailey & Blair 2015; Blair et al., 2010; Donellan et al., 1985; Sears et al., 2012; Vaughn, Wilson & Dunlap, 2002), with the second reviewer being involved in delivering intervention in a further three studies (Bynnendyk & Lucyschyn, 2008; Lucyshyn, Albin & Nixon, 1997; Lucyschyn et al., 2015). In two studies it was unclear who was acting as raters (Barry & Singer, 2001; Moes & Frea, 2000). This is important as it has been shown that ratings are impacted when the raters receive feedback from the interventionists or researchers (Kent & Foster, 1977). It could, therefore, be expected that ratings may also be affected when done by the interventionists or primary researchers themselves.

Outcomes

How are outcomes measured?

All but one study (Bynnendyk & Lucyschyn., 2008) included some measure of problem behaviour. Five studies used standardised measures (Bailey & Blair, 2015; Inchley-Mort et al., 2014; Reid et al., 2013; Durand et al., 2012; Dunlap & Fox, 1999). All studies aside from the two service evaluations (Inchley-Mort et al., 2014; Reid et al., 2013) used ratings of behavioural observations.

Behavioural observations are the primary method of assessment used in single-case designs and are also particularly appropriate for studies evaluating PBS, as the primary goal is generally to reduce the frequency or severity of a target behaviour. However, as with any other measure, it is important to ensure that the observations and the resulting ratings are reliable and valid. Key to reliability is ensuring that targets are clearly and fully defined and that a measure of inter-rater agreement is obtained.

Kazdin (2011) recommends two measures of inter-rater agreement be provided, a point-to-point ratio (number of instances agreed/number agreed and disagreed x 100), which is generally recognised as being the preferred method of obtaining reliability, as well as a measure of total agreement. The use of point to point measures of inter-rater reliability was an area of strength for the studies in this review with all except Lee et al. (2007) and Bailey and Blair (2015) including this. Only Dunlap and Fox (1999) provided an additional measure of agreement on totals.

Ensuring validity of behavioural observations can be more challenging and Kazdin (2011) recommends the use of multiple measures, probes and social validity checks in order to achieve this. The use of multiple probes and social validity checks were areas of strength in the studies included in the current review. All studies which report the use of behavioural observations conducted multiple probes and ten studies used a measure of social validity (Bailey & Blair, 2015; Binnendyk & Lucyschyn, 2008; Blair et al., 2010; Carr et al., 1999; Donellan et al., 1985; Koegel et al., 1998; Lucyshyn et al., 1997; Lucyshyn et al., 2007; Lucyshyn et al., 2015; Sears et al., 2013). Four studies also used either an additional standardised measure or parent rating of behaviours (Bailey & Blair, 2015; Durand et al., 2012; Dunlap & Fox, 1999; Lucyshyn et al., 1997)

One of the limitations identified in the earlier research was a lack of data on outcomes other than those related to problem behaviours (Carr et al., 1999). It appears that this is something which is beginning to be addressed as, although, reduction in challenging behaviour still appeared to be the primary measure of effectiveness, a variety of other measures of success were also identified in the current review. Nine studies used behavioural observations to obtain a measure of appropriate or adaptive behaviours, such as steps in routine completed or a specific targeted behaviour (Bailey & Blair, 2015; Barry & Singer, 2001; Blair et al., 2011; Bynnendyk & Lucyschyn, 2008; Carr et al., 1999; Lee et al., 2007; Lucyshyn et al., 2015; Moes & Frea, 2000; Sears et al., 2013; Vaughn et al., 2002). Three studies included a specific quality of life measure, two (Lucyshyn et al., 1997, Lucyschyn et al., 2007) used the Resident Lifestyle Inventory (RLI; Newton et al, 1987) and one the Family Quality of Life Inventory (Binnendyk & Lucyschyn, 2008). Reid et al. (2013) used the three concerns questionnaire, where carers rate their top three concerns and the level of worry related to each concern. Inchley-Mort et al. (2014) measured social and mental functioning using the HoNOS, mental health status using the PASS-AD and unmet needs using the CANDID-s. Durand et al. (2012) also included the Questionnaire on Resources and Stress–Short Form (QRS-SF) – Pessimism Scale, as they were specifically targeting pessimism as part of their intervention.

Behavioural Outcomes

An overview of study results including the PND measures of effect calculated specifically for this review can be found in table 1. All studies found some evidence for the effectiveness of PBS, as indicated by a statistically significant result or a PND greater than 0.70, for at least one measure of problem or appropriate behaviour. Studies with mixed results will be discussed in more detail.

Lucyschyn et al. (1997), in their longitudinal study of a single participant found a significant result for two of the daily routines the intervention was targeting, with an additional routine approaching significance. However, they failed to find a significant result for a final routine used as a test of generalisation (i.e. was not directly targeted by interventionists). Bynnendyk & Lucyschyn (2008) in their study evaluating PBS to improve food refusal found a strong effect for overall food acceptance according to the PND score calculated as part of this review, but only a marginal effect for self-initiated food consumption. The Donellan et al. (1985) study of 16 participants found a strong effect for nine participants, a questionable effect for four and no observable effect as assessed by PND for three participants, although for two the level of problem behaviour was still lower than at baseline.

Of the two service outcome evaluations, Inchley-Mort (2014) found a reduction for only some types of problem behaviour, stereotypy and irritability, with only stereotypy still significant at follow-up whereas Reid et al. (2013) found a significant effect on total score for the Developmental Behaviour Checklist and parent concerns.

There were two studies where the choice of PND as a measure of effectiveness may not have been appropriate. Lee et al. (2007) found only a low effect after the withdrawal condition for problem behaviour and Koegel et al. (1998) found no effect for one child out of three. Both of these results were caused by outliers in the baseline data, which were taken as the lowest data point for comparison with intervention effects. A visual inspection of both of these results showed that the intervention appeared to be effective.

Quality of Life

Three studies included a quantitative measure of quality of life (Binnendyk & Lucyschyn, 2008; Lucyshyn et al., 1997; Lucyschyn et al., 2007). The latter two studies specifically looked at participant quality of life as measured by increased participation in activity, whilst the Bynnendyk & Lucyshyn (2008) study reported on overall subjective reports of family quality of life. All three studies found an increase in quality of life.

Dunlap and Fox (1998) interviewed the six families in their study and comments related not only to a reduction in challenging behaviour but also to families being better able to engage with their child, the child being able to play more independently, use specific new skills (i.e. communication or coping) and access new activities.

Social Validity

As mentioned previously, ten studies provided a measure of social validity. This assessed how acceptable, effective and relevant participants and their families believe the intervention to be. The majority of studies used individually developed measures which rated six to ten items on a Likert scale including factors related to intervention goals, strategies and procedures and outcomes. One study (Koegel et al., 1998) asked a group of support staff to view videos of intervention sessions and provide ratings of child happiness, parent happiness and stranger comfort with interacting with the child. They found an increase in ratings from baseline to the end of intervention. Donellan et al. (1985) interviewed those implementing the behaviour support plans regarding their perceptions of the success of the intervention.

Bailey and Blair (2015) provided the most comprehensive measure of social validity including family member ratings as well as an interview. In addition, they also used independent observers who were familiar with the principles of PBS but naïve to the study to rate the acceptability of the behaviour plan and the child’s behaviour. All three measures showed high levels of social validity with some of the qualitative comments including the intervention preparing them to independently implement strategies and to apply the principles to novel situations.

Although no statistical analysis was done, all studies which included a measure of social validity either showed an increase over the course of treatment or in the case of qualitative interviews found the intervention to be appropriate and effective. The exception to this was the study by Moes and Frea (2000) which found low levels of social validity and consumer satisfaction for the prescriptive PBS intervention and high levels for the contextualised intervention.

Other outcomes

As the specific measures have been previously described these will not be repeated. Durand et al. (2012) included a measure of parental pessimism related to their child's challenging behaviour and found a significant decrease after receiving PBS. Interestingly the study was an RCT comparing PBS with PBS plus optimism training but there was no significant difference between groups on pessimism. Reid et al. (2013) found a significant decrease in parents' levels of worry related to challenging behaviour. Lucyschyn et al. (2015) found a decrease in parent-child coercive interactions and an increase in constructive parent-child interactions. The Inchley-Mort (2014) study found no significant difference between groups on unmet needs, social and mental health functioning or mental health status.

Generalisation and follow-up

One of the criticisms of early PBS research was that it tended to focus interventions only on targeted behaviours and did not include sufficient follow-up periods (Carr et al., 1999; Lucyshyn et al., 2007). As one of the central tenets of PBS is that it should occur within the context of the whole of the person's life and take a lifespan perspective this was a significant limitation of the research. In the Carr et al. (1999) review they found that only 1% of studies included a follow-up period greater than one year. Significant improvements appear to have been made since then with five of the included studies having follow-up measurements over one year (Inchley-Mort et al., 2014; Lee et al., 2007; Lucyshyn et al., 2015; Lucyschyn et al., 2007; Binnendyk & Lucyschyn, 2008). An additional eight studies (Bailey & Blair, 2015; Barry & Singer, 2001; Blair et al., 2010; Carr et al., 1999; Donellan et al., 1985; Lucyschyn et al. 1997; Moes & Frea, 2000; Sears et al., 2013) included either a shorter follow-up or a maintenance period, although in the case of Sears et al. (2013) this was only two weeks. All of the studies which included follow-up measures found evidence for the continued effectiveness of PBS.

Additionally, this review also found increasing use of generalisation phases. A generalisation phase is where changes in behaviours not directly targeted by intervention are measured in order to determine whether the skills learnt can generalise to other areas of the person's life. An example of this is Sears et al. (2012) who utilised a generalisation phase for one family, who were asked to design an intervention for a behaviour not previously targeted following the Prevent Teach Reinforce model. The family were allowed to ask for advice regarding specific interventions but were not provided any coaching. Six studies included generalisation phases where family members applied the skills or strategies learnt to target additional or new behaviours (Blair et al., 2010; Carr et al., 1999; Lucyshyn et al., 1997; Lucyschyn et al., 2007; Lucyschyn et al., 2015; Sears et al., 2012). Two additional studies included generalisation phases where existing strategies targeted the same behaviour but in a different context (Binnendyk & Lucyschyn, 2008; Moes & Frea, 2000). Of the studies which included a generalisation phase, all but Lucyshyn et al. (2007) found a significant effect.

Outcomes for different types of PBS

In the Durand et al. (2012) study comparing PFI to PBS, both groups showed a significant decrease in challenging behaviour and parental pessimism. The PFI group also showed significant improvement in scores on the standardised measure of challenging behaviour, The General Maladaptive Index of the Scales of Independent Behavior–Revised, when compared to the PBS group.

The two studies investigating PTR (Bailey & Blair, 2010; Sears et al., 2012) both found significant decreases in problem behaviour and increases in appropriate behaviour.

The Moes and Frea (2000) study found no effect for task engagement when using a prescriptive PBS plan, which then increased to a significant effect when the plan was contextualised to fit with family needs.

Discussion

Summary of main findings

This is the first systematic review to specifically look at the evidence for PBS's effectiveness in family contexts. Eighteen studies were identified as fitting with the inclusion criteria; one RCT, two service evaluations, one longitudinal cohort study and 14 single-case designs. These studies unanimously showed evidence for the effectiveness of PBS, although the effectiveness was reported as being limited to only some types of behaviours in one of the service evaluation studies (Inchley-Mort et al., 2014).

How do we define effectiveness?

All studies, with the exception of Bynnendyk and Lucyschyn (2008), included a measure of problem behaviour. This is not surprising given that PBS is an intervention designed primarily to target challenging behaviours. However, it is also an intervention which aims to improve skills and create alternative ways for people with disabilities to get their needs met. Therefore an equally appropriate outcome is whether there is a change in the use of alternative and adaptive behaviours. This was is in some way reflected in the literature with nine of the studies in this review including a measure of adaptive behaviour. It is worth noting that the four largest studies (Durand et al., 2012; Donellan et al., 1985; Reid et al., 2013 & Inchley-Mort et al., 2014) did not include such a measure.

The principles behind PBS also demand outcomes beyond those simply related to behaviour. At its heart, PBS is an intervention that aims to improve the quality of life of the person and their family, granted with the assumption that this will then also reduce the occurrence of challenging behaviour. Although this has been an area of improvement since the Carr et al. (1999) review, where quality of life outcomes were only reported for 2.6% of participants, there is still a need to address this in future research. Three of the eighteen studies in this review included a specific measure related to quality of life and one included qualitative interviews with family members which also addressed quality of life factors. In all of the studies which reported it, quality of life was found to have increased.

Methodological Quality of Studies

The evidence for the effectiveness of PBS in family contexts needs to be taken into consideration in relation to the methodological quality of the included studies. Despite the QA Tool being adapted to better incorporate single case-design research thirteen studies were identified as being of low methodological quality.

The nature of PBS however, does not necessarily lend itself well to large, well-controlled RCTs which are traditionally considered the ‘gold standard’ in terms of levels of evidence. Part of this difficulty comes from the nature of PBS interventions themselves, as the intervention delivered to each person is highly individualised, not only in terms of the strategies but also the length and intensity of the intervention. It is likely that that this is what has driven PBS research to be largely focussed on single-case design. It is not necessarily dissimilar to other forms of psychological/behavioural therapy, however, which whilst following a prescribed structure, are likely to be highly individualised in everyday practice. Durand et al. (2012) did manage, after all, to conduct an RCT of PBS vs PFI by structuring the intensity and duration of the intervention and providing a session by session protocol which allowed for individualisation and flexibility within this. Further RCT studies similar to this but comparing PBS to alternate interventions may provide some of the higher level evidence required in today’s age of evidence-based practice. Although some may argue that this would significantly limit the external validity of PBS, which will always remain a highly individualised intervention.

There are also a number of advantages to single-case study designs, including that they demonstrate how PBS can be effective on an individual level. They allow for a level of detail regarding what works and does not work in an intervention that RCTs cannot address. An example of this is the Moes and Frea (2000) study which found a significant increase in task engagement only after the intervention had been contextualised to meet family needs. This provides important information for those looking to implement PBS on the ground. Like RCTs, single-case designs also have limited external validity, although in this instance it is due to the fact that they are highly individualistic and generally representative of highly motivated participants.

The two evaluation studies identified as part of this review (Reid et al., 2013; Inchley-Mort et al., 2014), are an important first step in seeing whether PBS can be effectively implemented in clinical settings. Although they were generally supportive of PBS, the results of the Inchley-Mort (2014) study found significant results for only some types of behaviour. Additional research is therefore needed.

Strengths and limitations in the literature

The studies included in this review showed that there has been significant attempts to address some of the limitations in the literature identified by Carr et al., in their 1999 review. This includes longer follow up periods and the inclusion of generalisation phases, which are important given that the goal of PBS is to create enduring change and for skills and outcomes to be generalised to aspects of the person's life beyond those originally targeted by the intervention. Additionally, there has been an increased focus on including measures beyond those related to problem behaviours including measures of adaptive behaviours, quality of life and social validity. Measures of social validity are important as they assess how acceptable and effective participants and their families believe the intervention to be. In this review, ten studies included measures of social validity with all indicating that PBS was an intervention that was acceptable to participants and their families.

A limitation which occurred throughout the literature and impacts upon the generalisability and external validity of the studies was participant selection. Almost all of the studies required family members to be willing to commit to participating in the intervention as a key inclusion criteria. This means that the participants reflected in the research are likely to be highly motivated and have the time and resources needed to commit to implementing the behaviour plans. It is true that all research requires the engagement of participants to some extent, although there is a question as to how reflective this is of services in the real world.

Although PBS is a comprehensive intervention there is also a question as to whether the level of support provided to families in some of the studies is reflective of what is able to be delivered in a clinical setting. For example, in one study where the family did not feel able to implement the intervention as originally intended the researchers stepped in, implementing the majority of the intervention themselves (Barry & Singer, 2001). In a clinical setting it is possible that there would not be the resources to provide this and there is, therefore, a question as to whether the intervention would still have been effective.

Additionally, few studies reported information related to how participants were selected, how many were initially approached and how many withdrew or dropped out of intervention. This was largely the result of the majority of single-case study designs not reporting on this, although it is likely that attrition rates are very low for this type of design, due to the nature of the intervention and the probable care interventionists would have taken to ensure participants are likely to participate fully (Bailey & Burch, 2002). For larger studies reporting of attrition rates was an area of strength with three of the four non-single-case designs including information about attrition. For these studies there appeared to be evidence of reasonable rates of retention ranging from 63-80% suggesting that this is likely an intervention which is acceptable to many families. Further research is needed, however, to support these findings and to better understand the barriers for those who choose not to participate or do drop out of the intervention.

Issues related to family member motivation to engage and attrition are of significant importance when considering the effectiveness of PBS. This is due to the fact that not only is carer involvement one of the key principles of PBS, but evidence also suggests that outcomes are impacted by how well they engage with the intervention and make changes themselves (Carr et al., 1999). It is possible that the lack of reporting of attrition rates, in combination with the fact that family members who participate in PBS research are generally highly motivated and have the time and family resources needed to engage fully, has led to a lack of consideration of facilitators and barriers to engagement in PBS (Durand & Rost, 2005).

Another limitation is the fact that the majority of studies were conducted in either the United States or Canada and with male children. This has significant implications when considering the generalisability of the study to other countries and cultures and with female and adult participants. Further research in a variety of contexts is recommended.

Due to the fact that the majority of research stems from single-case designs, it is also worth considering how the methodology can be improved to provide higher quality studies. A strength of the existing literature is the use of experimental designs, such as ABAB and withdrawal designs, which allow for inferences regarding causality to begin to be made. Areas which should be addressed in future research are the use of independent (and preferably blind) raters and the control of confounds. Some confounds such as the impact of history and maturity are impossible to control for beyond the use of an experimental design, although factors such as the impact of increased attention and observation or intervention being provided in a novel way may be able to be addressed. In the current review, no studies specifically referred to controlling for these factors, although the Moes and Frea (2000) study could be considered to have done this by implementing the prescriptive PBS plan prior to the contextualised version.

Limitation of this review

A decision was made to limit the review to only studies which had been published in peer-reviewed journals. This was due to there being a large number of case studies presented in book chapters, conference presentations and unpublished dissertations, of which the quality and level of detail were varying. This decision is a potential limitation given the recommendation to include both published and unpublished studies in systematic reviews to reduce the risk of publication bias (Tacconelli, 2010). A review of the studies that were initially identified and then later excluded as a result of not being peer-reviewed, found that all papers reported a positive effect for PBS, as measured by a decrease in challenging behaviour or an increase in alternative adaptive behaviours. Given this, it was felt that the benefits of excluding non-peer reviewed studies outweighed the risks. However, the effects of publication bias in general still need to be considered, particularly given that the majority of studies identified were single-case study designs. Shadish, Zelinsky, Vevea, and Kratochwill (2016) found that single-case study researchers are more likely to attempt to publish when there are large effects and even found that up to 15% of researchers would consider dropping cases in order to increase a small effect size.

A decision was also made to exclude studies with participants with primarily physical disabilities or acquired brain injury. This was due to the likelihood that the nature and function of challenging behaviours in these groups are likely to be different. However, it is recognised that in the general population there is likely to be significant overlap and services will likely be working with people with co-morbidities.

In regards to estimating the effectiveness of single-case study designs, a decision was made to use PND as a measure. Traditionally visual inspection is the most common form of analysis for observational data, providing useful and valid information on an individual basis. It does not, however, lend itself well to systematic reviews and meta-analysis. PND was chosen as it has been shown to be a more conservative measure as well as a more accurate reflection of the data (Chen, Hyppa-Martin, Reichle, & Symons, 2016; Lenz, 2012), although it is significantly affected by outliers in the data, as was seen in two of the studies included in this review. Additionally, PND can only tell us how many data points were higher or lower than baseline, it tells us nothing of the magnitude of this effect.

The decision to use a single adapted quality assessment tool in order to appraise the methodological quality of all studies also presented some challenges and may have limited the validity of the quality assessments. Despite the adaptations to the tool, some items were difficult to score for single-case studies, due to these items not routinely being discussed in many of the articles, such as attrition and dropout rates, and control of confounds. For attrition, this resulted in the item being scored as unclear when not reported by the authors. As the control of confounds item specified that the authors should have specifically addressed how they controlled for confounds this was scored as weak unless there were clear attempts to use a methodology that addressed control of confounds, such as the use of a concurrent-multiple-baseline design, which is considered to exert greater control over history and maturation effects than non-concurrent designs.

An alternative means of evaluating the studies would have been to use a separate tool specifically designed for evaluating single-case study designs, such as the Risk of Bias in N-of-1 Trials (RoBiNT; Tate et al., 2013). This would have had the advantage of being able to use both measures in their original forms, both of which have been found to be reliable and valid (Tate et al., 2013; Thomas et al., 2004). The decision not to use the RoBiNT meant that some areas of methodological quality were not explored, such as randomisation and data analysis. Tate at al. (2013) suggest that randomisation can be achieved in single-case study designs by randomly allocating when individuals will receive the intervention, rather than randomly allocating to groups. Data analysis relates to incorporating some form of statistical analysis into the interpretation of results, rather than relying on visual inspection only (Tate et al. 2013). None of the studies in this review used either randomisation or additional data analysis. An advantage to using the adapted model meant that drop out and attrition rates were considered, as this is an item which is not included in the RoBiNT.

Despite the differences, there were also significant areas of overlap between the adapted quality assessment tool and the RoBiNT, including higher ratings for; the use of experimental design; independence of raters; detailed descriptions of the study context, such as participant selection and behaviours; training of raters; the use of multiple raters and adequate levels of inter-rater reliability. Given the level of overlap and the fact that no studies would have scored for items related to randomisation or data analysis, it is not felt that significant differences in study quality would have been found by using the RoBiNT.

Conclusion

Despite limitations, the results of all included studies were highly homogenous and fit with previous reviews of PBS which support its use within family contexts. Further research is needed, however, to compare PBS with other interventions and treatment as usual in order to fully establish its effectiveness when working in family contexts. One of the significant limitations of the studies included in this review was the representativeness of family member participants, given that highly motivated and engaged family members are more likely to be represented. Additional further research specifically considering processes of engagement for family members is warranted.

Despite the lack of 'higher level' evidence for PBS with families, it is still recommended as best practice within the UK by the NICE Guidelines (NICE, 2015) and the Royal College of Psychiatrists et al. (2004). This is likely due in part due to its focus on non-aversive practice and improving quality of life, which fit with the person-centered approach valued by health professionals and services. In the studies included in this review, negative consequences were not reported for any participants as a result of receiving PBS. Therefore in light of the relatively low risk of harm, the consistency of PBS values with health and social service goals and the results of the current review PBS can be considered a good choice of intervention when working with families managing challenging behaviour in someone with an intellectual disability or ASD.

Chapter 2

Developing an Understanding of Family Engagement in Positive Behaviour Support

Abstract

Young people with intellectual and developmental disabilities are significantly more likely to display challenging behaviour. This has a significant impact on not only their quality of life but also that of their family. Positive Behaviour Support (PBS) is currently considered best practice for managing challenging behaviour and has been found to be effective in family contexts. A key principle of PBS is that all members of the young person's support network participate in the assessment and intervention. It is therefore important to understand which factors are important in facilitating family member engagement. Six family members of young people with intellectual or developmental disabilities who have received PBS were interviewed about their experiences and factors they found helpful and hindering in terms of their engagement. The interviews were analysed using thematic analysis. All participants reported that they had found PBS helpful, although some reported being unable to implement specific strategies. All were able to reflect on both facilitators and barriers to their engagement in PBS. Five superordinate themes were identified; 1. PBS is more than just strategies; 2. Considering the family context; 3. The therapist/family relationship; 4. Acknowledging challenges and the ongoing nature of the problem; and 5. Supporting family member change. Family members described a variety of factors which contributed to their success in engaging with PBS as well as a number of barriers. The results are considered in relation to the existing literature and implications for clinical practice and future research are discussed.

Introduction

Young people with intellectual and developmental disabilities are at increased risk of displaying challenging behaviour (Baker et al., 2003; Murphy et al., 2005). ‘Challenging behaviour’ is a term which is used frequently in the literature and is defined in a paper by the Royal College of Psychiatrists, British Psychological Society, and Royal College of Speech and Language Therapists (2007) as behaviours that threaten the safety or quality of life of the person or others and result in responses which are aversive or restrictive. Key to this definition is the recognition that it is the responses to the behaviour which determine whether it is challenging, not the behaviour itself. This is due to the understanding that challenging behaviours serve a purpose for the person and are the result of mismatches between their needs and their environment (Royal College of Psychiatrists et al., 2007). Challenging behaviour not only has a significant impact on the young person, but also their family, with consequences including increased constraints on family activities, negative impacts on siblings and increased family stress (Doubet & Ostrosky, 2015).

Whereas traditional approaches to behaviour management generally focussed on addressing individual behaviours utilising a primarily behavioural approach, Positive Behaviour Support (PBS) uses a combination of behavioural, systemic, environmental and cognitive approaches (Carr et al., 2002) to not only manage behaviour but also to improve quality of life. It is currently considered best practice when addressing challenging behaviour (Royal College of Psychiatrists et al., 2007; National Institute for Health and Care Excellence [NICE], 2015).

PBS is a multicomponent framework that involves first conducting an in-depth functional assessment in order to develop an understanding of why the behaviour occurs. A comprehensive set of interventions or strategies is then developed based on this assessment and taking into account the values of the client and their family (Gore et al., 2013). Interventions aim to promote desired behaviours and make challenging behaviour unnecessary by developing new skills in the individual, changing the environment, for example, by removing stimuli that leads to challenging behaviour or by changing the responses of those in the support system. The idea is for the interventions to be implemented at all levels and by all those involved in the person's care (Carr et al., 1999; Dunlap, Carr, Horner, Zarcone, & Schwartz, 2008; Kinkaid et al., 2016).

There is a large evidence base supporting the use of PBS in reducing challenging behaviour including one large-scale research synthesis by Carr and colleagues (1999). This included 109 PBS studies, although many were single-case studies, published between 1985 and 1996. They found that 51.6% of PBS based interventions were effective, with effectiveness defined as a 90% reduction in the target challenging behaviour. Marquis et al. (2000) conducted a meta-analysis using the Carr et al. (1999) data and found a large overall effect size for the effectiveness of PBS in reducing challenging behaviour. A later systematic review by La Vigna and Willis (2012) investigating PBS for severe challenging behaviour concluded that PBS was effective, although they did not provide a specific definition of what was considered effective. They included 12 studies, although again relied heavily on single-case studies. These reviews are limited, however, in regard to their applicability to use in family contexts as many of the included studies were conducted in institutional settings with plans being implemented by healthcare professionals. When delivering PBS to young people living in the family home there is a need for professionals to work directly with family members as well as the young person (McCart, Wolf, Sweeney, Markey, & Markey, 2009).

Family members are generally central figures in a young person's support system, and it is recognised that they play a pivotal role in implementing PBS interventions. Systemic theories, stemming originally from Bronfenbrenner's ecological systems theory (1979), suggest that a person's environment, the people in their networks and how those people interact with them have a significant impact on challenging behaviour and quality of life (Franklin, 1980). Additionally, family members tend to have the most in-depth knowledge of the person and their contributions to functional assessments and the resultant interventions are therefore likely to be instrumental (Dunlap, Newton, Fox, Benito, & Vaughn, 2001).

Initial research appears to support the beneficial role of actively involving family members in PBS. Carr and colleagues (1999) found that PBS is more effective when people who typically provide support for the person are responsible for implementing interventions rather than professionals, with 61% of interventions being successful compared to 44.3%, with success defined as a 90% reduction in problem behaviour. The systematic review included as part of this thesis also found evidence that PBS can be used to reduce challenging behaviours and increase alternative appropriate behaviours when delivered in a family setting.

Although not investigating PBS specifically, a meta-analysis by Harvey, Boaer, Meyer and Evans (2009), looking at the effectiveness of behaviour intervention for people with learning disabilities, found that interventions were most successful when based on a functional assessment and involved teaching new skills, particularly when combined with either system change or more traditional behavioural interventions. On the surface this appears to fit well with the PBS model, however, their analysis also showed no direct benefit from family member involvement. This would appear to contradict the earlier findings of Carr et al. (1999), although these findings related to both professional and non-professional carers.

Harvey et al. (2009) suggested that a possible reason for their finding may be a more traditional mindset of the family member's role as being that of continuing to implement strategies developed by professionals. Consistent with this is the finding from Berryhill (2014), who interviewed six parents regarding their experiences of accessing support for challenging behaviour. She found that although families identified professionals as generally being helpful, services tended to be professional rather than family-centred. If this is the case, family members who have not been involved in the assessment process and the development of strategies may not be confident in their ability to implement them or feel that the strategies do not fit within their family context.

It would, therefore, seem important to understand how professionals can better work with families to ensure that interventions meet family needs; however, research in this area has been limited with a focus on professional opinions. Dunlap and colleagues (2001) made recommendations as to how best involve families based on their clinical experiences. These included recognising and respecting the fact that each family is unique, basing interventions on the family's priorities and identity, including interventions that will affect the quality of life of both the person and the family and working collaboratively.

Two qualitative studies have also investigated key stakeholder perceptions of outcomes and engagement in PBS (Ethridge, 2011; Hieneman & Dunlap, 2000). Hieneman and Dunlap (2000) interviewed 15 key stakeholders, including five family members, five service providers, and five consultants/trainers, to determine what factors affect outcomes in community-based PBS. Although not specifically looking at family engagement, of the 12 factors identified six could be considered to be closely related to family participation, including; system responsiveness, capacity of support providers, buy-in with the intervention, integrity of implementation, match with prevailing philosophy, and the contextual fit of the plan within the support network. Etheridge (2011), in an unpublished dissertation, conducted a similar study this time specifically investigating factors which impact on family member implementation of PBS strategies. She conducted 12 interviews with key stakeholders including organisation administrators, direct service providers, family support organization leaders, and researchers. She identified three major themes, one of which was the need to understand and match interventions to family need and resources. Two other themes related to service delivery systems and policy and community supports were also identified.

Although valuable insight can be gained from professionals working with families it would also seem important to consider family members' perspectives directly. Although both the Hieneman and Dunlap (2000) and Ethridge (2011) studies included family members in their samples of key stakeholders, they were chosen for their involvement, level of knowledge and training in behaviour support and were not specifically interviewed about their experiences of receiving a PBS service. Two additional studies, both unpublished dissertations, attempted to address this limitation by interviewing participants about their experiences of PBS but were limited by the fact that in the end participants did not necessarily receive PBS (Berryhill, 2014; Thomas, 2010).

Thomas (2010) interviewed three parents of children with autism who received PBS, although this was primarily within a school rather than a community-based setting. Unfortunately, one of the findings of this study was that none of the behaviour support plans appeared to be based on a functional assessment, parents were not generally included in the process and there was little evidence that plans were designed to be implemented in the home. Berryhill (2014) considered the experiences of six parents of preschool children with challenging behaviour in regards to PBS. However, the families interviewed had not necessarily received any formal behaviour support, with the interviews focussed on their general experiences of accessing help rather than participating specifically in PBS.

It is possible that the lack of attention to facilitators and barriers of family engagement may in part be due to the majority of previous PBS research being conducted with families who are highly motivated and have the time and resources needed to engage successfully in intervention, as found in the current systematic review, as well as a lack of reporting on attrition rates (Durand & Rost, 2005).

As discussed, PBS has been shown to be an effective way of managing challenging behaviour in people with learning and/or developmental disabilities. Central to PBS is the inclusion of all people in the person's support network and in the case of young people, particularly their families. This is due to the fact that families have a more in-depth understanding of the person and are the ones most likely to be implementing the interventions (Dunlap et al., 2001). Despite this, the literature related to engaging and working with families is still lacking and where it does exist is limited by the fact that it is generally based on the experiences of professionals. To date, there has been no study exploring family members' experiences of being involved in family-based PBS and factors they find to be helpful and hindering in regards to their participation. The proposed study, therefore, aims to fill this gap by interviewing the family members of young people with a learning or developmental disability who have received family-based PBS in order to identify factors they find helpful and hindering in terms of their engagement in PBS. It is hoped that this knowledge may then be used by services to better tailor interventions to meet families' needs and to support family engagement in PBS.

Method

Participants

Participants were recruited from two National Health Service (NHS) sites in London providing individualised PBS to young people with intellectual disabilities or autism. As a diagnosis was required by the referring services this was not assessed as part of this study. Participants were considered eligible if they were family members of a young person up to age 21 with a learning or neurodevelopmental disability who had undergone or were undergoing PBS and had received either an individualised behaviour support plan or been given strategies to implement within the home. The reason for including young people to age 21 is the increasing trend in the United Kingdom for child services to remain involved throughout the transition to adult services (Singh, Paul, Ford, Kramer, & Weaver, 2008). The reason for requiring family members to have already been given strategies or a behaviour support plan was to ensure that families would be able to talk about facilitators and barriers to implementation. Additionally, family members had to have been aged over eighteen, had capacity to consent and able to speak sufficient English to participate in the interview. This was due to funding limitations meaning the use of interpreters was not feasible and the fact that the role played by children in implementing strategies was likely to be different to adult family members. In order to increase the transferability of results, there were no restrictions on the type or level of challenging behaviour, type of disability, level of functioning, family structure or background.

Recruitments sites were encouraged to invite all eligible families to participate, regardless of their experiences with the service, in order to obtain a variety of participant perspectives.

Six family members expressed an interest in participating. All six met the inclusion criteria and were subsequently interviewed. This included five mothers and one father, none of whom were parents of the same child. Interviews were conducted on an individual basis. Three participants described themselves as white British, one as coming from a different white background, one as Asian-British and one preferred not to say. Young people were aged between seven and seventeen at the time of interview and displayed a range of challenging behaviours including aggression, self-injury and disruptive behaviour. Two young people had a diagnosis of autism, one had a diagnosis of profound and multiple learning disabilities and three had a dual diagnosis of both autism and intellectual disability. Four parents spoke of additional diagnoses such as ADHD, sensory processing disorder and hearing impairment. All young people lived in the family home and parents had day-to-day responsibility for their care. Three parents were currently engaged in intervention and three had received PBS services between six months and three years previously.

Methodological design

A thematic analysis of participant's perspectives of their experiences was conducted in order to identify and develop a thorough understanding of common patterns. Qualitative methods are generally recommended for developing knowledge in areas where there has been limited previous research (Pope & Mays, 1995).

Thematic analysis was chosen as it is flexible in terms of epistemological stance, allowing the methodology to be tailored to the research question (Braun & Clark, 2006). For this study a contextualist framework was considered appropriate as the researcher was interested in identifying patterns which could be applied beyond the participant group, suggesting a reality that can be accessed through interview, whilst also acknowledging that perspectives are influenced by a range of social factors. Consistent with this, an inductive approach to data collection and coding was used, where themes were developed from a systematic gathering and coding of all data rather than by attempting to fit data to existing theories or research. Consideration of the underlying processes which may have given rise to the themes identified based on existing literature was also undertaken in order to develop a deeper understanding of engagement processes in families.

Interview

Semi-structured interviews were used in order to encourage participants to speak openly and reflect on their experiences whilst also acting as a prompt and exploring emerging areas of interest.

The interview schedule, figure 2, was developed based on literature relating to aspects specific to PBS, factors early research found to be important for families receiving behaviour intervention and factors found to influence engagement in parent training. The decision to use factors identified in parent training more generally was due to the limited evidence base for family member engagement in PBS specifically. Two factors which emerge from this literature are parents' confidence in the intervention and belief that they have the skills and resources needed to make changes (Solish & Perry, 2008). The interview schedule was then reviewed by two family members of a young person with an intellectual disability who displayed challenging behaviour, one of whom was also a study participant, to ensure that it was clear and addressed all areas they felt were relevant.

Although interviews were guided by the interview schedule they were also shaped by the interviewer exploring emerging areas of interest; for example, asking for more detail or asking questions designed to follow emerging areas of interest. This is consistent with the inductive data collection approach discussed earlier (Charmaz, 2006).

Interview Schedule

Prior to commencing there was an introductory discussion about the purpose of the study, participants were asked if they had any questions and informed consent was obtained.

1. To start off with could you tell me a little about your son/daughter (prompts for their disability, any challenging behaviours etc.)

2. And how did you come to receive behaviour support services? (prompts for whose idea, who made the referral, were you in happy to be referred, waiting times)

3. Could you tell me what your understanding of positive behaviour support is? How did you come to this understanding? Did XXX service discuss what positive behaviour support is with you?

4. When XXX team started working with you what were your thoughts about whether PBS would work or not?
    a. Did this belief change at any time? And if so why?

5. Do you feel that you have a good understanding of why your child uses challenging behaviours? Can you tell me more about that? If so did you have this understanding

prior to starting PBS? Has your understanding changed over time of the PBS intervention? If so, how?

6. Can you tell me a little about what the PBs process for you was (i.e. the assessment process, developing strategies)? *

7. Do you remember receiving a behaviour support plan as part of the PBS process?
    a. Can you tell me more about the BSP?
    b. What did you think about the BSP?
        i. Further prompts if needed; Do you feel that the plan has been helpful; Do you feel like the therapist listened to and included your thoughts and opinions when developing the plan?
        ii. Do you feel that the recommended strategies are/were able to fit within your family's usual routines or activities?
        iii. Do you feel like the strategies in the plan can be/could be applied to all areas of your child's life (i.e. school and home)?
        iv. Do you/did you feel that, if followed, the strategies would lead to an improvement in your child's and your quality of life?

8. How confident did you feel about implementing the plan?
    a. What gave you confidence or what might have helped you to feel more confident?

9. What, if any, changes have you made as a result of PBS? (possible prompts such as the way you respond, changes to the physical environment etc.)
    a. If none why not? What made this difficult?
    b. If yes what do you think helped you make changes?

10. Did you experience any changes in yourself as a result of PBS? (i.e. attitudes, beliefs, wellbeing) If so what were these? *

11. Could you tell me generally how you have found receiving PBS?
    a. What were the most helpful aspect(s) if any?
    b. The most unhelpful aspect(s) if any?
    c. Do you feel like PBS has worked for you and your child? (Why? Why not?)

12. What, if anything, would you have wanted to be done differently?

13. Can you think of any barriers of difficulties other families might experience in terms of participating in PBS? *

14. Any other comments?

Each interview ended with a debrief and discussion to gather feedback from the participant as to how they found the interview as well as any suggestions for how it could be improved.

* Questions added as interview progressed based on participant feedback and emerging areas of interest

Figure 2. Interview Schedule.

Procedure

This study received ethical approval from the NHS Health Research Authority (ref: 17/LO/1110; Appendix 2) as well as Royal Holloway, University of London.

Clinicians from recruitment sites contacted family members directly and provided them with the study information sheets (Appendix 3) before obtaining consent to pass on their contact details to the researcher. The researcher then contacted potential participants by phone or email, based on their preference, to answer any questions and to organise an interview time and location. Interviews were conducted face-to-face in interview rooms at local services and in family members' homes.

Prior to commencing interviews participants were again given study information, informed of confidentiality processes and were able to ask any additional questions. Informed consent was then obtained prior to commencing the interview. Interviews took between 60-90 minutes and were audio recorded. All recordings were transcribed and anonymised by the researcher who also conducted the interviews.

Analysis

Interviews were analysed based on the six stage methodology described in Braun and Clark (2006). 1) the researcher read each transcript twice to become familiar with the data; 2) the researcher coded for any data which may have been useful, considered what each piece meant and gave it an initial name; 3) Coded data was compared and contrasted, and considered in terms of how they clustered together to generate initial themes; 4) Themes were reviewed, refined and considered as to how they might relate to each other; 5). Themes were further refined and defined and given names and 6) a report of the analysis was written which links the themes and data extracts to the initial research aim. This was done with the use of NVivo, an analysis software package.

As analysis in qualitative research is a subjective process, details about the researcher's background are provided for the purposes of transparency. The researcher is a white Australian woman with no children. She has an interest in and previous experience providing PBS to people with intellectual disabilities and autism. A reflective journal was also used to track the analytic process. This documented the researcher's thoughts and ideas regarding the coding process and allowed for constant reflection and engagement in the analysis and stimulated new ideas.

Reliability

Coding was done by the researcher. In order to improve validity, the coding for the initial interview was reviewed by one of the project supervisors and any areas of disagreement discussed and reconciled. Additionally a doctoral student studying clinical psychology who had no involvement in this study reviewed the coding of a different transcript to determine a level of inter-rater agreement. This was determined by counting the number of codes agreed/disagreed with. The percentage agreement was 92.90%. Areas of discrepancy were again discussed until an agreement was reached.

Other methods used to ensure the quality of the study and consistent with Mays and Pope's (1995) recommendations for conducting high quality qualitative research were; the inclusion of negative cases, such as participants who had been unable to implement strategies; a detailed description of the methodology; respondent validation, where a participant of this study reviewed the results to ensure they were reflective of their experiences and reflection and reflexivity through the use of a reflective journal.

Results

Interviews revealed that although participants had a diverse range of experiences with PBS they all felt that it had been helpful. Although generally the participants in this study appeared motivated and engaged in intervention, two spoke of not being able to implement some specific strategies, one spoke of a regression in their child's behaviour and all participants acknowledged challenges that they had faced.

Thematic analysis yielded five superordinate themes; *1. PBS is more than just strategies; 2. Considering the family context; 3. The therapist/family relationship; 4. Acknowledging challenges and the ongoing nature of the problem and 5. Supporting family member change.* Within these superordinate themes thirteen subordinate themes were also identified and are described in table 3.

Table 3.

Superordinate and Subordinate Themes Related to Family Engagement in PBS

Superordinate Theme	Subordinate Theme
PBS is more than just strategies	Working with the whole system
	Emotional support
Considering the family context	Matching the intervention to the family's resources.
	Keeping things simple
	The family's priorities and goals
The therapist/family relationship	Therapist qualities
	Working as a team
	Being open-minded and willing to try
Acknowledging challenges and the	Not everything will work

ongoing nature of the process.	It is an ongoing process
Supporting family member change	Becoming more relaxed
	Feeling more confident and in control
	Better understanding of behaviour

PBS is more than just strategies

Working with the whole system

Sharing of information and the need for consistency throughout the child’s support system is a key component of PBS and was seen as one of the most beneficial aspects by the family members in this study.

> “The good thing is that it brought everybody together… it saved a lot of time because otherwise, it would be me who would have to go through each one of them (Jamie).”

Although not unexpected given the age group, all participants reported some sort of school involvement in the PBS process and found this helpful. In some instances, school involvement was seen as a way to facilitate implementation of strategies at home by first introducing them at school.

> “that took some going, and some patience, but the school did that, I didn't have to do that, so sometimes, that's why it's a brilliant school, sometimes they say well we'll work on this…and then as soon as they have implemented it, I implement it at home (Jesse).”

Experiences prior to PBS with schools were mixed, with three participants specifically reporting positive experiences and two expressing frustration that schools did not appear to be addressing their concerns.

> “School kept saying ‘oh well, this is [his/her] way of expressing, you know, anxiety,’ and I, what I was saying was, you know, we have to come to the bottom of what it is, we cannot continue like this (Robin).”

In these instances, PBS was able to be supportive of family needs by incorporating the school in the process. The two family members who had previously experienced frustration felt that there was a benefit to having recommendations made by other professionals as the school was more likely to listen and to implement changes.

> “It was more about making everybody working together rather than me just nagging every teacher and everybody around as if it was my idea, so this, the service has legitimised the work (Jamie).”

This form of support also went beyond the school, with three participants identifying having the PBS therapist support them in getting additional services or support from other external agencies.

> “Because also [service] helped me out getting me in touch with social services, getting me extra support there, which I wouldn't have been able to do without [service] (Jesse).”

> “Sometimes when we have a problem with the, with the places, um, they cannot understand you and then they do not bother with you, but if you have a letter from [service]…they cannot say no (Taylor)”

Emotional support

Four family members identified emotional support for the family and just having someone listen to them as being something that they valued and was helpful.

> "It was about supporting the family and um, you know my health is just as important as [child]'s because if I'm not in the right frame of mind, um, and in the right head space and have the confidence to tackle some of the things that [service] were asking me to try it would have failed (Jesse)."

Three participants also spoke about feeling a sense of separation or isolation from their friends and peers, "I feel alone as far as, I feel isolated from the outside world (Ashley)," and identified having someone to talk to and normalise their feelings and concerns with as being helpful, with one participant likening it to having "a more experienced friend or village elder or like a granny who had seen it all before (Alex)."

Two participants spoke of feeling increased stress as a result of implementing PBS strategies themselves. This increased stress was the result of strategies leading to increased challenging behaviours or the parent having to suppress their natural instinct to be reactive to behaviour. Participants felt that it was important for the therapist to be supportive and understanding of this increased stress.

> "So it's really difficult to, to kind of keep my mouth shut, and I mean sometimes I see myself and I am sitting there and my body is so tense (Ashley)."

Considering the family context

Matching the intervention to the family resources

The families interviewed in this study identified a range of different strengths, limitations and resources, which impacted on their ability to participate in the intervention. Two family members spoke about having good family support, two spoke about a lack of support from co-parents, two spoke of having additional financial resources, three spoke of time constraints, and two spoke of having flexible employment situations.   Considering these different contexts and tailoring interventions was considered important in the success of PBS in the family home.

> “It’s something that has to fit into the lifestyle of the people (Alex).”

All six participants identified time or resource constraints as a factor which could impact on engagement and appreciated when PBS was able to be flexible around their needs

> “The biggest thing for anybody that has a child with special needs, let alone whether you are working full time or not, is time because everything takes much longer (Jamie).”
>
> “They have always been very accommodating around, around my work so it worked very well (Jamie).”

Families identified a need for the PBS process to specifically consider the fact that strategies would be implemented in a home, rather than school, environment and that there were limitations in terms of resources and differences in the needs of the young person between environments.

> "I go to the school and I copy what they are doing, so I try to do it here. Then it make [him/her] aggressive, when it make [him/her] aggressive I have to leave that one. Because in the school they have got a lot of, not only staff, it is a big place, special rooms, special things (Taylor)."

Keeping things simple

Some families highlighted the importance of keeping strategies simple with a recognition that if they were too complex or time-consuming then they were less likely to be implemented. Four participants spoke of the ease of implementation being a facilitating factor when they were able to successfully implement strategies and two participants spoke about not having sufficient time or resources as being a barrier.

> "I think the trickier things are to do, or more laborious they are to do, the less likely you are to do them (Alex)."

> "Maybe, you have only one child maybe, maybe it can work…but when I have four children (Taylor)."

Families also identified it as being helpful when PBS was able to make things easier in their daily life, when they were able to recognise that strategies which may seem more effort in the short term may make things easier later, or when it helped to simplify or break down issues they were dealing with.

> "So that is the most obvious thing that we have done, just moving things around, making it easier for things to happen (Alex),"

> "The easiest options aren't making it easier for you long-term (Jesse)"

> "It's helpful, and I mean also to help isolate problems rather than seeing it as a field of carnage (Alex)."

It was also important to consider the ease of implementation on a psychological as well as a practical level. One family member spoke of strategies being pleasant to implement; “it's lovely to do things with our [child] (Alex),” whereas another spoke about implementing a strategy as causing additional stress;

> “Dealing with [him/her] in the correct way actually had a negative effect on my health because I was holding it all in (Ashley).”

The family’s priorities and goals

Although only one family member specifically spoke about the importance of having clear goals as a means of facilitating engagement, two additional family members spoke about the importance of having a plan more generally.

> “If you don't have clear objectives that is just one of the things that you have to include in thirty others (Jamie).”

One thing that was clear throughout the interviews though, was that the family members all had different priorities in terms of the support they wanted from PBS. For example, some family members identified wanting to be given practical advice and support to use themselves as a priority, whereas for others the priority was more about bringing others in the support network on board.

> “What works for me is that I have specific targeted help understanding the behaviours and what to do with those behaviours (Robin),”

> “It was more for people at school to feel comfortable with dealing with it…rather than for us at home (Jamie).”

One family member also suggested that a potential barrier to engaging in PBS might be not wanting to focus on the negatives, suggesting it may be more beneficial for some to focus on quality of life goals rather than reducing behaviours.

> "Sometimes you just want to be like any other family and enjoy something positive rather than being constantly focussed on the most negative aspects of your life (Jamie)."

An overall benefit of understanding and considering the family context is that it helped families to feel understood and listened to and this built a sense of trust in the relationship.

> "[Therapist] really, really understood my child, really understood my family and the setup. I didn't feel like I was just another…you know because some people, you get the professionals and you think, are you talking about another family are you talking about the same child? Have you got me muddled up with someone else…and I thought, yeah I trust this person (Jesse)."

The therapist/family relationship

Therapist qualities (knowledgeable, honest and sensitive)

All families in this study reported feeling that the therapists they were working with were skilled and knowledgeable in the area and valued the advice that they were given. There was also a recognition, however, of the need to offer this advice in a tentative and non-blaming way.

> "She is probably very experienced. I think it probably comes with experience, I think it comes with knowledge (Alex)."

> “Advice was tentatively, very delicately, because when you are telling somebody ‘you could try this as an alternative to what you have been doing,’ you are really saying ‘actually what you have been doing isn't working’… but without it coming across as some sort of criticism (Alex).”

This was the case not only when dealing with the families themselves but also when dealing with other professionals and three family members specifically mentioned this as a strength they felt the therapists they worked with had.

> “I think [therapist] was very good about, you know very nicely saying, well this has to be done to school, and um and um also to us…so it is also about the way the messenger can make sure the information about what is learned is actually carried forward to a meaningful resolution (Robin).”

Other factors which were identified as being important in the therapist were a sense that they were listening and genuinely interested in their child, honesty, and patience.

> “I think the fact that [therapist] was very patient is a biggie, she listened to me, she, she would go over again and again and again until I got it, which was great (Ashley).”

Working as a team

Although families in this study had varying levels of understanding related to behaviour support all six stated that they were the expert in their child. Two family members felt that the understanding of their child’s behaviour which was developed and the resulting interventions were not things that were overly complex or beyond their abilities but that for whatever reason they had been unable to implement themselves.

> “I don't think there is, I mean anybody better to determine how things are with your child other than the parent (Alex).”

> “At the end, you probably could have done all this yourself but I just wasn't in the right frame of mind at the time, you were low, you were fragile and you just didn't quite know how to (Jesse).”

The role of the therapist can, therefore, be seen more as supporting families to overcome these limitations. And in fact, all six families interviewed in this study described a process of combining their expertise with the therapist in a two-way process, with the therapist in a position more like a guide.

> “She would come up with just some ideas, but I'd then come up with the solution if you know what I mean, because at the end of the day I know [child] the best but just her ideas, I kind of thought ‘Oh yes, I could do that, I know how I'm going to tackle this’ (Jesse)."

Being open-minded and willing to try

A common characteristic that all parents in this study had was a sense of being open-minded about the PBS interventions and willing to try, despite all participants also acknowledging some uncertainty as to whether PBS would work.

> “I can't say I was confident they would work, but I didn't lack confidence either, it was a bit of the unknown. So I was just, I was open-minded to it (Ashley)."

Acknowledging challenges and the ongoing nature of the problem

Not everything will work

A common theme that came during the interviews, and perhaps linked to the theme of being open-minded and willing to try, was, in fact, a recognition that not everything would work and that in the short term behaviours may get worse. One thing that was reported as helpful in managing this was simply acknowledging it, which then allowed for the therapist and the family to plan for it.

> "Obviously not everything worked straight away or we, or worked at all and we would have to change it (Jesse)."

> "In the beginning it is hard and then sometimes it is working and sometimes not working (Taylor)."

> "We knew that we were going to go through a rough patch because all of these things were a huge change for [child] and then it was working out things, well what can we do to soften that (Jesse)."

Five participants also spoke about not being able to implement strategies all the time, even if they were effective. Reasons for this included it being impossible in a practical sense due to the frequency of the behaviour or time limitations, the mood of the young person, and also that family members are not always going to be perfect. Again what families found helpful was acknowledging this and encouraging the family members "just to do your best (Taylor)" and to plan for challenges.

> "You start off being confident, yes I'm going to do it and then, but um, you never, never, do it religiously, you can't, you just can't, it just doesn't work that way (Robin)."

It is an ongoing process

All families in this study described PBS as an ongoing process, with behaviours and challenges varying along the way, whether this was the result of changes in circumstances or through the natural progression of time.

> "Because with autism you got always new problems, always you have, because since they are a baby they are developing, changing (Taylor)."

For this reason, most families felt that it was important for there to be some form of ongoing support not only during implementation but also into the future. One parent attributed their ability to implement the strategies to frequent appointments with the therapist during implementation and two parents described this as something that would be helpful for families who were struggling with engagement. Five of the family members in this study spoke about wanting some form of ongoing support into the future to prevent deteriorations and meet new challenges.

> "I think the motivation of having [therapist] round every week. Because I wanted to give her good news every week, we've tried it (Jesse)"

> "Things can just, disintegrate really, very quickly, if, if there is not that [ongoing support] (Alex)."

Three participants also spoke about being able to adapt the information and strategies they had learnt themselves to meet new challenges.

> "I tackled something again, the same principles I realised, the same principles applied with a lot of what [therapist] told me, applied with a lot of behaviours that [child] done (Jesse)."

Supporting family member change

Feeling more confident and in control

Most of the family members in this study described feeling more confident and in control as a result of PBS. "Actually [therapist] did lots for me, for my confidence as well (Jesse)." One parent talked about the biggest change for them being the shift in power dynamics in the house.

> "And so the biggest thing [therapist] gave me, it put control back on to my family situation, where I actually was back in charge (Jesse)."

Parents in this study spoke about the change in confidence being linked to having a plan in place as well as a greater understanding of the behaviours and how to respond.

> "It helped that she said 'Ok, this is what we do, we have a plan' (Robin);"
>
> "I still had a bit of that problem this summer but I knew how to tackle it (Jesse)."

Becoming more relaxed

One of the consequences of feeling more confident and in control was that family members were also more relaxed about their child's behaviour.

> "I actually became more relaxed about it because now I understood the behaviour (Robin)"
>
> "He will have his meltdowns and I will have my tough days but I'm quite calm about it now, not getting stressed too much about it (Jesse)."

This change came about not only as a consequence of increased confidence but also as the result of a better understanding of the behaviour and a realisation that it is an ongoing process.

> “It also was quite good at saying to us ‘calm down, don't worry, within this one element don't project all your fears of the future onto it’…so just calm down, deal with this, deal with it step by step (Alex).”

There was a recognition that this more relaxed approach then, in turn, enabled them to be able to think more clearly and respond more appropriately to behaviours.

> “By me taking a step back and taking a breath I was actually able to recognise ‘hang on, this is one I need to step in and deal with, that one I don't’ (Ashley).”

Better understanding of behaviour

Families felt that gaining a better understanding of why their child engaged in the behaviour was particularly valuable to them.

> “Probably the most important thing…I got into my head the realisation that [child] is not doing this on purpose, [he/she] actually has no control over this (Ashley).”

The parents in this study originally had varying levels of understanding of why their child displayed challenging behaviour; however, even those who felt that they had some understanding acknowledged benefits such as a deeper or more formalised understanding. For two family members, they felt that they still did not always know the specific reasons for their child’s behaviour but the knowledge that there is a cause and that the behaviour is functional was helpful anyway.

> “Sometimes you don't know what the problem is and just, I think through trying to define it and address it with positive behavioural support, it just formalises it much more in my head (Jamie).”

> “You see she give us the key, there is, even if you can't know the reason for him, even the small things, the small thing it is for him a mountain. So there is a reason (Taylor).”

Discussion

The current study was the first to ask family members about their experiences of participating in family-based PBS and adds to the limited literature related to engaging with family members in PBS. All of the families in this study described their experiences as generally being positive and all reported that they had found it helpful. Despite this, some family members spoke of not being able to implement specific strategies and one of a regression in their child’s behaviour. All families were able to speak about some of the challenges they faced or that other families in similar situations may face when engaging in PBS.

The aim of this study was to identify factors that may facilitate or hinder family member participation in the PBS process. Five superordinate themes were identified; *1. PBS is more than just strategies; 2. Considering the family context; 3. The therapist/family relationship; 4. Acknowledging challenges and the ongoing nature of the problem and 5. Supporting family member change.* Each theme will be discussed with consideration to how it relates to the previous literature and implications for clinical practice.

PBS is more than just strategies

PBS has been defined as a multicomponent framework which aims to improve quality of life for the person and those that support them (Carr et al., 2017; Gore et al., 2013). Inherent in this is the focus on broader forms of support than simply providing individual behavioural strategies. An important finding of this study was that families identified some of these broader supports as also being a facilitator for them in engaging in intervention.

The system-wide approach is key to PBS and family members in this study very much valued this as not only a means of bringing everybody together but also of sharing the burden of intervention; for example, having the school introduce a new strategy before it is introduced at home. Some families also described therapists as going beyond this by supporting them to access additional services. This fits with previous research, which suggests that external supports are a factor which may impact on family members' ability to engage with PBS (Ethridge, 2011) and is consistent with previous reports of PBS services incorporating or helping families connect with other services (Hienemen & Dunlap, 2000).

Another form of support identified as beneficial was the therapist addressing the emotional well-being of the family. This is important as parents of children with disabilities who display challenging behaviour generally experience increased levels of stress, anxiety, and depression (Falk, Norris, & Quinn, 2014; Lecavalier, Leone, & Wiltz, 2006). These findings are consistent with previous research which suggests that engagement in family-based interventions and outcomes are influenced by the family's emotional well-being. (Hieneman & Dunlap, 2000; Singer, Ethridge, & Aldana, 2007; Singer et al., 2002; Webster-Stratton & Hammond, 1990). This has important clinical implications for services which should consider how best to incorporate family well-being considerations into interventions. In the current study families reported that this was achieved by having the therapist spend time talking with them regularly in "a bit of a counselling session (Jesse)," but this could also include integrating additional interventions focussed on family well-being into PBS, such as increasing external support or individual/family therapy.

Considering the family context

The importance of context or "goodness of fit" (Albin, Lucyshyn, Horner, & Flannery, 1996) to positive behaviour support is not new. This involves ensuring that the intervention takes into account the values and priorities of the family and fits with their available resources. Families in this study highlighted this as being vital to how well they were able to engage with PBS.

Of particular importance was the need for PBS interventions to fit within the families' available resources, with two family members identifying time and resource limitations as being the primary reason they were unable to implement strategies and four family members identifying ease of implementation as a facilitating factor. This fits with the growing idea that PBS interventions and strategies should be incorporated into existing routines rather than being considered additional activities, which may be seen as burdensome (Lucyshyn, Blumberg, & Kayser, 2000). By addressing existing everyday routines this not only reduces the burden of participating in PBS but also helps families to see interventions as practical tools that can help simplify their lives.

Families in this study also reported a greater feeling of confidence and trust in the therapist as a result of feeling listened to and that their individual family context was understood. This leads to interventions that are individualised, consistent with family priorities and able to be implemented within the families' resources. These factors have been linked to 'family buy-in' of interventions and increase the chances that they will be able to successfully implement interventions (Hieneman & Dunlap, 2000; Marshall & Mirenda, 2002).

The therapist/family relationship

In the meta-analysis conducted by Harvey and colleagues (2009) mentioned earlier, a potential explanation for the finding that there was no benefit to including family members was that services were still primarily "professional led" with family members having little say in the development of strategies. This appeared to be supported by Berryhill (2014) who found that only one out of six parents were offered a truly family-based service to help manage their child's challenging behaviour. Interestingly all families in this study identified a process of collaborative working, combining the therapist's knowledge of behaviour intervention with their knowledge of their child. This is consistent with the movement towards person-centred services in which PBS in part emerged. The person-centred approach is based on the work of Carl Rogers (1957) and is built on the idea that people have an innate ability and predisposition to grow and to reach their full potential. It is, therefore, the person who is the expert and the role of a therapist is to empower them to do this. There is evidence in the literature to support the fact that when families and professionals work together PBS is more successful (Lucyshyn, Blumberg, & Kayser, 2000; Hienaman & Dunlap, 2000).

There is also a well-established literature on the importance of therapist qualities in building a therapeutic alliance, engagement in therapy and on outcomes in family-based interventions (Holdsworth, Bowen, Brown, & Howat, 2014; Karver, Handelsman, Fields, & Bickman, 2005). The qualities identified by families in this study are consistent with those in the literature, such as being empathic and non-blaming. In addition to therapist qualities, however, it is also important for the family member to have reached a point where they are ready and willing to engage in an intervention (Dunlap & Fox, 2007). This was evidenced by the fact that all participants in this study described themselves as generally being open-minded and willing to try.

Acknowledging challenges and the ongoing nature of the process.

The family members in this study went into the PBS process with an awareness that not all strategies would work and that some may even lead to an increase in behaviour, even if only as a temporary response to change. This would appear to contrast with research, which has suggested that parents who participate in parent training programmes are more likely to make changes when they are confident that it will work and are able to see early results (Hieneman & Dunlap, 2000; Kazdin, Holland, & Crowley, 1997; Moore & Symons, 2011). This has important implications for clinical practice as family members appeared to appreciate this possibility being acknowledged and prepared for early on and it is possible that this then acted as a protective factor if strategies were then not immediately effective.

There was also a recognition amongst family members that it is not always possible to implement strategies one hundred percent of the time. This finding is particularly interesting as consistency in implementation of PBS has been identified as a key factor related to outcomes (Hieneman & Dunlap, 2000). If this contrast is not handled correctly this could alienate families who may feel that they have failed when they are not able to follow plans completely. Therapists need to be able to support families to implement behaviour plans as consistently as possible, to empower them to use best judgement and to plan for obstacles and challenges. The family members in this study felt that support to “just do your best” was helpful in normalising this but also identified regular and ongoing support with implementation as being beneficial.

Lucyshyn and colleagues (2000) saw ongoing support with implementation throughout the entirety of the process as being a key principle in the provision of PBS with families. Although this initially requires additional time and resources from services, by working with families in a collaborative manner and supporting them to develop, implement and adapt strategies, these skills will be developed in the family members themselves meaning they may require less support in the future (Lucyshyn et al., 2000). In this study there was evidence of families starting to use the knowledge and skills they had learnt to meet new challenges. This is important given the recognition by all family members in this study that behaviours are always changing.

Supporting family member change

The theme “supporting family member change” focuses more on the processes of change rather than helpful or hindering factors. The reason for including this theme was the clear link between these changes and family members feeling more able to engage in intervention. By being aware of and actively facilitating these changes the therapist may be able to foster greater engagement in PBS earlier on.

One of the biggest changes reported by family members was a better understanding of why their child engages in challenging behaviour. The use of functional assessments as an integral part of PBS suggests that this should always be an early component of any PBS intervention. This is also an area which has been well researched in the parent training literature and there is evidence to suggest that family members are more likely to implement interventions and less likely to use more authoritarian parenting approaches as a result of better understanding the motivations and causes of behaviour (Allen, 1999; Dix, Ruble, & Zambarano, 1989; Whittington, Sofronoff, & Sheffield, 2009).

Parents in this study also identified themselves as feeling more confident in being able to manage their child’s behaviour as a result of PBS. There is already an evidence base supporting the impact of parental self-efficacy on challenging behaviour as well as the likelihood of engaging in therapy and implementing strategies (Hastings & Brown, 2002; Solish & Perry, 2008). This was the basis for Durand and colleagues (2012) adding an additional optimism component to a standard PBS intervention. When comparing results for the two interventions they found that although behaviour improved for both groups those parents who were in the group with additional optimism training reported a greater improvement. Interestingly, they found that parental pessimism decreased for parents in both conditions supporting the findings of this study that engagement in PBS alone increased parental self-efficacy.

One less-explored aspect of family member change identified in this study is parents feeling more relaxed or calm about the problem behaviour. Although this occurred in part as a natural consequence of understanding the behaviour and learning skills to manage it one parent also specifically mentioned being supported and encouraged not to catastrophise behaviours and "project all your fears of the future onto it (Alex)." An important implication for clinical practice could, therefore, be thinking about how to support family members with this earlier in the process. Ideas and techniques taken from third wave interventions such as acceptance and commitment therapy (ACT; Hayes, Strosahl, & Wilson, 1999) are one potential way this could be addressed. ACT aims to support people to be accepting of current situations and to move forward in a way that helps them to achieve their goals, an idea that fits well with the aim of PBS being to improve quality of life. This idea is supported by a study of the experiences of participants of a group-based program for parents of children with a disability who display challenging behaviour, which incorporated elements of ACT (Thompson-Janes, Brice, McElroy, Abbott, & Ball, 2016). The findings indicated that parents felt calmer and more confident after attending the group and linked this to specific elements of ACT such as considering their values and mindfulness exercises, which encouraged them to be more present in the moment.

Limitations

The aim of this study was to explore the experiences of family members who had participated in PBS in order to identify common factors which facilitated or acted as barriers to family engagement, with a view to these guiding future research and clinical practice. The small sample size is a significant limitation when considering how transferable the results are likely to be beyond the original sample. Traditionally when conducting thematic analysis sample sizes are larger, with Braun and Clarke (2013) recommending ten to 20 participants for medium scale projects and over 30 participants for large-scale studies. Although this suggests that the aims of the study may not have been fully met, it is felt that meaningful themes were able to be identified, which can offer tentative suggestions for future research and clinical practice. This is supported by the fact that there was a high level of consistency in the experiences of the current participants, with no new themes emerging in the last interview, as well as research which suggests that when conducting qualitative research the majority of themes are identified in the first five to six interviews (Francis et al., 2010; Guest, Bunce & Johnson. 2006; Morgen, Fischhoff, Bostrom, & Atman, 2002).

The question of how applicable the results of qualitative research are beyond the original sample is an issue which has been heavily debated in the literature (Ritchie and Lewis, 2003). Lincoln and Guba (1985) suggest that in order to combat this, researchers should provide ‘thick descriptions’ of the sample and the research process, which then allows the reader to determine how transferable to results are to other populations. Therefore detailed descriptions of the study population, data collection, and analysis process have been provided for the current study.

A second limitation of this study is the inherently subjective nature of qualitative research, as the researcher’s interpretation of the data is impacted on by their own previous experiences and cultural background (Charmaz, 2014). A number of controls were utilised in this study in order to minimise the effects of this based on the recommendations of Mays and Pope (2000). This included the use of a second coder, respondent validation, and the use of a reflective diary.

A third limitation is that the sample may not be reflective of all family members of young people with challenging behaviour. Participants in this study reported an overall positive experience with PBS, therefore, the views of those who did not find PBS helpful are not represented. Participants were also all parents and it is possible that other family members and carers may have different experiences and perceptions. Further, the majority of participants came from white backgrounds and all had lived in the UK for at least 15 years, limiting transferability. Future research could consider exploring the experiences of different family members, such as siblings, or interviewing whole families as well as including participants from a variety of ethnic backgrounds.

Reflexivity

Consideration of a researcher's background and culture are important when using qualitative methodologies as these can influence the interview process, the responses given by participants during interview (Richards & Emslie, 2000) and the way that the researcher interprets the data (Kacen & Chaitin, 2006).

An important part of this is considering the impact of potential power imbalances. Wang (2006) suggests that power imbalances in interviews can develop as the result of differences in gender, educational levels, socioeconomic status, and cultural background. As the researcher was a white middle-class female in a professional role it was felt that there was a need to consider the possible role power imbalances might play and to attempt to address these. This was done by the researcher deliberately adopting a curious rather than expert position, consistent with the stance taken in clinical settings. Additionally, the majority of participants in this study were women and research has suggested that power imbalances can be reduced in woman to woman interviews (Oakley, 1981).

Through discussions prior to the interview, some family members were also aware that the researcher had previously worked in a PBS service and all participants were aware that the researcher was connected with the NHS, particularly as they were recruited directly by the services from which they received PBS. This may have caused some participants to feel less comfortable speaking critically about PBS, should that have been their experience. Reassurance was provided to participants about the independence of the researcher from the recruiting services as well as the bounds of confidentiality and anonymity. Additionally, the use of the semi-structured interview format was thought to be helpful in managing this as it specifically invited participants to discuss any unhelpful factors. In general, the researcher's past experience in addition to the curious rather than expert position was felt to have been helpful in developing rapport.

In terms of analysis, the researcher was aware that her previous experiences and background may impact on the interpretation of the data. Having previously delivered PBS in a clinical setting it was important for the researcher to recognise that she was likely to have her own pre-conceived ideas about possible facilitators and barriers to family engagement. In addition to being mindful of this and making deliberate attempts to put any preconceived ideas aside a number of controls were put in place to help manage the risk of researcher bias based on the recommendations of Mays and Pope (1995). This included coding all instances of data that may be relevant, the use of a second researcher, a trainee clinical psychologist who was familiar with PBS, reviewing the results as well as one of the original study participants.

Implications

Despite its limitations, this study makes an important contribution to the literature around family engagement and PBS. This is the first study to explore family members' perceptions of participating in PBS and factors they found helpful and hindering and gives professionals providing PBS valuable insight into their experiences and the importance of considering family perspectives when developing services. It has also resulted in a number of considerations for future clinical practice. These include; incorporating elements to address family members' emotional well-being, collaboratively working with family members to develop interventions which fit with their priorities and available resources, acknowledging and planning for challenges and supporting family members to feel more calm and confident. Given the study's limitations, these recommendations should be considered preliminary.

There are also implications for future research. All of the participants in this study identified PBS as being helpful and it would be interesting to see whether the challenges to engagement identified are consistent with those who have not found PBS helpful. Future research could also look at the perspectives of other family members as well as those from a variety of cultural backgrounds. Finally, future research could more specifically investigate the impact of some of the factors identified in this study as contributing to family member engagement, such as the role of families' sense of open-mindedness and confidence in the intervention.

The results of the current study also fit well within the general PBS model, with some of the factors identified by family members as facilitating engagement being the same as some of the key elements and hypothesised mechanisms of change in PBS. These include the importance of achieving system change by incorporating the whole of the young person's support system, as well as tailoring the intervention to the priorities and goals of the person and their family. This study did not specifically explore the relationship between family engagement and outcomes in PBS. However, if it is assumed that increased family engagement in the intervention leads to better outcomes, as discussed in the introduction, then this would appear to lend some support to the inclusion of these components of PBS.

Conclusion

This study explores the experiences of six family members of young people with a developmental disability who underwent PBS and considers factors that they felt were helpful and hindering in terms of their engagement. Overall the findings of this study are generally consistent with the literature related to engaging with families, including the importance of the therapeutic relationship, collaborative working and supporting family member well-being. A number of more novel findings such as the importance of family members being open-minded and recognising that not all interventions will be effective were also identified.

Although the results of this study are promising in terms of the consistency within the experiences of the family members, as well as with past research, the limitations mean that these findings should be considered preliminary. Further large-scale qualitative and quantitative research is needed to fully understand the factors which impact on family member engagement in PBS.

Chapter 3

Integration, Impact and Dissemination

Introduction

This chapter aims to provide a reflection on my experiences of conducting the systematic review and empirical study, as well as to provide a synthesis between the two papers. This will include descriptions of decision making processes, strengths and limitations of the studies and considerations related to impact and dissemination of findings.

Empirical Study Choice

The general topic area of Positive Behaviour Support (PBS) was chosen due to my interest in intellectual and developmental disabilities and previous experience in delivering PBS. An initial search of the literature was conducted in order to gain a general understanding of the state of the research base and to identify potential gaps. Whilst there was a strong evidence base for PBS in educational settings, the literature was more limited in community and family-based contexts, with the majority of these studies being small in scale. I had initially thought to add to this evidence base by looking to conduct a larger-scale study evaluating effectiveness of PBS in community settings; however, I realised that this was likely to be beyond the scope of a clinical psychology doctorate project.

In reviewing the literature it also became clear that there was a lack of research related to PBS implemented specifically in family contexts. There appeared to be a general recognition of the importance of working with families but research into how best to do this was limited. I had previous experience in conducting qualitative research and felt that this methodology could be used to gain a better understanding of family members' experiences and to address this gap.

Rationale for Systematic Review

Although PBS is considered best practice when working with people with intellectual or developmental disabilities who display challenging behaviours (Royal College of Psychiatrists et al., 2007; National Institute for Health and Care Excellence [NICE], 2015), a search of the literature suggested that the bulk of the evidence-base has come from American educational settings. Only two systematic reviews have been conducted to review the findings specifically related to PBS in community settings (Carr et al., 1999; La Vigna & Willis, 2012) and none have focussed specifically on delivering PBS in a family context. Additionally the Carr et al. (1999) review was conducted over 30 years ago and the La Vigna and Willis (2012) review was limited to only those displaying severe challenging behaviours. As I was aiming to understand family member engagement in PBS for my empirical study, it was felt that a systematic review specifically looking at the effectiveness of PBS in family contexts was warranted to validate the need for this.

Systematic Review Reflections

Conducting the search

In keeping with recommendations on conducting systematic reviews (Tacconelli, 2010), two databases were chosen based on their suitability to the research question: PsycINFO and PubMed. Additional searches of reference lists were also conducted. Developing the search terms was challenging given that many interventions consistent with PBS are not necessarily identified as being PBS. The search terms were therefore chosen to maximise the likelihood of including all relevant studies. “Behaviour support,” “behaviour intervention” and “behaviour management” were chosen, with both American and British English spelling. However, it is possible that due to the variability in terminology used, some studies may have been missed. An additional strategy to increase the likelihood of all relevant articles being included would have been to include a manual search of relevant journals. Due to time constraints this was not feasible. A strength of the literature search is attempt to maximise transparency by detailing all steps of the process consistent with PRISMA guidelines (Moher et al., 2015).

Quality analysis

Although studies were not excluded based on methodological quality a means of assessing this was considered necessary in order to consider the validity of the overall review findings. Due to the high number of single-case studies identified, careful consideration needed to be given as to how best to do this. A search of existing evaluation tools found that the majority are designed for use with larger-scale control studies which would result in even well-designed and controlled single-case study designs being identified as weak. A decision was therefore made to adapt an existing measure, The Quality Assessment Tool for Quantitative Studies (QA Tool; Effective Public Health Practice Project, 1998). The adaptations were based on recommendations from Kazdin (2011) and are described in the review.

A limitation of the methodological assessment tool used, and the majority of other tools available, was that it did not evaluate external validity (Downs & Black, 1998). Two service outcome evaluations were identified in the systematic review (Inchley-Mort, Rantell, Wahlich, & Hassiotis, 2014; Reid, Scholl, & Gore, 2013); the methodological quality of these were assessed as ‘medium’ and ‘poor’ respectively. It was felt that this may not be an entirely fair assessment as the nature of service outcome studies lends itself to difficulties with things such as randomisation, control groups, blinding and the presence of confounders. However, there are significant advantages in terms of increased external validity.

Analysis

Given the high proportion of single-case design studies, consideration needed to be given as to how best to evaluate results. A criticism of some systematic reviews is that they simply restate the conclusions of the author without objectively evaluating the outcome (Scruggs, Mastropieri, & Casto, 1987) and I was conscious of not wanting to do this here. All studies in the current review used observational measures of behaviours prior to, during and after intervention with a visual analysis of graphical representations being the primary means of analysis. A number of methods for quantifying what is a significant result in single-case study design research have been suggested and were discussed in more detail in the systematic review. A decision was made to use percentage of non-overlapping data (PND), which measures the percentage of data points in the intervention phase that are greater, or lower than, the highest data point in the baseline phase, as it has been used more frequently in the literature and has also been found to be a more conservative measure of effect size (Chen, Hyppa-Martin, Reichle, & Symons, 2016; Lenz, 2012). This decision came with limitations as two studies were found to have had partially non-significant results due to the impact of outliers.

Inter-rater reliability

I found conducting the systematic review challenging as this was not something I have done previously. In addition to supervision and support in developing search terms and inclusion and exclusion criteria, the use of second reviewer checks allowed me to feel more confident in my decisions. The rate of agreement between myself and the project supervisor, who checked nine full text articles against the inclusion/exclusion criteria, was 100% and the agreement rate between myself and a different second reviewer, a trainee clinical psychologist, who co-rated four of the included articles using the quality assessment tool was 83.33%. Given that inter-rater reliability for tools assessing methodological quality are generally low (Armijo-Olivo, Stiles, Hagen, Biondo, & Cummings, 2012), I was pleased with this level of agreement.

Empirical Article Reflections

Rationale for methodology

There are a number of reasons for choosing a qualitative methodology. In addition to qualitative methods being recommended when there is a limited knowledge base, qualitative studies can also provide a rich and detailed analysis of experiences which would be difficult to explore with quantitative methods (Pope & Mays, 1995). Specifically relevant to this study, qualitative methods are a means of understanding the motivations behind behaviours, in this case engaging with PBS, and of finding out from service users themselves what they value and find helpful in services (Berkwits & Inui, 1998).

Initially a grounded theory methodology was considered with the aim of the study being the development of a model of family member engagement. This decision was reviewed after receiving feedback from the reviewers of my research proposal, who suggested that a thematic analysis might be more appropriate. After discussing with my project supervisors what I ultimately hoped to achieve from the study – an in-depth understanding of family members' experiences that could be used to make practical recommendations for services – it was agreed that this could be achieved without developing a specific theory of engagement, and a decision to use thematic analysis was made.

Interpretative Phenomenological Analysis (IPA) was also considered. IPA aims to understand participants' perceptions of personally significant experiences and has a very individual focus which relies heavily on researchers' interpretations (Smith & Osborn, 2004). As this study aimed to develop an understanding of patterns in participants' perspectives, which would be more broadly applied outside of the initial participant group, it was felt that this approach would not be appropriate.

Participant choice and recruitment

A purposive sampling method was used in that participants were family members of young people (aged under 21) who displayed challenging behaviour and had received PBS. The inclusion criteria specified that they had to have actually received PBS strategies or been given a behaviour support plan. This decision was made in order to gain richer data by allowing participants to reflect on the facilitators and barriers to implementing strategies, a key component of family engagement. The limitation of this was that the perspectives of those who had declined or dropped out of intervention in the early stages are not represented. In order to facilitate the ethics process a decision was made to only include participants over the age of eighteen. This was due to the need for different procedures related to risk and consent for minors. Additionally it was felt that the expectations of and the role played by younger family members in implementing strategies was likely to be different.

A decision was also made to recruit directly from services offering PBS rather than advertising for participants more broadly. One reason for this was that PBS was felt to be a term largely used by professionals and it was felt that families would likely not be aware they were receiving PBS specifically, meaning that they would be less likely to respond to advertisements and those that did would likely be family members who were more knowledgeable in the area, thus limiting transferability. This was something that was confirmed in the interviews as when participants were asked what PBS was the majority spoke of only some elements such as reinforcing positive behaviour or of the intervention being a positive experience in general.

Given that one of the aims of the study was to generate recommendations that would be transferable beyond the original participant group, the representativeness of participants was an important consideration. Two measures taken to address this were including detailed descriptions of the participant context as well as the use of maximum variation sampling. Lincoln and Guba (1985) suggest that the key to transferability of findings is providing a detailed description of the research context, which then allows others to make decisions as to how well the results transfer to other settings and contexts. Maximum variation sampling refers to sampling methods which aim to capture a wide range of perspectives in order to increase transferability. In order to achieve this there were no restrictions on the type of familial relationship to the young person, the type or level of disability or the type, severity or frequency of challenging behaviour. Additionally, services were encouraged to approach all potential participants who met the inclusion criteria. Unfortunately, the final sample only included participants who found PBS helpful and was also limited in terms of other demographic factors, such as most participants being largely white and all participants being parents, with five being mothers. This limitation is common in qualitative research and relates to a self-selection bias where people who are more interested in the research area, proactive about responding to recruitment requests and open to sharing their experiences are also more likely to participate (Robinson, 2014). It is possible that people who have had more positive experiences with PBS may be more interested as a result of their experience, or the very fact that they are more proactive, interested and open may have contributed to them having better results. As women are often more open to self-disclosure (Dindia & Allen, 1992) this can also lead to predominantly female samples (Robinson, 2014).

The recruitment strategy originally involved recruiting participants from a single service, although concerns emerged early on about the likelihood of there being enough participants. Two additional services were approached and agreed to be involved, which also had the benefit of increasing the variability of the sample. It was felt that this would be sufficient to secure twelve participants. After obtaining ethical approval, recruitment commenced with two sites and the third several months later. The delay was a result of several factors including focussing on the initial sites first, the availability of the contact person at the service, and ethics and research and development approval delays. Unfortunately the response rate was very low from one service and no participants were recruited from another, although there were only a few weeks between receiving final approval to recruit and the end of the recruitment period for that site. Several measures were taken to try to improve the response rate. The second service, which had initially sent a letter to service users followed this up with phone calls but this did not result in any additional participants. A number of third sector organisations were also contacted and although one had initially expressed interest they did not respond to further communications. At this point it was felt that there was not sufficient time to approach new NHS services and obtain research and development approval.

Initially it had been felt that the population would not be particularly difficult to recruit from, hence measures such as incentives for participation were not put in place. Interestingly family members who were interviewed identified a lack of time as being a barrier to engagement in PBS and also as being a general issue in families with children who have a disability. This may in part account for the low response rate, as potential participants may have found it difficult to find the time needed to participate in the interview.

Ultimately six participants were recruited for this study and it was felt that meaningful themes were able to be derived from these interviews. This is supported by a number of previous papers which have suggested that five to ten participants are sufficient for identifying the majority of themes in qualitative research (Braun & Clarke, 2013; Francis et al., 2010; Guest, Bunce, & Johnson 2006; Namey, Guest, McKenna, & Chen, 2016).

Interview schedule

A semi-structured interview format was chosen for the interviews. This form of interview allows participants to speak in an open manner without restricting them to specific topics, whilst also acting as a prompt for family members in order to gain more detailed information (Charmaz, 2006). The questions are considered a guide and can be changed and adapted to explore emerging areas of interest. For example, an additional question was added to ask about changes that had occurred within the family member themselves after the first four participants all spoke about aspects of personal change. This process is consistent with the inductive approach described earlier.

The interview schedule was developed by me, with support from the project supervisor, and aimed to draw information about participants' experiences of PBS at different stages as well as things they found helpful and hindering. Additional questions were drawn from the literature related to PBS, family needs and parent training. One challenge at this point was balancing the need to develop questions that would prompt participants to give detailed and thorough responses without shaping them to fit with the existing literature or researcher expectations. To manage this the initial literature review was kept very wide and aimed to get a more general understanding of the area rather than to develop specific hypothesis and theories. Additionally the questions in the interview were broad with prompts being used to get more specific information only when needed. Both were written so as to be non-biased and non-leading.

An additional advantage of using the semi-structured interview over a standardised interview meant that it allowed for the wording to be changed or simplified to meet the needs of non-native English speakers. This was relevant to the current study as one participant, although having sufficient English to participate in the study, required wording to be carefully chosen and adapted to meet their needs. There is evidence in the literature of a link between language barriers and not participating in research (Woodall, Morgan, Sloan, & Howard, 2010) and without this flexibility the representativeness of the sample would likely have been further limited.

Analysis

Consistent with recommendations from Charmaz (2006), data analysis occurred concurrently with data collection and transcription. All interviews were transcribed by me and I also conducted all interviews, allowing for increased familiarity with the data which is an important part of analysis.

Data analysis was conducted according to the six step process recommended by Braun and Clarke (2006) as described in the empirical article. Consistent with recommendations from Glaser (1999), all data which could be potentially relevant was coded even if it did not appear to be directly related to family member engagement. This was to avoid selecting only data that fits with existing literature or preconceived/early ideas. Given delays with recruitment and the deadline for submission I felt significant time pressure whilst conducting the analysis as it is a time intensive process, which involves going through the interviews line by line. More time to review and refine the emerging themes would likely have been beneficial, although, there are also benefits to this process being conducted in a more intense manner, such as being fully immersed in the data.

Ethics

Potential risks to the participant and me were important considerations in the planning stages of the study. Although it was not expected that the interviews themselves would be distressing it was considered important to be mindful of the fact that families with a child displaying challenging behaviour may already be experiencing high levels of on-going stress and that discussing this may potentially raise issues of distress or risk. The initial NHS research ethics committee submission included measures to address this such as using my skills as a trainee clinical psychologist, debriefing participants at the end of the interview, and considering additional sources of support that could be offered if needed. A benefit of the ethics process was that it identified the need to have a more robust distress protocol in the event that family members experienced distress or if a risk issue was raised (Appendix 5).

Service user involvement

There is increasing recognition of the importance of service user involvement in health research as a means of improving the quality of the results and of future service delivery (Department of Health, 2000; 2001). Given that the aim of this study was to contribute to improved services for families it was deemed important to include family member representatives beyond the role of participants. This was achieved by having two family members review the initial interview schedule, asking participants for feedback at the end of each interview and having one of the participants review the results to see how well they fit with their experiences.

This resulted in practical amendments being made to the study, such as the inclusion of a question in the interview about potential barriers for other families. An additional positive aspect was in helping me to maintain motivation and believe in the importance of my project. I was genuinely touched at the level of interest shown, with several family members specifically bringing up there hopes that this study would be able to help other families in the future.

Integration of Results for the Systematic Review and Empirical Article

Despite its limitations, the results of the systematic review suggest that PBS can be effective in managing challenging behaviours in family contexts. This lends support to current best practice recommendations and also justified the need for further research into understanding what factors may play a role in its effectiveness, such as family member engagement.

Of particular relevance to the empirical article was the finding that outcomes from highly motivated and engaged family members were significantly more likely to be represented in the literature. It has been suggested that this in combination with a failure to consider attrition rates has led to a lack of research into understanding family member engagement in PBS (Durand & Rost, 2005). The aims of the empirical study were therefore to address this gap.

Five themes were identified by the empirical article as being important factors which contribute to family member engagement. Although these results should be considered tentative, given the small sample size and limitations around transferability, they give valuable insight into the experiences of family members which can be used by services when considering how best to engage with families.

Impact

The systematic review conducted as part of this thesis is the first to synthesise the research literature related to outcomes in family-based PBS, whilst the empirical study was the first to ask family members about their experiences of receiving PBS. In general the results of both studies confirm those of previous research related to the effectiveness of PBS in reducing challenging behaviour and the importance of factors such as the therapeutic relationship, collaborative working and addressing family member emotional-wellbeing on engagement. There were also some novel findings such as family members not always being confident that strategies will work but finding it helpful when this is addressed and planned for early on.

Potential research and clinical implications of the two studies have been discussed throughout. Possible avenues of future research suggested by the systematic review include larger-scale studies into the effectiveness of PBS in family contexts and considering ways to improve the methodological quality of single-case design research.

In terms of further research into engaging family members, suggestions include gaining an understanding of the experiences of families who do not find PBS helpful, other family members and families from a variety of cultural backgrounds as well as research to address the specific aspects of family member engagement identified as being important in the empirical study such as the impact of family emotional well-being or open-mindedness on engagement and outcomes.

Both studies also have important implications in clinical practice, although due to the studies' limitations, the recommendations should be considered preliminary. The systematic review lends support to best practice guidelines that recommend the use of PBS when working with people who display challenging behaviour. This is important as although PBS fits well with the person-centred movement the evidence-base related to providing PBS in family contexts was limited. The empirical article gives clinicians valuable insight into the experiences of family members of young people with challenging behaviour who receive PBS. The themes identified led to practical suggestions for engaging family members in PBS including finding ways to address family members' emotional well-being, working collaboratively with family members, designing interventions that fit with the families' priorities, strengths and limitations, acknowledging and planning for challenges and set-backs and supporting family members to feel more calm and confident. It is hoped that this study will therefore be able to play a role in developing services which are designed to better meet family member needs.

Dissemination

In order to achieve these aims it is necessary for the results to be disseminated and to reach the researchers and clinicians most likely to be able to build upon them.

The first consideration is publication in a peer-reviewed journal. The decision as to which journal to submit to will be influenced by the areas of research addressed by the journal, word counts and willingness to publish qualitative research and systematic reviews. One possibility at the moment is the Journal of Positive Behavior Interventions as there is close match with the topic of interest, they welcome qualitative and systematic reviews and the word count is approximately 7500 words, which will allow for much of the detail in the two articles to be retained. Due to the two articles being highly related it is felt that submission of both to the same journal is appropriate.

It is also intended to present the results of these studies at a minimum of one conference. Again a primary consideration is the likely target audience so initial submissions will be to those which focus on PBS such as the British Institute for Learning Disabilities (BILD) PBS International conference held in the United Kingdom and the Association for Positive Behaviour Support international PBS conference held in America.

Other means of dissemination are also being considered in order to reach professionals providing direct support, service users and family members who may be less likely to attend conferences and read professional journals. As a first step the results of this study will be presented to the recruiting services and lay summaries of the results will also be sent to participants. A plain language summary of the study and results will also be developed and forwarded to services known to provide PBS in the UK.

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Appendix 1

Adapted Quality Assessment Tool

QUALITY ASSESSMENT TOOL FOR QUANTITATIVE STUDIES

EPHPP
Effective Public Health Practice Project

COMPONENT RATINGS

A) SELECTION BIAS

(Q1) Are the individuals selected to participate in the study likely to be representative of the target population?

1 Very likely 2 Somewhat likely 3 Not likely 4 Can't tell

(Q2) What percentage of selected individuals agreed to participate?

1 80 - 100% agreement 2 60 – 79% agreement 3 less than 60% agreement 4 Not applicable 5 Can't tell

RATE THIS SECTION	STRONG	MODERATE	WEAK
See dictionary	1	2	3

B) STUDY DESIGN

Indicate the study design

1 Randomized controlled trial 2 Controlled clinical trial 3 Cohort analytic (two group pre + post) 4 Case-control 5 Cohort (one group pre + post (before and after)) 6 Interrupted time series 7 Other specify ______________________________ 8 Can't tell

Was the study described as randomized? If NO, go to Component C.

No Yes

If Yes, was the method of randomization described? (See dictionary)

No Yes

If Yes, was the method appropriate? (See dictionary)

No Yes

RATE THIS SECTION	STRONG	MODERATE	WEAK
See dictionary	1	2	3

C) CONFOUNDERS

(Q1) Were there important differences between groups prior to the intervention?

1 Yes 2 No 3 Can't tell

The following are examples of confounders:

1 Race 2 Sex 3 Marital status/family 4 Age 5 SES (income or class) 6 Education 7 Health status 8 Pre-intervention score on outcome measure

(Q2) If yes, indicate the percentage of relevant confounders that were controlled (either in the design (e.g. stratification, matching) or analysis)?

1 80 – 100% (most) 2 60 – 79% (some) 3 Less than 60% (few or none) 4 Can't Tell

RATE THIS SECTION	**STRONG**	**MODERATE**	**WEAK**
See dictionary	1	2	3

D) BLINDING

(Q1) Was (were) the outcome assessor(s) aware of the intervention or exposure status of participants? 1 Yes 2 No 3 Can't tell

(Q2) Were the study participants aware of the research question?

1 Yes 2 No 3 Can't tell

RATE THIS SECTION	**STRONG**	**MODERATE**	**WEAK**
See dictionary	1	2	3

E) DATA COLLECTION METHODS

(Q1) Were data collection tools shown to be valid?

1 Yes 2 No 3 Can't tell

(Q2) Were data collection tools shown to be reliable?

1 Yes 2 No 3 Can't tell

RATE THIS SECTION	**STRONG**	**MODERATE**	**WEAK**
See dictionary	1	2	3

F) WITHDRAWALS AND DROP-OUTS

(Q1) Were withdrawals and drop-outs reported in terms of numbers and/or reasons per group?

1 Yes 2 No 3 Can't tell 4 Not Applicable (i.e. one time surveys or interviews)

(Q2) Indicate the percentage of participants completing the study. (If the percentage differs by groups, record the lowest).

1 80 -100% 2 60 - 79% 3 less than 60% 4 Can't tell 5 Not Applicable (i.e. Retrospective case-control)

RATE THIS SECTION	**STRONG**	**MODERATE**	**WEAK**	
See dictionary	1	2	3	Not Applicable

GLOBAL RATING

COMPONENT RATINGS

Please transcribe the information from the gray boxes on pages 1-4 onto this page. See dictionary on how to rate this section.

A	**SELECTION BIAS**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	
B	**STUDY DESIGN**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	
C	**CONFOUNDERS**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	
D	**BLINDING**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	
E	**DATA COLLECTION METHOD**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	
F	**WITHDRAWALS AND DROPOUTS**	**STRONG**	**MODERATE**	**WEAK**	
		1	2	3	NA

<u>GLOBAL RATING FOR THIS PAPER (circle one):</u>

1	STRONG	(no WEAK ratings)
2	MODERATE	(one WEAK rating)
3	WEAK	(two or more WEAK ratings)

Quality Assessment Tool for Quantitative Studies Dictionary

The purpose of this dictionary is to describe items in the tool thereby assisting raters to score study quality. Due to under-reporting or lack of clarity in the primary study, raters will need to make judgements about the extent that bias may be present. When making judgements about each component, raters should form their opinion based upon information contained in the study rather than making inferences about what the authors intended.

A) SELECTION BIAS

(Q1) Participants are more likely to be representative of the target population if they are randomly selected from a comprehensive list of individuals in the target population (score very likely). They may not be representative if they are referred from a source (e.g. clinic) in a systematic manner (score somewhat likely) or self-referred (score not likely).

(Q2) Refers to the % of subjects in the control and intervention groups that agreed to participate in the study before they were assigned to intervention or control groups.

B) STUDY DESIGN

In this section, raters assess the likelihood of bias due to the allocation process in an experimental study. For observational studies, raters assess the extent that assessments of exposure and outcome are likely to be independent. Generally, the type of design is a good indicator of the extent of bias. In stronger designs, an equivalent control group is present and the allocation process is such that the investigators are unable to predict the sequence.

Randomized Controlled Trial (RCT)

An experimental design where investigators randomly allocate eligible people to an intervention or control group. A rater should describe a study as an RCT if the randomization sequence allows each study participant to have the same chance of receiving each intervention and the investigators could not predict which intervention was next. If the investigators do not describe the allocation process and only use the words 'random' or 'randomly', the study is described as a controlled clinical trial.

See below for more details.

Was the study described as randomized?

Score YES, if the authors used words such as random allocation, randomly assigned, and random assignment.

Score NO, if no mention of randomization is made.

Was the method of randomization described?

Score YES, if the authors describe any method used to generate a random allocation sequence.

Score NO, if the authors do not describe the allocation method or describe methods of allocation such as alternation, case record numbers, dates of birth, day of the week, and any allocation procedure that is entirely transparent before assignment, such as an open list of random numbers of assignments. If NO is scored, then the study is a controlled clinical trial.

Was the method appropriate?

Score YES, if the randomization sequence allowed each study participant to have the same chance of receiving each intervention and the investigators could not predict which intervention was next. Examples of appropriate approaches include assignment of subjects by a central office unaware of subject characteristics, or sequentially numbered, sealed, opaque envelopes.

Score NO, if the randomization sequence is open to the individuals responsible for recruiting and allocating participants or providing the intervention, since those individuals can influence the allocation process, either knowingly or unknowingly.

If NO is scored, then the study is a controlled clinical trial.

Controlled Clinical Trial (CCT)

An experimental study design where the method of allocating study subjects to intervention or control groups is open to individuals responsible for recruiting subjects or providing the intervention. The method of allocation is transparent before assignment, e.g. an open list of random numbers or allocation by date of birth, etc.

Cohort analytic (two group pre and post)

An observational study design where groups are assembled according to whether or not exposure to the intervention has occurred. Exposure to the intervention is not under the control of the investigators. Study groups might be nonequivalent or not comparable on some feature that affects outcome.

Case control study

A retrospective study design where the investigators gather ‘cases’ of people who already have the outcome of interest and ‘controls’ who do not. Both groups are then questioned or their records examined about whether they received the intervention exposure of interest.

Cohort (one group pre + post (before and after)

The same group is pretested, given an intervention, and tested immediately after the intervention. The intervention group, by means of the pretest, act as their own control group.

Interrupted time series

A time series consists of multiple observations over time. Observations can be on the same units (e.g. individuals over time) or on different but similar units (e.g. student achievement scores for particular grade and school). Interrupted time series analysis requires knowing the specific point in the series when an intervention occurred.

C) CONFOUNDERS

By definition, a confounder is a variable that is associated with the intervention or exposure and causally related to the outcome of interest. Even in a robust study design, groups may not be balanced with respect to important variables prior to the intervention. The authors should indicate if confounders were controlled in the design (by stratification or matching) or in the analysis. If the allocation to intervention and control groups is randomized, the authors must report that the groups were balanced at baseline with respect to confounders (either in the text or a table).

D) BLINDING

(Q1) Assessors should be described as blinded to which participants were in the control and intervention groups. The purpose of blinding the outcome assessors (who might also be the care providers) is to protect against detection bias.

(Q2) Study participants should not be aware of (i.e. blinded to) the research question. The purpose of blinding the participants is to protect against reporting bias.

E) DATA COLLECTION METHODS

Tools for primary outcome measures must be described as reliable and valid. If 'face' validity or 'content' validity has been demonstrated, this is acceptable. Some sources from which data may be collected are described below:

Self reported data includes data that is collected from participants in the study (e.g. completing a questionnaire, survey, answering questions during an interview, etc.).

Assessment/Screening includes objective data that is retrieved by the researchers. (e.g. observations by investigators).

Medical Records/Vital Statistics refers to the types of formal records used for the extraction of the data.

Reliability and validity can be reported in the study or in a separate study. For example, some standard assessment tools have known reliability and validity.

F) WITHDRAWALS AND DROP-OUTS

Score **YES** if the authors describe BOTH the numbers and reasons for withdrawals and drop-outs.

Score **NO** if either the numbers or reasons for withdrawals and drop-outs are not reported.

The percentage of participants completing the study refers to the % of subjects remaining in the study at the final data collection period in all groups (i.e. control and intervention groups).

Component Ratings of Study:

For each of the six components A – F, use the following descriptions as a roadmap.

A) SELECTION BIAS

Strong: The selected individuals are very likely to be representative of the target population (Q1 is 1) **and** there is greater than 80% participation (Q2 is 1).

Moderate: The selected individuals are at least somewhat likely to be representative of the target population (Q1 is 1 or 2); **and** there is 60 - 79% participation (Q2 is 2). 'Moderate' may also be assigned if Q1 is 1 or 2 and Q2 is 5 (can't tell).

Weak: The selected individuals are not likely to be representative of the target population (Q1 is 3); **or** there is less than 60% participation (Q2 is 3) **or** selection is not described (Q1 is 4); and the level of participation is not described (Q2 is 5).

B) DESIGN

Strong: will be assigned to those articles that described RCTs and CCTs.

Moderate: will be assigned to those that described a cohort analytic study, a case control study, a cohort design, or an interrupted time series. **(For single case designs use of experimental design)**

Weak: will be assigned to those that used any other method or did not state the method used.

C) CONFOUNDERS

Strong: will be assigned to those articles that controlled for at least 80% of relevant confounders (Q1 is 2); **or** (Q2 is 1).

Moderate: will be given to those studies that controlled for 60 – 79% of relevant confounders (Q1 is 1) **and** (Q2 is 2).

Weak: will be assigned when less than 60% of relevant confounders were controlled (Q1 is 1) **and** (Q2 is 3) **or** control of confounders was not described (Q1 is 3) **and** (Q2 is 4).

D) BLINDING

Strong: The outcome assessor is not aware of the intervention status of participants (Q1 is 2); **and t**he study participants are not aware of the research question (Q2 is 2).

Moderate: The outcome assessor is not aware of the intervention status of participants (Q1 is 2); **or** the study participants are not aware of the research question (Q2 is 2); **or** blinding is not described (Q1 is 3 and Q2 is 3). (**For behavioural observations the rater is independent of the research team)**

Weak: The outcome assessor is aware of the intervention status of participants (Q1 is 1); **and** the study participants are aware of the research question (Q2 is 1).

E) DATA COLLECTION METHODS

Strong: The data collection tools have been shown to be valid (Q1 is 1); **and** the data collection tools have been shown to be reliable (Q2 is 1).

Moderate: The data collection tools have been shown to be valid (Q1 is 1); **and** the data collection tools have not been shown to be reliable (Q2 is 2) **or** reliability is not described (Q2 is 3). **(For ratings of behavioural observations this will be considered reliable when raters receive training and a minimum of 80% inter-rater reliability is achieved)**

Weak: The data collection tools have not been shown to be valid (Q1 is 2) **or** both reliability and validity are not described (Q1 is 3 and Q2 is 3).

F) WITHDRAWALS AND DROP-OUTS - a rating of:

Strong: will be assigned when the follow-up rate is 80% or greater (Q2 is 1).

Moderate: will be assigned when the follow-up rate is 60 – 79% (Q2 is 2) **OR** Q2 is 5 (N/A).

Weak: will be assigned when a follow-up rate is less than 60% (Q2 is 3) or if the withdrawals and drop-outs were not described (Q2 is 4). **(Single Case studies will be considered weak if there is no maintenance or follow-up phase)**

Appendix 2

NHS Ethics Approval

London - Stanmore Research Ethics Committee
Ground Floor
NRES/HRA
80 London Road
London
SE1 6LH

Telephone: 020 797 22567

<u>Please note</u>:
This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

17 August 2017

Mrs Sinead Botterill
Trainee Clinical Psychologist
Camden and Islington NHS Foundation Trust
Royal Holloway, University of London
Clinical Psychology - John Bowyer Building
Egham Hill, Egham
TW20 0EX

Dear Mrs Botterill

Study title:	**Developing an Understanding of Family Engagement in Positive Behaviour Support.**
REC reference:	**17/LO/1110**
Protocol number:	**N/A**
IRAS project ID:	**224751**

Thank you for your letter, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Alternate Vice-Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [University Research sub-committee approval]	1	16 February 2017
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [Initial Research sub-committee response]	1	19 January 2017
Covering letter on headed paper [Cover letter for Stanmore REC (Request for further Information)]	1	06 August 2017
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Indemnity insurance]		31 August 2016
Interview schedules or topic guides for participants [Interview guide/schedule]	1	19 January 2017
IRAS Application Form [IRAS_Form_25052017]		25 May 2017
Letters of invitation to participant [Invitation/information sheet]	2	08 May 2017

Letters of invitation to participant [Invitation/information sheet]	3	06 August 2017
Other [Participant Demographic Sheet]	1	17 May 2017
Other [Initial research proposal (v1)]	1	13 December 2016
Other [Participant Demographic Sheet]	2	06 August 2017
Other [Participant Demographic Sheet v2 (changes highlighted)]	2	06 August 2017
Participant consent form [Consent Form]	1	25 April 2017
Participant consent form [Consent to Contact]	2	08 May 2017
Participant consent form [Consent Form]	2	06 August 2017
Participant consent form [Consent Form v2 (changes highlighted)]	2	06 August 2017
Participant consent form [Consent to Contact]	3	06 August 2017
Participant information sheet (PIS) [Participant Information Sheet]	2	08 May 2017
Participant information sheet (PIS) [Participant Information Sheet]	3	06 August 2017
Participant information sheet (PIS) [Participant Information Sheet v3 (changes highlighted)]	3	06 August 2017
Research protocol or project proposal [Research Proposal v2]	2	19 January 2017
Summary CV for Chief Investigator (CI) [Sinead Botterill CV]	1	01 May 2017
Summary CV for supervisor (student research) [KT Primary supervisor CV]	1	08 May 2017
Summary CV for supervisor (student research) [SC Supervisor CV]	1	08 May 2017
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Research flowchart]	1	08 May 2017

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

<u>Reporting requirements</u>

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports

- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

17/LO/1110 Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr Anthony Kaiser Chair

Email:nrescommittee.london-stanmore@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: *Mrs Annette Lock*
Noclor , Central and North West London NHS Foundation Trust

NHS
Health Research Authority

Email: hra.approval@nhs.ne

Mrs Sinead Botterill
Trainee Clinical Psychologist
Camden and Islington NHS Foundation Trust
Royal Holloway, University of London
Clinical Psychology - John Bowyer Building
Egham Hill, Egham
TW20 0EX
Sinead.Botterill.2015@live.rhul.ac.uk

13 September 2017

Dear Mrs Botterill,

Letter of HRA Approval

Study title:	**Developing an Understanding of Family Engagement in Positive Behaviour Support.**
IRAS project ID:	**224751**
REC reference:	**17/LO/1110**
Sponsor	**Royal Holloway, University of London - Research Services**

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to

give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.

- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from www.hra.nhs.uk/hra-approval.

Appendices

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

After HRA Approval

The document *“After Ethical Review – guidance for sponsors and investigators”,* issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the HRA website, and emailed to hra.amendments@nhs.net.
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the HRA website.

Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/.

HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

Your IRAS project ID is **224751**. Please quote this on all correspondence.

Yours sincerely

Gemma Oakes Assessor

Email: hra.approval@nhs.net

Copy to: *Mrs Annette Lock, Royal Holloway University of London [Sponsor Contact]* Annette.Lock@rhul.ac.uk
Noclor, Central and North West London NHS Foundation Trust [Lead NHS R&D Contact] contact.noclor@nhs.net
Dr Kate Theodore, Royal Holloway University of London [Academic Supervisor] Kate.Theodore@rhul.ac.uk

Appendix A - List of Documents

The final document set assessed and approved by HRA Approval is listed below.

Document	*Version*	*Date*
Confirmation of any other Regulatory Approvals (e.g. NIGB) and all correspondence [University Research sub-committee approval]	1	16 February 2017
Confirmation of any other Regulatory Approvals (e.g. NIGB) and all correspondence [Initial Research sub-committee response]	1	19 January 2017
Covering letter on headed paper [Cover letter for Stanmore REC (Request for further Information)]	1	06 August 2017
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Indemnity insurance]		31 August 2016
HRA Schedule of Events [Schedule of Events]	1	05 June 2017
HRA Statement of Activities	1	05 June 2017
Interview schedules or topic guides for participants [Interview guide/schedule]	1	19 January 2017
IRAS Application Form [IRAS_Form_25052017]		25 May 2017
IRAS Application Form XML file [IRAS_Form_25052017]		25 May 2017
IRAS Checklist XML [Checklist_25052017]		25 May 2017
Letters of invitation to participant [Invitation/information sheet]	2	08 May 2017
Letters of invitation to participant [Invitation/information sheet]	3	06 August 2017
Other [Participant Demographic Sheet]	1	17 May 2017
Other [Participant Demographic Sheet]	2	06 August 2017
Other [Participant Demographic Sheet v2 (changes highlighted)]	2	06 August 2017
Participant consent form [Consent Form]	2	06 August 2017
Participant consent form [Consent Form v2 (changes highlighted)]	2	06 August 2017
Participant consent form [Consent to Contact]	3	06 August 2017
Participant information sheet (PIS) [Participant Information Sheet]	3	06 August 2017
Participant information sheet (PIS) [Participant Information Sheet v3 (changes highlighted)]	3	06 August 2017
Research protocol or project proposal [Research Proposal v2]	2	19 January 2017
Summary CV for Chief Investigator (CI) [Sinead Botterill CV]	1	01 May 2017
Summary CV for supervisor (student research) [KT Primary supervisor CV]	1	08 May 2017
Summary CV for supervisor (student research) [SC Supervisor CV]	1	08 May 2017
Summary, synopsis or diagram (flowchart) of protocol in nontechnical language [Research flowchart]	1	08 May 2017

Appendix B - Summary of HRA Assessment

This appendix provides assurance to you, the sponsor and the NHS in England that the study, as reviewed for HRA Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England to assist in assessing and arranging capacity and capability.

For information on how the sponsor should be working with participating NHS organisations in England, please refer to the, *participating NHS organisations, capacity and capability* and *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* sections in this appendix.

The following person is the sponsor contact for the purpose of addressing participating organisation questions relating to the study:

Name: Sinead Botterill

Tel: 075 535 94732

Email: Sinead.Botterill.2015@live.rhul.ac.uk

HRA assessment criteria

Section	HRA Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	No comments.
2.1	Participant information/consent documents and consent process	Yes	No comments.
3.1	Protocol assessment	Yes	No comments.
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	The sponsor has provided statement of activities and schedule of events for use as the agreement for participating in the study. The sponsor has confirmed that no other form of agreement will be used, or will be required.

Section	HRA Assessment Criteria	Compliant with Standards	Comments
4.2	Insurance/indemnity arrangements assessed	Yes	Where applicable, independent contractors (e.g. General Practitioners) should ensure that the professional indemnity provided by their medical defence organisation covers the activities expected of them for this research study
4.3	Financial arrangements assessed	Yes	External funding has not been obtained to run the study at site.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	The applicant confirmed she would be anonymising the recordings herself. The audio recordings will be made and stored on an encrypted device and transcribed within 2 weeks of the interviews. The transcriptions will be anonymised immediately, as they are transcribed, and the recordings deleted as soon as the transcriptions are completed. The transcriptions will be recorded on a password and fingerprint protected laptop in a private office space.
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments.
5.3	Compliance with any applicable laws or regulations	Yes	No comments.
6.1	NHS Research Ethics Committee favourable opinion received for applicable studies	Yes	REC Favourable Opinion was issued on 17 August 2017.
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments.
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments.

6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments.

Participating NHS Organisations in England

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.

There is one site type participating in this study. All research activity is the same at each participating NHS site, as detailed in the study protocol and supporting documentation.

The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. For NIHR CRN Portfolio studies, the Local LCRN contact should also be copied into this correspondence. For further guidance on working with participating NHS organisations please see the HRA website.

If Chief Investigators, sponsors or Principal Investigators are asked to complete site level forms for participating NHS organisations in England which are not provided in IRAS or on the HRA website, the Chief Investigator, sponsor or Principal Investigator should notify the HRA immediately at hra.approval@nhs.net. The HRA will work with these organisations to achieve a consistent approach to information provision.

Please note that the remit of HRA Approval is limited to the NHS involvement in the study. Research activity undertaken at non-NHS sites is therefore not covered and the research team should make appropriate alternative arrangements with relevant management at these organisations to conduct the research there.

Confirmation of Capacity and Capability

This describes whether formal confirmation of capacity and capability is expected from participating NHS organisations in England.

Participating NHS organisations in England **will be expected to formally confirm their capacity and capability** to host this research.

- The sponsor should ensure that participating NHS organisations are provided with a copy of this letter and all relevant study documentation, and work jointly with NHS organisations to arrange capacity and capability whilst the HRA assessment is ongoing.
- Further detail on how capacity and capability will be confirmed by participating NHS organisations, following issue of the Letter of HRA Approval, is provided in the *Participating NHS Organisations* and *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* sections of this appendix.
- The Assessing, Arranging, and Confirming document on the HRA website provides further information for the sponsor and NHS organisations on assessing, arranging and confirming capacity and capability.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and the minimum expectations for education, training and experience that PIs should meet (where applicable).
The sponsor has confirmed that a Local Collaborator would be required at each participating site and these have already been identified. GCP training is not a generic training expectation, in line with the HRA statement on training expectations.

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken
Access arrangements are not expected for local members of staff undertaking research activities within participating NHS organisations. A letter of access is not expected for researchers if the research activity is being carried out in an office within the participating NHS organisations. However, a letter of access is expected for researchers to carry out research activities for this study if the research activity is being carried out within a care setting on the premises of participating NHS organisations. If the researcher holds an NHS contract, an NHS to NHS letter of access will be required. No Disclosure and Barring Service or Occupation Health checks will be needed where a letter of access is required.

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England to aid study set-up.
The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Appendix 3

Participant information sheets

Royal Holloway University of London
Doctorate in Clinical Psychology
Egham Hill, Egham TW20 0EX, UK

Developing an Understanding of Family Engagement in Positive Behaviour Support

Name of Researcher: Sinead Botterill

I am carrying out a research study into the experiences of family members involved in Positive Behaviour Support (PBS). I would very much appreciate your input if you:

- Are the family member of a young person (aged under 21) with a learning disability and/or neurodevelopmental disability (such as Autism) who displays or has displayed challenging behaviour: and
- Have received PBS services

If this is you then your participation would be very much appreciated!

What is the purpose of this study?

Research has shown that family members generally want to be involved in the PBS process and that there may be greater reductions in challenging behaviour when this occurs. However, there has been very limited research into families' experiences of being involved in PBS, and aspects they find helpful or unhelpful. This study aims to gain an understanding of families' experiences of PBS, which can then be used to improve the way services work with families.

What does the study involve?

If you agree to take part you will be interviewed by the researcher about your experiences of positive behaviour support. The interview will take place in a location that is convenient for you and will probably take between 1 – 2 hours.

Do you have to take part?

No, taking part is completely voluntary – it is your choice if you take part or not. The services you or your family member receive now or in the future will not be affected in any way, whether you take part or not.

What to do if you are interested in learning more or participating in this research?

Please look at the more detailed information sheet. Then fill in the consent form to be contacted by the principle researcher.

Alternatively you can contact Sinead Botterill, the principle researcher directly at (email: Sinead.Botterill.2015@live.rhul.ac.uk phone: 01784414012) or Dr Alex Fowke, project supervisor (email: Alex.Fowke@rhul.ac.uk or phone: 01784 443600).

Royal Holloway University of London
Doctorate in Clinical Psychology
Egham Hill, Egham TW20 0EX, UK

Developing an Understanding of Family Engagement in Positive Behaviour Support

Participant Information Sheet

Name of Researcher: Sinead Botterill

Introduction

I would like to invite you to take part in this research study, which is investigating the experiences of family members of young people with a learning or neurodevelopmental disability (such as Autism) who have received positive behaviour support (PBS) services. Specifically, I am interested in what things you may have found helpful or unhelpful in terms of your involvement in the PBS process.

Before you decide to take part, you need to understand why the research is being carried out and what taking part will involve. Please take the time to read this information sheet carefully. Please ask questions if anything is unclear or you would like more information.

What is the purpose of this study?

This project is being completed as part of my clinical psychology doctorate at Royal Holloway, University of London. It is hoped that the project could provide useful information for healthcare professionals about how best to involve families in PBS.

Research has shown that family members generally want to be involved in the PBS process and that there may be greater reductions in challenging behaviour when this occurs. However, there has been very limited research into families' experiences of being involved in PBS, and aspects they find helpful or unhelpful. This study aims to gain an understanding of families' experiences of PBS, which can then be used to improve the way services work with families.

Why have I been invited to take part?

You have been invited to take part in this study because your family member has received or is receiving PBS services. Approximately twelve participants from London will take part in this study.

What will taking part involve?

If you agree to participate you will be asked to undergo a single interview about your experiences of PBS. The interview will be conducted in a location that is convenient for you and will take approximately 1 – 2 hours.

Before starting the interview you will also be asked to complete a brief questionnaire about you. This will include demographic questions such as your age, gender, cultural background, family member's disability etc. This information will be used to develop an understanding of the backgrounds of the people who participate in the study. This sheet will be completely anonymous and will not be linked to your interview. All of the questions on this sheet are voluntary and you can choose not to answer any you do not want to.

Do you have to take part in the study?

No, taking part is completely voluntary – it is your choice if you take part or not. You have been approached as a family member of a young person who has received PBS and may be interested in taking part, this does not mean you have to.

If you do not wish to take part you do not have to give a reason and you will not be contacted again. Similarly, if you do agree to participate you are free to withdraw at any time during the project if you change your mind. The services you or your family member receive now or in the future will not be affected in any way, whether you take part or not.

What are the possible benefits of taking part?

It is not expected that there will be any direct benefits to you or your family, however you may find the interview interesting and a chance to talk about your experiences. It is hoped that the study will provide useful information to services on how to involve families in PBS services.

Are there any potential risks or disadvantages?

The interview will involve talking about a family members challenging behaviour and what may have been, or is currently a distressing and difficult time for you. It is possible that during the interview you may find talking about your experiences distressing or emotional. If this happens you are free to end the interview at any time. I will debrief with you after the interview and discuss with you further sources of support should they be needed.

Equally it is possible that if you choose to complete the interview with another family member they may also say something that you find distressing or disagree with. It will be your choice as to whether you complete the interview with another family member or individually and this will be discussed with you prior to scheduling the interview. Again you will be free to end the interview at any time or to change your mind at any time and choose to participate individually or with another family member.

What if there is a problem?

If you have a concern about any aspect of this study, please contact Sinead Botterill, principle researcher (*Sinead.Botterill.2015@live.rhul.ac.uk or 01784414012)* in the first instance or the research supervisor, Dr Alex Fowke (Alex.Fowke@rhul.ac.uk or *01784 443600*).

If you remain unhappy and wish to complain formally you can do this by contacting Noclor research support through their website: https://www.noclor.nhs.uk/complaints

Will your information be kept confidential?

Yes. All information which is collected about you during the course of the research will be kept strictly confidential.

If you agree to participate the interview will be audio-recorded and transcribed. The audio recording will be destroyed as soon as it has been transcribed and the transcript will be anonymous with any identifiable information being removed. Both the recording and transcript will be stored electronically and secured with encryption and password protection. The transcription will be kept for five years and then destroyed. The contact details on your consent form will be stored in a locked cabinet inside Royal Holloway. Your name will not be disclosed to third parties and the researchers will have no access to your health information. No one from your PBS service or outside of the research team will have access to your interview.

What if I want to withdraw from the study?

You can choose to stop the interview or withdraw from the study at any time. The service or care that you or your family receive will not be affected in any way. If you withdraw from the study all the information collected from you will be destroyed and removed from all the study files.

What will happen to the results of this study?

The results of this study will be published in professional journals and may be presented at conferences. If you would like, a summary of the results can be sent to you.

We will not use any information that identifies you or your family in any report, publication or presentation. Direct quotes from the interviews may be used in reports and publications; however, the quotes will be anonymised to ensure that you cannot be identified.

What if I have further questions?

Please contact Sinead Botterill, the principle researcher (email: Sinead.Botterill.2015@live.rhul.ac.uk phone: *01784414012*) or Dr Alex Fowke, project supervisor (email: Alex.Fowke@rhul.ac.uk or phone: *01784 443600).*

Thank you for taking the time to read this information sheet

Appendix 4

Participant Consent Form

Royal Holloway University of London
Doctorate in Clinical Psychology
Egham Hill, Egham TW20 0EX, UK

Participant Identification Number:

Name of Researcher: Sinead Botterill

Developing an Understanding of Family Engagement in Positive Behaviour Support

CONSENT FORM

1. I confirm that I have read the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my or my family members care or legal rights being affected. ☐
3. I agree to the interview being audio recorded ☐
4. I agree to the use of anonymised quotes in publications ☐
5. I agree to take part in the above study. ☐
6. Would you like to receive a summary of the results of this study? **Yes** ☐

   If so please provide an address: **No** ☐

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________________________	____________________	______________
Name of Participant	Date	Signature
________________________	____________________	______________
Name of Person Taking Consent	Date	Signature

Appendix 5

Distress and risk protocol

Distress and Risk Protocol

Participants in this study are likely to be experiencing on-going heightened levels of stress related to having a family member with a disability who is currently or has previously displayed challenging behaviour. It is possible that some family members may find talking about their experiences to be distressing or that in the event of joint interviews one family member may say something that another family member finds upsetting or contentious.

These risks will be managed in a manner consistent with those used for managing risk in clinical settings including:

- Where more than one family member wishes to participate they will be asked to consider the possible risks and benefits of participating in interviews individually or jointly (as described in response to the previous point).
- Prior to commencing all interviews it will be reinforced with all participants that they do not have to answer any question they are not comfortable with and that they are free to end the interview at any time.
- Where multiple family members are participating in the same interview, ground rules will be established beforehand reinforcing the importance of respecting each other's opinions and not speaking over each other.
- The researcher will use her skills as a trainee clinical psychologist to manage any signs of distress or conflict as they arise during the interview and to redirect or end the interview as and when needed.
- A full debrief will be conducted at the end of every interview in order to address any sensitive or contentious issues raised during the interview. If needed appropriate sources of support will discussed and offered to the participant.
- Sources of support will be considered prior to contacting family members to schedule interviews. In most instances it is anticipated that this will be the service who referred them, however in the event that they are not currently receiving support from the service (i.e. have been discharged) options will be discussed and considered with the referring service to identify appropriate alternatives.
- In the event that the interview or debrief suggest that there is any risk to the participant, young person or their family then this will be discussed with the research supervisor and appropriate action taken. This may include either passing details of the risk onto the referring service or in the case of more serious or immediate risks, liaising directly with appropriate social or emergency services.

Appendix 6

Interview extract and coding

I: Yes, that sounds pretty good. The reason I am asking is because PBS is a model that a lot of services use but it's not always explained that that is what they are using. And so I am just interested in what parents understanding of PBS is. And what you were saying, it's about understanding the causes of the behaviour, that's a big part of it.	
B: Going right back to the roots, and actually [therapist] did do that, going back to really why and what triggered it of in the first place and it's not always the obvious thing. So it wasn't the fact that [his/her] speech and everything else, it was because I was just doing it for [him/her], because you do think it is the easiest option sometimes and then the easiest options aren't making it easier for you long term. And so the biggest thing [therapist] gave me, it put control back on to my family situation, where I actually was back in charge [I: Yes, that's great] and we worked on, we knew that we were going to go through a rough patch because all of these things were a huge change for [child] and then it was working out things, well what can we do to soften that, so we had, we introduced a calm box for [child] a special place for [child], we had, there was different stages so if the calm box didn't work and [he/she] was in and our safety or [his/her] safety was a concern then [his/her] room would be the best option, because that is somewhere, which is just, it's just a bed, nothing else, very plain, the toys [he/she] has in [his/her] room are soft toys so if [he/she] throws them, [he/she] is not going to hurt [his/her]self.	Changing understanding of the cause Therapist support to understand cause Family member role Easiest option not always easiest Increased control Collaboration Behaviour might get worse acknowledging challenges Planning for challenges Specific strategies Need to be safe
I: Could you tell me a little about the PBS process, just briefly, the assessment process, number of meetings that kind of thing?	
B: Oh God yeah, First of all was the initial assessment sort of, where I had a load of forms to fill out, so I had to fill out a questionnaire, so on a scale of I think five how often does [he/she] do this and I think it was like "never" "often" "a lot" kind of thing, and uh, so I filled in that and that was kind of what we were discussing at the initial meeting and that was when I had [his/her] assistant there, that was at [child]'s school and that was with [support person] [I:Yep] and then, then what was it, because she obviously had all the information from the school as well and then actually, this just amazed me, [therapist] was brilliant, she kind of wrote up the whole summary meeting, because she asked me there and then what specific areas did I want to work on and I wasn't quite sure because a lot of them overlapped, because a lot of it was about communication really so she helped me sort of narrow it down, what it could be, but we didn't sort of come to a final	PBS components/Initial Appointment PBS components/ Questionnaires External support School role Helpful factors/summarising Goals Simplifying or breaking things down

decision because we didn't want to say this is what would work, you know, and actually something else could be more important.	Collaboration
But the amazing thing was when she summarised the meeting, because I was sitting there at the time, thinking she is just listening, she is not making any notes, she may have made one or two but she really got what I was talking about and uh, I got this letter you know summary through, because she had arranged another meeting to come and give me it, so she could talk again to me. And um, she had, wrote out our meeting pretty much word for word, [I: Wow] and she kind of straight away got [child] without even meeting [him/her], she hadn't met [child] yet and I thought, yeah I trust this person and that's the thing, trusting that person that they completely understand your child and are going to work with your child. It's wasting my time if you're going to tell me to do this, do that and I'm going to be sitting here thinking that's absolutely impossible because [child] is not going to do that, you don't know my child, you know, she got [him/her] straight away without even meeting [him/her] and so I agreed, and that's when we had another discussion and we pinpointed the three things we were going to work on um, and um, how we were going to approach, initially approach it, because it kind of evolved over the weeks, how we were sort of adapting and changing and then something else might be thrown into the mix but it would still sort of be under that umbrella, because [child] has a very good way of you overcome one challenge [he/she] will find another one for you. So um it's keeping on our toes and trying to pre-empt [child].	Helpful factors/summarising Listening Therapist understood child/context Trust Time Unhelpful/not understanding Need for individualisation Collaboration Goals Ongoing process Adapting strategies Behaviour/problem changes Planning for challenges
So we narrowed it down to three things and it was [behaviour] um was it [behaviour] and about with [him/her] a bit more and um [his/her] like [behaviour] and oh and [his/her] [behaviour] so [his/her] behaviour and I had to every time I saw [therapist] I had to keep a record of all these things, what the triggers were, why it happened, how long, um, how long they happened, how bad they were um, so I kept a little sort of a tally, chart thing and that was good because even then once it was all down in front of me [therapist] would be there, but I would actually be coming up with the answers myself, because you can see it then, and you can see "Oh God, that's why it happened", because at that time or it could be because it's later on during the day [he/she] is getting more tired or it could be because [he/she] has been stuck in all day and it's been a miserable day outside and [he/she]'s not been able to release that energy.	Goals Specific behaviours PBS Components/functional assessment Collaboration Therapist as guide Family member coming up with answers Changing understanding of the cause

Appendix 7

Additional quotes to support themes

Additional Quotes

Superordinate Theme	Subordinate Theme
PBS is more than just strategies	**Working with the whole system** *“she is going and then if there is contact with anyone with doctors she is doing that, she is very good, she is supporting a lot… If I ask the doctors, I say he/she don't like a check-up do you think they are going to come to home, never, they are not going to do it. And they are not going to listen to me (Taylor).”* *“So it's, it's...I think it started a discussion in school, I think it opened up the school's eyes, because while the parents can go on about something, it's always better when a professional says ‘Yes, you have to do something’ (Robin).”* *“[therapist] would go into the school to assess [child] she would have meetings with the OT, with the teacher, so yeah they were involved um, but they, you know they were very respectful of the fact that [therapist] is who she is and she is in that job because she has been trained to be in that job, therefore she is to be listened to (Ashley).”* *“I think it's the good thing is that it brought everybody together because I know the majority of these health professionals do talk to each other but they don't, do talk to each other individually whilst this was, it saved a lot of time, because otherwise it would be me who would have to go through each one of them (Jamie).”*
	Emotional Support *“just talking about it and finding someone listening at the other end and trying to help, is in itself helpful… you have no idea what it feels like to see your child come home every day and everything and know that he/she, this is not normal, he/she must be suffering on some level and you cannot, nobody is doing anything. So um, it might seem like something inconsequential, but it’s a no brainer when somebody just listens (Robin).”* *“also because I think because I had quite a good little counselling session with [therapist] every week about my feelings and then, you know how, if I've had a bad week, or you know, well mainly with my [other family member] really. But she really, we would talk about it, it was good, it gave me a bit more, you know that I wasn't going mad, that kind of thing (Jesse).”* *“Like um, always the family they have more stress, because this, this that they are facing (Taylor).”* *“Yeah but it's um, it's, it's affecting my health, very much*

	affecting my health and um you know I feel very sad, very um I'm not going to say depressed because I think people use the word depression too easily (Ashley).”
Considering the family context	**Matching the intervention to the family’s resources.** *“so to follow the strategy of the school at home it is not working… um the facilities they are not the same, so always they tell us, uh follow the strategy of the school and then we try to tell them, not excuse, we like it, but the facilities are not the same (Taylor).”* *“Um that, [laugh] um we haven't managed to implement that a lot. Um but it, it's hard because the time is so limited (Jamie).”* *“you know if somebody has eight kids maybe they aren't going to be, or a job, or I don't know whatever, it’s not going to be so easy for them (Robin).”* *“And also I have support from my family as well, so I have my dad supporting me and my mum, um whereas some people might not have that support (Jesse).”* **Keeping things simple** *“Yeah, I think that was a realisation, that if you make it tricky it just won’t get done (Alex).”* *“You might look at it, so there is sometimes, things need to be a little bit briefer (Alex)” [re: Behaviour Support Plan]* *“so, that's quite embedded, those two strategies, were big one for me, but very simple, yeah, so um (Jesse).”* *“They weren’t you know, the strategies weren’t hard (Robin).”* *“So it's very complex, it's not the actual therapy, it's how you take [him/her] there, who's going to pick [him/her] up, you know um, incorporating that in [his/her] daily routine because [he/she] is in school for most of the day (Jamie).”* **The family’s priorities and goals** *“she asked me there and then what specific areas did I want to work on and I wasn't quite sure because a lot of them overlapped… so she helped me sort of narrow it down (Jesse).”* *“we have friends who have been through this and they just thought it was a waste of time… I guess they didn't have the idea what would be an outcome. For us the outcome was that the*

	school was brought on board…So that is a positive outcome for us. For them it was more of a, you know they were explained the behaviours that they already knew about (Jamie).” *“Umm, then they actually said that, they again offered us family therapy, which I found was not really what would help me...I want practical advice (Alex).”* *“We are not talking about behaviour, just sleep. This one is the big problem for us, the one we are concentrating on and we said maybe [he/she] is not sleeping and then the aggression is coming (Taylor).”*
The therapist/family relationship	**Therapist qualities** *“but [therapist] she has more experience, she has a lot of experience and then, she tell us the things, a lot of points. (Taylor).”* *“Sometimes professionals do tend towards the positive, but I like to have just a bit of honesty (Jesse).”* *“It's wasting my time if you're going to tell me to do this, do that and I'm going to be sitting here thinking that's absolutely impossible because [child] is not going to do that, you don't know my child (Jesse).”* *“even though I am sure [therapist] deal with lots and lots of different people I have never felt like I haven’t got their attention, which is great (Ashley).”* **Working as a team** *“no one knows my [child] like I do (Ashley).”* *“we talked through everything and she would listen and then she would come up with just some ideas but I'd then come up with the solution (Jesse).”* *“I think they did listen, I think it was a two way street (Alex).”* *“So, we were doing this ABC, right, and it was a bit like a light bulb moment and I just suddenly went, ‘ahhh, ok, right so when I'm doing that it's actually having a complete opposite effect to what I am aiming for (Ashley).”* **Being open-minded and willing to try** *“if you are in a situation where you think you are fighting fire any*

	support whatsoever you just think oh here is another bucket of water, fine. It's not going to put the fire out but it's help (Alex).” *“I just wanted help, so I was willing to try anything (Robin).”* *“Why do I try? [Yes] no, no, no because I have first of all you have to know if it is working or not with you, you have to try it (Taylor).”* *“Yeah, I just thought well I'm just going to give it a go, because [he/she]'s my [child] (Jesse).”* *“what I was doing wasn't working so I was well and truly open, my mind was open and whatever she suggested I was like, ‘OK, let's try’ (Ashley).”*
Acknowledging challenges and the ongoing nature of the process.	**Not everything will work** *“you know, we can only try, it doesn't mean that it always works (Alex).”* *“that one hasn't worked and I think we won’t be able to find a working solution for that one we just have to get on with it (Jamie).”* *“But I think what we recognised, that [he/she] would have a period of time, whatever it is, watching Peppa Pig on the iPad and [he/she] is going to be, you're banging your head if you are going to be trying to enforce it (Alex).”* *“I mean it was a very conscious thing I was doing, I would most of the time, not all of the time because honestly [he/she] would test the patience of a saint (Ashley).”* **It is an ongoing process** *“We recognise that the problem that you see in front of us that happens to be something which is on the problem box. It can have different contents at different stages (Alex).”* *“I think there should be continuity in terms of seeing if the strategy has helped, um, I think we had maybe one meeting to ask about that (Robin).”* *“if I stop for a while, new things come in and then we talk about it (Taylor).”* *“that's why I keep my feet in the door as much as I can, because I know I am going to need them at some point again in the future (Jesse).”*

Supporting family member change	**Becoming more relaxed** *“It's something that we were doing before, but we were worrying a lot more about it…we are a bit more relaxed (Alex).”* *“organically the process helped me to, to um, work on myself and how I deal with it as well and that, it used to upset me a lot, the behaviour, I mean really, I was just beside myself every day. I actually became more relaxed about it because now I understood the behaviour, a lot more (Robin).”* *“Always think of what could happen and be ready for it, but staying positive, not losing my temper, um, ah, just trying to remain calm (Ashley).”* *“you have to be yourself relaxed (Taylor).”* **Feeling more confident and in control** *“at one point in time I don't think I was brave enough and now I am and you know and I'm um, I'm um you know we just, you know, we are just doing a lot more with [him/her] than we used to and I thinks that's helped…I'm a lot braver, the changes I have made (Alex).”* *"[Daddy/mummy]'s back in charge now (Jesse)."* *“for me the biggest change is that people other people who work with him feel more confident, apart from [behaviour], that was me definitely, giving myself the permission to [use strategy] (Jamie).”* **Better understanding of behaviour** *“I got more of an understanding, yeah, um, yeah, you know when [young person] is feeling um more anxiety and worry and you know, it made me think about, ‘hold on a second, if I was in [child]'s shoes now, how would I be feeling?’ (Jesse).”* *“Initially I thought it was to do with frustration, and I realised that I approached it completely the wrong way (Robin).”* *“I mentally try to say ok ‘Why is that behaviour happening?’ and so it's made me more, you know, thoughtful, rather than just react to something (Alex).”*