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Practitioner Review: Current best practice in the use of parent training and other behavioural interventions in the treatment of children and adolescents with attention deficit hyperactivity disorder (ADHD)

Running head: Best practice in the use of behavioural interventions for ADHD

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Background: Behavioural interventions are recommended for use with children and young people with ADHD, however specific guidance for their implementation based on the best available evidence is currently lacking. **Methods:** This review used an explicit question and answer format to address issues of clinical concern, based on expert interpretation of the evidence with precedence given to meta-analyses of randomised controlled trials. **Results**: On the basis of current evidence that takes into account whether outcomes are blinded, behavioural intervention cannot be supported as a front-line treatment for core ADHD symptoms. There is however, evidence from measures that are probably blinded that these interventions benefit parenting practices and improve conduct problems which commonly co-occur with ADHD, and are often the main reason for referral. Initial positive results have also been found in relation to parental knowledge, children's emotional, social and academic functioning – although most studies have not used blinded outcomes. Generic as well as specialised ADHD parent training approaches - delivered either individually or in groups – have reported beneficial effects. High quality training, supervision of therapists and practice with the child, may improve outcomes but further evidence is required. Evidence for who benefits the most from behavioural interventions is scant. There is no evidence to limit behavioural treatments to parents with parenting difficulties or children with conduct problems. There are positive effects of additive school based intervention for the inattentive subtype. Targeting parental depression may enhance the effects of behavioural interventions. **Conclusion**: Parent training is an important part of the multi-modal treatment of children with ADHD which improves parenting, reduces levels of oppositional and non-compliant behaviours and may improve other aspects of functioning. However,

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blinded evidence does not support it as a specific treatment for core ADHD symptoms. More research is required to understand how to optimise treatment effectiveness either in general or for individual patients and explore potential barriers to treatment uptake and engagement. In terms of selecting which intervention formats to use it seems important to acknowledge and respond to parental treatment preferences. **Keywords:** ADHD, behaviour therapy, conduct disorder, parent training, treatment trials.

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Introduction

Behavioural interventions are defined in this review as those interventions which are directed at improving an individual's conduct (increasing desired behaviours and decreasing undesired behaviours), using strategies based on reinforcement and social learning principles and other cognitive theories. This includes classical contingency management, behaviour therapy (mainly through mediators such as parents or teachers) and cognitive behaviour therapy (such as verbal self-instruction, problem solving strategies or social skills training). These treatments are usually offered in several sessions over time, and implemented either through training the mediator(s), the child or both, with training guided by an explicit protocol (Sonuga-Barke et al 2013). Interventions employing behavioural techniques are recommended, and commonly used, in the treatment of children and adolescents with ADHD (NICE 2008). However, detailed evidence-based guidance on what, why, when and with whom these should be employed is not well described.

In the past, systematic reviews and meta-analyses have been cited as evidence of the value of these approaches for ADHD (Charach et al 2013; Corcoran & Dattalo: 2006; Fabiano et al 2009). However, it is our opinion that these reviews have often been over-inclusive, combining both randomised and non-randomised studies and that they have also lacked transparency, making it difficult to understand which studies and outcomes contribute to the stated effect size estimates. This makes their relevance to clinical practice difficult to interpret. These reviews also failed to address the issue of over-reliance on unblinded outcomes that is known to be a major source of bias in treatment trials (Wood et al 2008).

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The European ADHD Guidelines Group (EAGG) have recently conducted several meta-analyses of randomised controlled trials (RCTs) of behavioural interventions using stringent inclusion and exclusion criteria which have addressed these shortcomings (Sonuga-Barke et al 2013; Daley et al 2014). These meta-analyses used recognized scales to evaluate the quality of the studies. Most importantly, the EAGG attempted to address the impact of blinding on estimates of treatment efficacy. To do this the outcomes "most proximal" to treatment delivery, which in behavioural interventions are nearly always unblinded (e.g., ratings of symptoms by parents who received the intervention) were compared with outcomes judged to be "probably blinded" (e.g., direct observation by independent researchers or ratings by informants not aware of treatment allocation). Not all studies had blinded measures, but where they did, the difference between most proximal, and probably blinded ADHD assessments was clear. There was a statistically significant positive effect of behavioural interventions on the most proximal, parent ratings (d = 0.40; CI 0.20 - 0.60), and a non-significant effect when probably blinded measures were used (d = 0.02; CI -0.30 - 0.34). The EAGG concluded that, on the basis of current evidence, that takes in to account whether outcomes are blinded, behavioural intervention could not be supported for the treatment of core ADHD symptoms. The situation was different for other important outcomes (Daley et al 2014). Behavioural interventions had significant effects on probably blinded measures of parenting (positive parenting d = 0.63; CI 0.47 - 0.78 negative parenting d = 0.43 CI 0.24 - 0.62) and childhood conduct problems (d = 0.31; CI 0.05 - 0.57).

In this practitioner review, we provide, for the first time since the publication of these analyses, detailed interpretation of the findings and guidance for commissioners and clinicians on the use of behavioural interventions for the treatment of children and adolescents with ADHD. Our review has a broad scope covering issues of treatment benefits, therapeutic content and delivery as well as indications and contra-indications. There is also some consideration of the relationship between behavioural treatments and other non-pharmacological approaches. The issue of the relationship with medication, although important, is outside the scope of the current review and will be the focus of future publications. We have attempted to cover all interventions based on behavioural principles for children and adolescents. However, as nearly all trials that met the inclusion criteria for the EAGG meta-analyses (31 out of 32 studies) focused on parent-based

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approaches (i.e., parent training), most of our guidance relates to parent training or interventions with a parent training component in preschool and school-aged children. In line with our previous practitioner review (Cortese et al 2013) we have employed a question and answer format. Questions were generated after consultation with clinicians and service users. Answers were based on expert interpretation of existing best available evidence. As much of the evidence is drawn from studies with a major parent training component we will use the terms behavioural intervention and parent training interchangeably unless there is evidence that the effects would be different for parent training and other behavioural interventions.

Methods

Generation of questions and answers

There was consultation at various levels during the development of this Practitioner Review. The clinical questions were first created by the EAGG Behavioural Interventions BIn group (an interdisciplinary group of academic clinicians, of whom the majority are behaviour therapists], circulated to the wider EAGG group as well as ADHD advocacy groups and ADHD clinician groups in the UK, Netherlands and Belgium for feedback. Questions were amended in line with feedback; and further questions of clinical relevance suggested by these groups were added. After preparation of the first draft of the manuscript by the EAGG BIn Group, the manuscript received a first round of feedback from the wider EAGG group. After adaptation by the BIn group, there was additional final feedback from the wider EAGG group who are all clinicians and academics working in the ADHD area.

First bottom-up questions were drafted by the Bin group without ordering them into the 4 subthemes (1.treatment benefits, 2.therapeutic content and delivery, 3.treatment indications and 4.contraindications and relationship to other non-pharmacological treatments). After reviewing the questions, these 4 logical main themes of questions emerged, and questions were re-ordered into these subthemes. Feedback on order and placing of these questions under subthemes was provided by the broader EAGG group and ADHD advocacy groups and clinicians in the UK, Netherlands and Belgium.

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Providing answers: In each case answers are based on expert interpretation of the best available evidence. In terms of evidence, precedence was given to systematic reviews and/or meta-analyses of RCTs. Where no RCT data were available to answer a specific question, other evidence, including that from more pragmatic trials and observational studies, was taken into account on a case-by-case basis Strength of evidence ratings are provided for all recommendations using the Scottish Intercollegiate Guidelines Network SIGN development guide which rates levels of evidence from the highest 1++ which is evidence based on high quality meta-analysis to the lowest 4 where evidence is based solely on expert clinical opinion (see text box). In the case where met-analyses were available the SIGN ratings took into account the quality of the trials in the meta-analysis – downgrading those where the trials had a high risk of bias or where there was a high level of heterogeneity between studies – even if the meta-analysis itself was high quality. Effect sizes were interpreted according to criteria outlined by Cohen (Cohen 1992) with an effect size of 0.2 representing a small effect, 0.5 a medium effect and 0.8 a large effect.

SIGN Guidelines levels of evidence (www.sign.ac.uk)

1++ High quality meta-analyses, systematic reviews of RCTs, or RCTs with a very low risk of bias

1+ Well conducted meta-analyses, systematic reviews, or RCTs with a low risk of bias

1- Meta-analyses, systematic reviews, or RCTs with a high risk of bias

2++ High quality systematic reviews of case control or cohort studies. High quality case control or cohort studies with a very low risk of confounding or bias and a high probability that the relationship is causal

2+ Well conducted case control or cohort studies with a low risk of confounding or bias and a moderate probability that the relationship is causal

2- Case control or cohort studies with a high risk of confounding or bias and a significant risk that the relationship is not causal

3 Non-analytic studies, e.g. case reports, case series

4 Expert opinion

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Results

Each section of this Practitioner Review is divided into three parts. For each question the same format is followed. First a rationale for the question is provided. Then the relevant evidence that addresses that question is reviewed. Finally, a short concluding statement providing clinical guidance is made.

A) Treatment benefits

In this section, we explore the beneficial effects of behavioural interventions for children and adolescents with ADHD in relation to parent and child outcomes.

Q1.1 – Do behavioural interventions enhance parental knowledge about ADHD?

Rationale: Many behavioural interventions have a psychoeducational component giving information about the nature of the disorder (Montoya, Colom, & Ferrin 2011). The primary aim of this is to increase parents' knowledge about the nature of ADHD, its possible causes and general course and the treatment options of the disorder. This may be a goal in itself but also a necessary basis for subsequent therapeutic intervention.

Evidence: There is no meta-analysis of the effects of behavioural interventions on parental knowledge of ADHD. One review of the effects of psychoeducation supported its value but also highlighted the poor evidence base and the methodological limitations of studies (Montoya et al 2011). A higher level of knowledge of ADHD has been shown to be related to more favorable parental opinions of behavioural interventions. Enhanced knowledge increases the likelihood of engagement in pharmacological and non-pharmacological treatments (Corkum, Rimer, & Schachar 1999). A recent study has also shown that receipt of psychoeducation may result in lower parental unblinded ratings of ADHD (over two standard deviations) as well as enhanced adherence to ADHD medication ($r = 0.42$) (Bai et al 2015).

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Guidance: Behavioural interventions that educate parents about ADHD may be used to help parents understand more about ADHD and encourage engagement in medication treatment.

SIGN rating *for the level of evidence that psychoeducation*

(i) enhances parents' knowledge about ADHD = ***4***

(ii) *enhances engagement with treatment =* ***1-***

Q1.2 – Do behavioural interventions improve parenting behaviour and the quality of parent-child relationships?

Rationale: An implicit assumption of the behavioural treatment model is that improving parents' behaviour towards their children with ADHD improves the behaviour of children with ADHD. This is also likely to improve the quality of the parent-child relationship more generally (i.e., the positive feelings and attitudes the parent and child have toward one another). Therefore, more appropriately targeted parenting should be a prerequisite for therapeutic effectiveness (Hinshaw et al 2000).

Evidence: Meta-analyses suggest that both blinded measures of parenting behaviour (positive parenting d = 0.63; CI 0.47 - 0.7(ii) negative parenting d = 0.43; CI 0.24- 0.62) and parent self-reports of parenting self-concept (d = 0.37; CI = 0.03 - 0.70) are improved by behavioural interventions (Daley et al 2014). During face-to-face interactions levels of positive parenting (e.g. warmth, reward) are increased and levels of negative parenting (e.g., harshness, criticism) are reduced. The quality of more general parent-child relationship as represented by parent and child feelings and attitudes to one another has only rarely been used as an outcome in behavioural intervention trials. As a result, we know little about whether the child and/or parent attitudes and feelings towards each other improve following intervention. However, there is blinded evidence for reduced child oppositional behaviour (Daley et al 2014; d = 0.31; CI 0.05 – 0.57), which may lead to increased engagement and cooperation from the child towards their parents and, potentially, improved parent-child relationships. Behavioural interventions that have directly tested the impact of intervention on parent's feelings about their relationship with their child (usually termed expressed emotion) do show a reduction in expressed emotion (enhancement of warmth and reduction in criticism) in children with behavioural

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problems (Scott et al 2010). Only one small scale ADHD-specific study to date has explored the impact of behavioural intervention on expressed emotion (Thompson et al 2009). Results showed that while overall expressed emotion was not significantly reduced in the intervention arm compared to treatment as usual, there were significant reductions in parents' negative comments (d = 0.73).

Guidance: Behavioural interventions can be used to improve parenting behaviour and increase parents' sense of self-worth. They may also lead to improvement in parent-child relationships, but there is limited evidence to support this latter aspect.

SIGN rating *for level of evidence showing behavioural interventions improve*

(i) parenting (and parental self-concept) = ***1+***

(ii) the quality of parent-child relationship more generally = 1-

Q1.3 - Can behavioural interventions reduce ADHD symptoms?

Rationale: Parent training interventions for ADHD often focus on reducing coexisting problems and impairments rather than ADHD symptoms (Tarver, Daley & Sayal 2015). These co-existing problems and impairments are often the main reason for referral (O'Connor et al 2015) and the treatment goal for many interventions. Nevertheless, behavioural interventions have also been recommended as a way to reduce core ADHD symptoms (O'Connor et al 2015).

Evidence: Meta-analyses have demonstrated positive effects with moderate (0.67 Fabiano et al 2009) to large (d = 0.87 Van der Oord, Prins, Oosterlaan & Emmelkamp 2008) effect sizes for the impact of behavioural interventions on parental reports of ADHD. Given that parents providing the ratings also received the intervention and were therefore aware of treatment allocation, these ratings could be considered to produce a high risk of bias. . In our meta-analyses (Sonuga-Barke et al 2013; Daley et al 2014) these effects reduced to approximately zero with broad confidence intervals when probably blinded ratings were considered (d = 0.02; CI -0.30 – 0.34). When the probably blind meta-analysis was limited to studies of no/little medication in the comparison arm, the effect remained non-significant (d=0.26; 95% CI=–0.08, 0.60) but the point estimate and confidence intervals do not exclude a small beneficial effect. The probably blinded measures are a mixture of teacher reports and direct observations which in some cases may not be optimal for identifying changes in ADHD behaviours in the home. This pattern does not appear to differ as a function of whether the assessed outcome is inattention or

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hyperactivity/impulsivity or which ADHD presentation the participants have (Webster-Stratton, Reid, & Beauchaine 2011; Hoath, & Sanders 2002; Pfiffner et al 2007).

Guidance: Based on evidence that parent training does not reduce ADHD symptoms when measured by individuals blinded to treatment allocation, it is not presently supported as a way of reducing core ADHD symptoms. However, the effects on parent's reports suggest that these interventions change parental perceptions of their child's behaviour and these could be important even if they don't change actual levels of ADHD. However, there is currently no evidence to support this view.

SIGN rating *for the level of evidence that parent training*

(i) *reduces non-blinded measures of ADHD symptoms =* ***1+***

(ii) Does not have effects on blinded ADHD outcomes likely to be of sufficient size to have clinical value = 1-

Q1.4 Do behavioural interventions reduce co-existing behavioural and emotional problems in children with ADHD?

Rationale: Many behavioural packages were initially developed to treat children with conduct problems (CP) rather than ADHD. Behavioural interventions used with individuals with ADHD continue to focus on reducing these behavioural problems which are very common in these children (Tarver et al 2015).

Evidence: Meta-analyses confirm that behavioural interventions reduce conduct problems in children with ADHD (Van der Oord et al 2008; Fabiano et al 2009). In the EAGG reviews this extended to probably blinded measures, where small to moderate effects (d = 0.31; CI 0.05 - 0.57) have been reported (Daley et al 2014). Few studies have examined the impact of behavioural interventions on emotional problems. One meta-analysis suggests a moderate positive effect on unblinded measures of internalising behaviours in pre-school children with ADHD (SMD -0.48; 95% CI -0.84 to -0.13; Zwi et al 2011), but this was based on just two studies.

Guidance: Behavioural interventions can be used to reduce conduct problems, but there is less evidence that behavioural interventions lead to improved emotional functioning in children with ADHD.

SIGN rating *for level of evidence that behavioural interventions*

(i) *improve behavioral problem symptoms =* ***1+***

(ii) *reduce emotional problems =* ***1-***

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Q1.5 Do behavioural interventions have benefits in terms of social and academic functioning.

Rationale: Children with ADHD often have impairments in social and academic functioning. Targeting ADHD and comorbid symptoms has the potential to enhance social and academic functioning, especially if the behavioural approaches include specific modules that target these deficits.

Evidence: Consistent with other meta-analyses (Van der Oord et al 2008), our meta-analysis demonstrated moderate but significant effects (d = 0.47; CI 0.15 - 0.78) on unblinded parental and teacher ratings of social skills (Daley et al 2014). Trials that report a positive effect typically include a specific social-skills component (Pfiffner & McBurnett 1997). With regards to academic functioning, Daley et al (2014) found small but significant effects (d = 0.28; CI 0.06 – 0.59) from 6 parent or teacher reports (performance ratings and homework problem checklists) and 3 objective measures (actual school grade performance) of academic functioning. Another meta-analysis reports similar results (Van der Oord, Prins, Oosterlaan & Emmelkamp 2008, d = 0.19). Again, studies showing the most benefit often incorporated an academic or organisational skills component often delivered at school (Daley et al 2014; Evans et al., 2016).

Guidance: When adapted to include specialist modules targeting social or academic skills, behavioural interventions may have beneficial effects on social skills and academic functioning.

SIGN rating *for level of evidence that behavioural interventions*

(i) improve social or academic functioning = ***1-***

Summary of Benefits of Behavioural Interventions

Based on current evidence the positive effects on ADHD symptoms reported by parents are not corroborated by independent blinded sources and may reflect a change in parents' attitudes and perceptions about their child with ADHD rather than any actual change in behaviour (Daley et al 2014). This is in contrast to the impact of behavioural interventions on conduct problems where the evidence from independent sources corroborates the view of parental reports. Behavioural interventions may improve academic and social functioning, but the lack of independent blinded measures for either outcome in our meta-analysis (Daley et al (2014) makes the un-blinded improvements difficult to interpret at the meta-analytic

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level. There is also evidence that behavioural interventions enhance parental behaviours towards their children. They increase positive and reduce negative parenting even on blinded measures, which may eventually have a positive effect on future outcomes.

B) Therapeutic content and delivery

In this section, we discuss the evidence relating to which types of behavioural intervention are most effective and how they should be delivered.

Q2.1 - What are the important elements in effective behavioural interventions?

Rationale: Behavioural interventions are generally based on reinforcement and social learning theory. Group-based interventions, grounded in the principles of social learning theory and behavior modification are recommended as interventions for ADHD (NICE 2008) but include a range of different components that may or may not be of value.

Evidence: There are no systematic reviews or meta-analyses assessing the relative value and impact of the different components of behavioural interventions for ADHD. One meta-analysis of programs for children 7 years and younger with more general behaviour problems (Kaminski, Valle, Filene & Boyle 2008) showed that components that aimed to increase emotional communication skills (d = 1.47 compared to d = 0.35 for interventions without this aim), taught parents to use time-out (d = 0.52 compared to d = 0.36 for interventions without this aim), and targeted parenting consistency (d = .59 compared to d = 0.36 for interventions without this aim) were consistently associated with larger effects sizes. However, it is not clear whether these findings would also be true for children with ADHD.

Guidance: Because it is unclear yet what the active components of behavioural interventions are, therapists should implement interventions in the way they were intended to be used and not use component parts of interventions in isolation.

SIGN rating for the level of evidence that therapists

(i) should not use components of interventions in isolation = **4**.

Q2.2 - Are there benefits of behavioural interventions adapted specifically for ADHD compared to more generic behavioural approaches?

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Rationale: At least one behavioural programme has been designed to target underlying features of ADHD – such as self-regulatory and cognitive problems (Sonuga-Barke, Thompson, Abikoff, Klein & Brotman 2006) on the grounds that this will lead to better effects on core symptoms.

Evidence: One RCT (Abikoff et al 2015) has compared a generic parent training approach (Helping the Non-compliant Child; McMahon & Forehand 2003) and an ADHD-specific programme (New Forest Parenting Programme (NFPP); Sonuga-Barke et al (2006)). The specific ADHD approach did not show greater efficacy on child behaviour (ADHD, conduct problems) or parental stress or parenting practices. A second large trial (Sonuga-Barke et al, submitted) also failed to demonstrate superiority of NFPP over a different generic approach (Incredible Years infant programme, Webster-Stratton 2015).

Guidance: ADHD specific programmes are not superior to generic programmes and therefore both approaches should be considered.

SIGN rating *for level of evidence that programmes designed specifically for ADHD*

(i) are no more effective than generic programs is ***1-***

Q2.3 - Is the treatment setting important (i.e., home versus clinic; individual versus group)?

Rationale: Home-based parent training programmes may be more effective than clinic based ones, as the behavioural techniques can be more easily contextualized and individualized. Alternatively, group-based programmes may facilitate the sharing of experiences between parents.

Evidence: There is little available evidence to support one treatment setting (home versus clinic) or delivery structure (individual versus group) over another. General engagement and drop-out rates for group-based programmes for children with conduct problems are high and usually between 25 and 40% (Scott et al 2009; Koerting et al 2013). A general review of parent training programmes concluded that programmes should include home visits to provide tailored support (Moran & Ghate 2005). A recent study comparing home-based individual parent training versus a group based parent training programme delivered in non-home-based settings showed no difference between the two interventions in terms of ADHD or conduct problem outcomes but the home based individual programme was associated with lower levels of participants drop-out and cost less than the group programme

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(Sonuga-Barke et al submitted). In this study cost differences were due to expensive facility costs (crèches, halls and refreshments and travel costs) and higher preparation/supervision and training costs for the group-based approach (Incredible Years).

Guidance: The effects of behavioural interventions do not vary across treatment setting and delivery structure. In considering where and how to deliver behavioural interventions it seems likely that patient preferences and cost of delivery will be the most important factors to consider.

SIGN rating of level of evidence that one setting or mode of delivery

*(i) is not better than another is **1-**.*

Q2.4 - Who should deliver the interventions? What level of training/supervision is necessary?

Rationale: Given the complex nature of many behavioural interventions levels of training and supervision are likely to impact on their success.

Evidence: There is no meta-analytic evidence to answer this question and no studies that have systematically varied the amount of training and supervision. Nearly all RCTs are implemented with highly trained, motivated and skilled therapists under careful supervision. Therefore, the most relevant evidence comes from studies which have looked at the effects of behavioural interventions delivered as standard care. One RCT found that effects were reduced to non-significance when interventions were implemented by randomly selected therapists delivering treatment as part of their everyday caseload compared to specialist therapists working on a clinical trial study (Sonuga-Barke, Thompson, Daley, & Laver-Bradbury 2004). In contrast, another study (Hautmann, Hanisch, Mayer, Plück, & Döpfner 2008) found positive effects on unblinded ADHD symptoms and behaviour problems when behavioural interventions were included in routine care; effects were equal in size to the original efficacy study. A third study (Van den Hoofdakker et al 2007) found positive effects of behavioural parent training delivered as an adjunct to routine care (including pharmacotherapy) by experienced psychologists on unblinded measures of behaviour problems and ADHD symptoms. Authors of these trials highlight the importance of therapist motivation and the need to deliver the intervention with fidelity (as intended) – factors shown to predict outcome of treatment for children at risk of conduct problems (Eames et al 2010).

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Guidance: Effective use of behavioural intervention is likely to require investment in training and supervision to ensure interventions are delivered with fidelity.

SIGN rating *of level of evidence that intervention*

(i) needs to be delivered by well trained and motivated therapists = ***4****.*

Q2.5 Should both mothers and fathers and their children be actively involved in behavioural interventions?

Rationale: The involvement of both parents is predicted to increase consistency of the implementation of strategies and shared understanding of ADHD and lead to better outcomes. Involving children increases the ecological validity of the training setting.

Evidence: In general, fathers have not been included in RCTs of behavioural interventions (Fabiano 2007). In relation to ADHD, only one programme, combining parent training with sports activities, has been specifically designed for fathers and demonstrated small to moderate effects on un-blinded observations of frequency of Total Praise (d = 0.54), and Total Negative comments (d = 0.57) for fathers (Fabiano et al 2012). However, to our knowledge there is no study directly comparing the effects of an intervention delivered to a single parent compared to both parents. With regards to child involvement a review of generic behavioural programmes not specifically targeting ADHD (Kaminski, Valle, Filene, & Boyle 2008) indicates that programmes which encourage parents to practice with their own child during sessions reported larger effect sizes (d = 0.91) than programmes without this treatment component (d = 0.33) although the authors did not directly compare the two sets of effect sizes. This may highlight the potential importance of including practice with the child in the therapeutic process,

Guidance: Despite the lack of direct evidence therapists should still try to include fathers and children in training where practical, but will need to take account of complexity of family composition and overcome barriers to achieve this.

SIGN rating *of level of evidence that parents and children should be involved:*

(i) fathers should be involved in intervention = ***4***

(ii) children should be involved in intervention = ***4***

Summary of evidence relating to therapeutic content and delivery.

High-quality evidence is lacking to help answer most of the questions relating to

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therapeutic context and delivery. There has been little attempt to identify the key elements necessary for effectiveness. Furthermore, based on limited evidence, behavioural interventions seem to be robust to setting and delivery type and specialised interventions do not show advantages over more generic approaches. However, in this regard individual patients and families may prefer a particular form of intervention and this is likely to have an impact on both engagement and outcome. The quality of therapist training and supervision are likely to be important but greater research is required to explore this. Involving fathers and children directly in their own treatment is likely to enhance their value. Choices between different behavioural interventions may ultimately depend on practical considerations and cost.

B) What are the treatment indications and contra-indications?

In this section, we will focus on individual differences that determine who should and should not use behavioural interventions.

Q3.1 - Should behavioural interventions be used only where parents have clear parenting deficits/difficulties?

Rationale: The aim of behavioural parent training is to provide parents with enhanced strategies that they can apply to help raise children with challenging behaviour, it therefore seems logical that it should target parents who lack these additional skills.

Evidence: In the past inclusion in RCTs has been based on children having ADHD and not on a lack of parenting abilities. Improvements in parenting, especially reductions in negative parenting and improvements in positive parenting, have been shown to mediate the relationship between receipt of intervention and change in behaviour problems for children at risk of conduct problems (Gardner et al 2010). However, there is no evidence to suggest that intervention-related improvements in parenting occur only for those families with low pre-existing parenting skills or deficits.

Guidance: Behavioural interventions should continue to be offered to parents irrespective of the absence of dysfunctional parenting.

SIGN rating *of level of evidence that parent training*

(i) should be available to all parents, independent of pre-existing parenting skills = ***4***

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Q3.2 - Is it important to take account of patient and parent preferences?

Rationale: It seems reasonable to assume that patients and parents will be less likely to engage with, or work at, interventions that they either do not want, do not believe work or do not value, and which are not delivered in the way that they would prefer.

Evidence: A recent large study showed that around two thirds of parents of children with ADHD had a preference for individual over group parent training or other alternatives (Wymbs et al 2015). The majority of parents were seeking to feel more informed about their child's problems and to understand as opposed to solve their child's difficulties. About one fifth of parents preferred group-delivered therapy and the same amount preferred a minimal information alternative (i.e., just information). Parents with a preference for minimal information reported the highest levels of depression and had children with the most complex problems. These findings suggest that not all help-seeking parents are looking or willing to engage in behavioural parent training interventions known to be effective. This suggests that services need to consider ways to help motivate parents to engage in behavioural parent training or provide alternative methods of intervention such as child-focused interventions. Gewirtz, Lee, Morrell, & August (2016) found that families accessing mental health clinics (not specifically for ADHD) displayed a clear preference for individual therapy, and those that were able to choose this option were more likely to remain in treatment. This evidence of a preference for individually delivered therapy is at odds with current guideline recommendations in the UK (NICE 2008), which recommends group over individual intervention for ADHD.

Guidance: Parent and patient preferences should be taken into account when planning behavioural interventions, although little is known about the relationship between preferences and treatment outcomes. A range of individual and group-based approaches should be available.

Sign rating *of level of evidence that patient and parent preferences about mode of intervention*

(i) should be taken into account = ***4***

Q3.3 - What are the barriers to initial engagement in behavioural interventions? How might these be overcome?

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Rationale: Parents need to engage with behavioural parent training for it to be effective – but many families are in complex circumstances and non-engagement is often a challenge for services.

Evidence: A qualitative review explored barriers to engagement in parent training programmes from both parental and clinician perspectives (Koerting et al 2013). Barriers identified by parents and clinicians included situational factors (e.g. transport and childcare problems, inconvenient timings), psychological factors (fear, stigma and distrust), lack of awareness or unavailability of programmes and issues with poor interagency collaboration. A second study (Smith et al 2015) explored how to overcome barriers to early behavioural intervention for ADHD from both parent and clinician perspectives. Their results indicated that enhancing parental motivation to change parenting practices and providing an intervention that addressed the parents' own needs was important (e.g. in relation to self-confidence, depression or parental ADHD), in addition to those of the child. Comparisons between the views of parents and practitioners highlighted a need to enhance awareness of parental psychological barriers among practitioners and for better programme advertising generally. However, there are no empirical studies of the effects of removing barriers to engagement on treatment outcome.

Guidance: Clinicians should be sensitive to the concerns of parents and actively try to address barriers to treatment engagement whenever possible.

SIGN rating *of level of evidence that barriers to engagement*

(i) need to be addressed = ***2++***

Q3.4 - Are there parental difficulties that reduce/improve treatment effectiveness?

Rationale: Behavioural parent training interventions use parents as agents of change to help their child. It seems plausible that certain parental characteristics (mental health problems, literacy intellectual abilities or motivation) could disrupt that process.

Evidence: The multimodal treatment of ADHD Study (MTA) group conducted several moderator analyses for their main outcomes (MTA Cooperative Group 1999). In these parental characteristics did not predict treatment outcome (Owens et al 2003). In contrast, Sonuga-Barke, Daley & Thompson, (2002) and Chronis-Tuscano et al (2011) showed that the effects of parent training were reduce by high levels of

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ADHD in mothers. Also, Dawson, Wymbs, Marshall, Moutone & Power (2014) showed that parents at risk for ADHD had particular difficulty maintaining treatment effects in the longer term. In contrast, one study showed no effect of either parental ADHD or depression but did report a moderating role for parental self-efficacy on unblinded ADHD and conduct problems (Van den Hoofdakker et al 2010). The impact of other parental characteristics such as intellectual ability, motivation and literacy on the outcomes of behavioural interventions has not yet been studied systematically.

Guidance: There is little systematic evidence to suggest that behavioural interventions will be less effective with parents with mild to moderate mental health problems, but therapists can consider adjusting delivery to take account of ADHD in parents.

SIGN rating *of level of evidence that parental ADHD*

(i) reduces the effectiveness of parenting training is ***2++***

Q3.5 - Are there family situations where behavioural interventions are contra-indicated?

Rationale: Behavioural interventions could exacerbate existing marital conflict or enhance the burden on already stressed parents.

Evidence: There is no evidence that contra-indicates behavioural interventions for particular families. However, when making referrals to behavioural programmes clinicians should reflect on the fact that family dynamics may be altered by participation in behavioural interventions. Chronis, Chacko, Fabiano, Wymbs & Pelham (2004) reviewed evidence that parents participating in behavioural interventions who displayed clinically significant levels of marital dissatisfaction at pre-treatment tended to direct aversive behaviours towards their spouses (e.g., negative feedback, argumentativeness, noncompliance, ignoring) when their child was misbehaving.

Guidance: There is no evidence to suggest that behavioural interventions are contra-indicated if specific family problems are present. However, therapists should be sensitive to the potential impact of behavioural interventions on family dynamics.

SIGN rating *of level of evidence that in families with poor functioning*

(i) parent training should not be used due to risk of negative effects of family functioning = ***4***

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Q3.6 - Does disorder severity and comorbidity reduce the effectiveness of behavioural intervention?

Rationale: More symptomatic and complex ADHD cases may have more deep-rooted and complex causes which could make behavioural approaches less effective.

Evidence: It is surprising how little is known about the effects of ADHD severity or comorbidity on treatment efficacy as no studies have sought to randomise participants on these factors. The MTA study found no evidence of the effect of symptom severity on psycho-social treatment outcome (Owens et al 2003). In contrast, Hautmann et al (2008) found that the most severely impaired children profit the most from behavioural interventions in terms of externalising behaviour improvement, although these findings were for a general externalising behaviour disordered group. With regard to comorbidity, a meta-analysis found that the presence of conduct disorder reduced the impact of behavioural interventions on unblinded ADHD measures (Lee et al 2012). In the MTA study comorbidity of ADHD with anxiety was associated with better outcomes for behavioural interventions on unblinded ADHD measures (Owens et al 2003). Number of comorbidities (anxiety/depression or oppositional defiant/conduct disorder) was negatively related to behavioural intervention efficacy in another study with children with no comorbidity or just one comorbidity displaying a superior response to behavioural intervention, compared to those with two or more (Van Den Hoofdakker et al 2010). A recent study, comparing a specialized ADHD intervention and a generic intervention developed specifically to treat non-compliance, found that the latter was generally more effective at treating conduct problems where individuals had comorbid ADHD and conduct problems (Forehand et al 2016).

Guidance: Behavioural interventions can be used for children with ADHD irrespective of the severity of their symptoms. Comorbidity may alter the effects of behavioural interventions but these are not contra-indicated for children with comorbidity.

SIGN rating *of level of evidence regarding symptom severity and comorbidity that*

(i) symptom severity does not impact on treatment efficacy = ***2++***

(ii) comorbidity does impact on treatment efficacy ***1-***

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Q3.7 – Is early intervention more effective? Does it reduce long-term risks of ADHD?

Rationale: Larger effects of behavioural interventions may be expected in preschool children when neuro-plasticity is greatest, before either the full-blown disorder is established or the development of comorbid disorders has occurred and while parent-child relationships are still relatively intact.

Evidence: RCT's have focused mainly on preschool and primary school aged children. Most meta-analyses do not report a significant impact of age on outcomes of behavioural interventions (Hodgson, Hutchinson & Denson 2012; Lee et al 2012; Mulqueen, Bartley & Bloch 2013). However, our recent meta-analysis (Daley et al 2014) found larger effects in younger children on unblinded ADHD measures ($t = -2.63$, $p = 0.03$), conduct problems ($t = -2.46$, $p = 0.05$) and positive parenting ($t = -2.63$, $p = 0.03$). With regards to long-term effects, significant treatment effects are maintained but their magnitude declines (Lee et al., 2012). However, evidence for these longer-term benefits may be contaminated by participants' exposure to other treatments during the follow-up period (Jones, Daley, Hutching, Bywater. & Eames, 2008). Given this, there is currently no evidence demonstrating that early intervention with behavioural approaches reduces the long-term risk of ADHD diagnosis or associated comorbid disorders.

Guidance: Clinicians are encouraged to commence behavioural interventions as early as possible before the child's ADHD becomes associated with more severity, comorbidity, anti-social tendencies and school failure. Behavioural interventions should also continue to be offered to older school aged children as well.

SIGN rating *for level of evidence that early intervention*

(i) has a special value = ***1+***

(ii) *reduces the long-term risk =* ***4***

Summary in relation to indications and contra-indications.

There are currently no clear contraindications for the use of behavioural interventions for children and adolescents with ADHD. Research into predictors of treatment outcomes is sparse and inconsistent. More generally, clinicians are advised to listen to parents' thoughts and opinions and to reflect on whether parents are ready to engage with behavioural interventions before commencing treatment. Comorbidity

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may alter the effects of behavioural interventions but these are not contra-indicated for children with comorbidity. Early intervention, where possible, is encouraged.

D) Relationship to other non-pharmacological treatments

Q4.1: Is there value in combining parent-focused interventions with school-focused or patient-focused behavioural interventions?

Rationale: Behavioural interventions often show limited generalizability in randomized controlled trials perhaps because they are often delivered by parents at home or in the clinic (Daley et al 2014). Adding school-based, and child-focused interventions may help to enhance generalisation to school-settings.

Evidence. A recent meta-analysis (Chan, Fogler & Hammeress 2016) of treatments for adolescents with ADHD has demonstrated that behavioural interventions (which were mostly adolescent focused but were sometimes augmented with teacher and/or parent components) were associated with robust (Cohen d range, 0.51-5.15) improvements in mostly parent rated academic and organizational skills, such as homework completion and planner use. Although studies have shown the effectiveness of integrated school/home programmes compared to control groups (Pfiffner et al 2007; Power et al 2012; Ostberg et al 2012), only one study has systematically assessed the additive value of school intervention (and a child skills training) to parent training in a sample of children with the inattentive subtype of ADHD (Pfiffner et al 2014). Results showed superior effects of integrated home-school treatment as compared to parent training alone on unblinded teacher-reported inattention, organizational skills, social skills, and global functioning at post-treatment. However, at follow-up during the subsequent school year, differences in teacher-reported outcomes were not statistically significant.

Although several treatment studies have combined child-focused and parent focused elements (e.g. Abikoff et al 2013; Webster-Stratton et al 2011; Pfiffner et al 2007) and reported positive results, few studies have systematically assessed the additional value of a child-focused element to parent training. Some early studies combined parent training with child-focused treatment (targeting child self-control) and assessed the separate and combined effects. In these studies, there was no evidence for additive effects of child-focused problem solving treatment on ADHD and conduct problems (Horn et al 1990; Horn et al 1987).

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Guidance: Adding school-based intervention may hold promise for the inattentive presentation/subtype of ADHD. There is little current evidence for combining child-focused problem-solving treatment with parent training.

SIGN rating *of evidence that adding further*

(i) school-based elements to parent training is advantageous = ***1-***

(ii) child- focused elements is advantageous = ***1-***

Q4.2 - Can behavioural interventions be combined with cognitive training and neurofeedback to improve outcomes?

Rationale: Adding interventions that are more directly targeted at underlying deficits in cognitive mechanisms may enhance the benefits of behavioural interventions.

Evidence: Recent meta-analyses have questioned the efficacy of both cognitive training and neuro-feedback as treatments for core ADHD symptoms in terms of data from blinded outcomes (Cortese et al 2015; Cortese et al 2016). Two recent studies assessed the separate and combined effects of cognitive training and parent focused behavioural training. Steeger et al (2015) found no benefit of the combination on unblinded measures of ADHD. Maleki et al (2014) found some evidence of benefits of combined cognitive training and parent training on unblinded outcomes (effect sizes not available) compared to parent training or cognitive training alone, however this study had a number of methodological limitations. To date, no RCTs have assessed the added combined effects of neuro-feedback and behavioural interventions in children/adolescents with ADHD.

Guidance: There is currently no reliable evidence to support the efficacy of working memory training or cognitive training for ADHD or the combination of behavioural and cognitive or neuro-feedback interventions.

SIGN rating *of level of evidence regarding combinations with cognitive interventions that*

(i) *working memory training does not enhance the effects of parent training* ***= 1-***

(ii) neurofeedback does not enhance the effects parent training = 4

Q4.3 - Should behavioural interventions be combined with treatment for parents' mental disorders/psychiatric diseases?

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Rationale: Given the fact that the parent is usually the agent of change in behavioural interventions, parental psychopathology and psychological states may impact on the effectiveness of behavioural interventions. In these cases, combining treatment for the child with treatment for the parent may enhance both child and parent outcome.

Evidence: In our recent meta-analysis, no effect of behavioural interventions was found on parental mental health (Daley et al 2014). Some studies have compared additive effects of parental treatment to parenting interventions.

Three different domains of parental psychopathology and functioning have been addressed; depression (Chronis-Tuscano et al 2013), parental stress and lack of social support (Chacko et al 2012; Rajwan et al 2014), parental ADHD (Jans et al 2015). With regards to the additive value of CBT in combination with regular BPT for mothers with at least mild depressive symptoms, Chronis-Tuscano et al (2013) showed the additive value of combining treatment for parental depression and child ADHD on child, parenting and parental outcomes (child impairment, family functioning, parental depression) at 3 months follow-up. Another study showed that enhanced parent training (enhanced to target parental stress and coping but also social skills training for the child) reduced drop-out, significantly raised engagement and social support for parents, as compared to standard behavioural treatment (Chacko et al 2012), although these benefits were too small to be considered clinically significant (Rajwan et al 2014). Additional multi-modal treatment of maternal ADHD did not enhance effects of a subsequent behavioural parenting intervention on the child's externalising problems; although it significantly reduced unblinded reports of parental ADHD (Jans et al 2015).

Guidance: Identifying and addressing mental health problems such as depression in parents of children with ADHD children is important. Although potentially beneficial for the parents, it may not increase the effectiveness of behavioural interventions or outcomes for their children, with the potential exception of treatment of parental depression.

SIGN rating *of level of evidence that behavioural interventions with treatment for parental mental health*

(i) is beneficial = ***1-***

Summary of findings for non-pharmacological treatment combinations:

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There is very little evidence that adding other non-pharmacological interventions to behavioural interventions improves outcomes. There are positive effects of additive school-based interventions for the inattentive subtype. Targeting parental depression may enhance the effects of behavioural interventions.

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Discussion

We have used a question and answer format to address questions about behavioural intervention most typically parent training for the treatment of ADHD that we feel are of particular significance for practitioners and policy makers. We have based our answers, as far as possible, on empirical and peer reviewed evidence. For every question, we have provided clinical guidance which we hope will be of practical use. We conclude that behavioural interventions have beneficial effects on conduct problems and parenting where evidence from independent sources corroborates parental report. Effects on ADHD symptoms, academic and social functioning are more difficult to interpret as the lack of evidence from independent sources does not rule out the possibility that reported improvements are merely changes in informant perception rather than actual behaviour**.** The essential elements of behavioural interventions are, as yet, unknown. What is known is that specialised ADHD behavioural interventions are not more effective than more generic behavioural programmes, but if delivered in an individual format may be more cost effective. Including children in the intervention process may also enhance outcomes. There are few specific indications or contraindications for behavioural interventions but considering whether parents are physically or psychologically able, and ready to engage and implement behavioural interventions may be clinically important. There is very little reliable evidence that adding other non-pharmacological interventions to behavioural interventions has any benefits.

This review does highlight a number of important gaps in the current evidence base. Firstly there is a need to enhance the number of studies that use blinded or independent outcomes across multiple measures, but especially for ADHD, academic functioning and social skills, to explore whether proximally reported improvements reflect actual improvement, or merely changes in informant perception. In doing this it will be important to be able to control for the influence of rater bias and context on differences between Most Proximal and Probably Blinded informants reports. Secondly, additional work is required to identify mediators and

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moderators that can help better understand the mechanisms and active treatment components which are associated with improvement as well as identifying which patients benefit the most. Improving our understanding in this area could allow clinicians to tailor the delivery of intervention to families and children who will benefit the most.

Our guidance is not without its limitations and constraints. Our review of evidence is not based entirely on systematic reviews and meta-analyses. However, we have taken a systematic approach to the synthesis of the evidence where possible, focusing on recent meta-analyses and RCTs. Second, the interpretation of the evidence and the subsequent clinical recommendations are the views of the membership of the EAGG, this applies to all questions but is particularly influential when evidence is weak or inconclusive. In such cases we have taken a pragmatic approach based on the principles and logic of good clinical practice referenced against the expert clinical opinion of EAGG members to guide our recommendations and have used SIGN evidence ratings to highlight where recommendations are based solely on expert opinion. Third, we have had to give the guidance with almost no reference to the relative financial costs and benefits of the various options. This of course is a major handicap for while we might consider that a certain approach is optimal in terms of efficacy it may be prohibitively expensive to implement in routine practice or costs may vary considerably between different healthcare settings. The questions relating to the mode of delivery, the involvement of fathers, the quality of training and supervision and the integration with adjunct therapies are especially likely to be affected by such considerations.

Our hope is that, in the future, stronger empirical evidence will guide clinical recommendations in a more direct way based on clearer evidence to guide day to day clinical practice.

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Key Practitioner message
On the basis of current evidence, that takes into account whether outcomes are blinded, behavioural intervention cannot be supported as a front-line treatment for core ADHD symptoms. However, there is evidence on probably blinded outcomes that behavioural interventions reduce conduct problems in children with ADHD and enhance parenting in parents of children with ADHD. Specialised ADHD behavioural interventions do not appear to be more effective than more generic behavioural programmes. There are few contraindications for behavioural interventions. There is no reliable evidence to date to suggest that adding other non-pharmacological interventions to behavioural interventions has benefit.
Areas for future research
There is a need to enhance the number and quality of studies that use blinded or independent outcomes especially for core ADHD symptoms, but also for co-morbid impairment domains. More research on moderators of outcome is required to help understand for whom behavioural interventions work best. More research on mediators of outcome is required to identify underlying mechanisms of action for behavioural intervention.

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