Psychological treatment strategies for challenging behaviours in neurodevelopmental disorders: what lies beyond a purely behavioural approach?

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Abstract
Purpose of review: Challenging behaviour (CB) shown by individuals with neurodevelopmental disorders (NDDs) has a major negative impact. There is robust evidence for the efficacy of treatments based on applied behaviour analysis. However, such approaches are limited in important ways – providing only part of the whole solution. We reviewed the literature to provide an overview of recent progress in psychological treatments for CB and how these advance the field beyond a purely behavioural approach.

Recent finding: We identified 1029 papers via a systematic search and screened for those implementing a psychological intervention with individuals with NDD (or caregivers) and measuring the potential impact on CB. Of the 69 included studies published since 2018, more than 50% implemented a purely behavioural intervention. Other studies could generally be categorised as implementing parent training, meditation, skill training or technology-assisted interventions.

Summary: Greater consideration of the interplay between behavioural and non-behavioural intervention components; systematic approaches to personalisation when going beyond the behavioural model; mental health and broad social communication needs; and models that include cognitive and emotional pathways to CB; is needed to advance the field. Furthermore, technology should not be overlooked as an important potential facilitator of intervention efforts.

Keywords
Challenging behaviour; Neurodevelopmental disorders; Psychological treatment; Behavioural management

Introduction

Aggression, self-injury, destruction of property, temper outbursts and other such behaviours that challenge show heightened prevalence in people with neurodevelopmental disorders (NDDs) [1]. Such challenging behaviour (CB) has a major negative impact on the lives of people with NDDs and their family members, being associated with increased family health problems and cost of care [2-5]. Stakeholder groups repeatedly identify CB as a priority target for research, not least because of the limits it places on functional independence [6-7]. Furthermore, a large proportion of referrals to healthcare services are linked to CB [8]. And the presence of CB limits educational opportunities [9].

Unfortunately, in treating CB, there are important concerns about the use of psychiatric medication [10-12]. On the other hand, there is a robust and long-standing evidence base for the efficacy of approaches based on the personalised application of behavioural principles to identify environmental contingencies that contribute to the maintenance of CB and manipulate these to bring about a desired behaviour change [13]. Applied behaviour analysis (ABA) and positive behavioural support (PBS) both take this approach [14]. Despite this robust evidence base for behavioural approaches, they are limited in important ways – notably, they provide limited scope for the consideration of cognitive and emotional processes. This is particularly relevant when we consider the wide range of CBs and associated contexts. Temper outbursts for example, appear to have a strong emotional component. Often, in their very nature, they preclude environmental consequences that would satisfy a need created by the environmental antecedents [15-18]. Thus, behavioural approaches, whilst an effective and necessary part of the solution to CB, cannot provide the whole solution for everyone. With this in mind, we reviewed the recent literature on psychological interventions for CB in people with NDDs. Our aim was to provide the reader with an overview of recent progress in the area and use this to identify recommendations that will help to advance research and practice.

Materials and methods

We systematically searched Web of Science, Pubmed, Psychinfo and Medline for articles published between January 2018 and August 2019. We selected multiple search terms referring to the core inclusion criteria – interventions, NDDs and CB – and searched for these in title, abstract and keywords (full search strategy available in supplementary materials).

Following duplicate removal, we identified 1029 articles, which we screened for inclusion. We included peer reviewed articles that reported on an intervention in an NDD population and measured its potential impact on CB. We defined CB as behaviour that can be harmful to the individual or to those around them. Articles which related only to medical interventions were excluded. Two researchers screened the articles independently, disagreements were discussed and where necessary resolved by discussion with a third researcher. The screening process led to the inclusion of 69 papers (see supplementary materials).

A tool for risk of bias assessment was created by our research team for a previous systematic review of interventions, in line with published guidelines ([19-20]; supplementary materials). Two reviewers conducted the risk of bias assessment of each article independently (mean Kappa inter-rater reliability was acceptable – 0.56), conflicts were discussed and if necessary, resolved by discussion with a third researcher.

Results and discussion
The included papers are summarised in Table 1 and corresponding risk of bias in Table 2.

Behaviour approaches
Thirty-six papers applied ABA (n=28) or PBS (n=8), with a US dominance for ABA and UK for PBS. ABA broadly focused on personalised interventions for children (n=23) with a primary diagnosis of ASD (n=22). PBS largely targeted staff training in adolescent/adult supported living services for intellectual disability (ID).

Behaviour strategies mostly demonstrated reductions in targeted behaviours. Where maintenance or follow-up was reported, improvements were stable [64, 84, 98, 81, 71, 74, 94]. Twenty-seven papers employed a case series (1-4 participants in all but 1 study) and five a multiple-baseline design (3-7 participants). Interestingly, Hassiotis et al. [99] employed the most robust sample, and was the only study not reporting improvements – perhaps in part reflecting a tension between wide scale application of behavioural strategies and the resource intensive requirements of effective personalisation.

ABA studies focused on reinforcer (R+) manipulation (n=9), functional communication training (FCT) (n=8), function-based interventions (n=4), multiple schedules fading (n=2), demand manipulation (n=2), previously developed intervention programmes (n=2), sleep manipulation (n=2), pivotal response parent training (n=1). In practice however, such strategies are combined within evolving packages, the efficacy of which is not examined in the present research.

PBS studies have administered strategies more likely to yield scalable impact. Five studies integrated training in residential services, one applied peer-mediation to improve engagement in physical activity [83] and one describes the impact of a community PBS team [82]. Like the ABA papers, Lee et al. [82] integrated augmented communication to provide a more holistic approach to the underlying motivations of behaviour difficulties. However, the limited detail given on training makes comparison difficult.

Cost-benefit is pertinent in understanding likely impact of an intervention. Reporting of contact hours across the current studies is varied, with unclear aggregates. As such, limited conclusions can be drawn about the comparative effectiveness. Behavioural approaches are clearly resource intensive. Despite this, only two of the studies measured social validity or feasibility [83, 94], which is essential for the potential scalability and real-world impact to be determined.

Only six interventions targeted home as the primary setting. A critique of behavioural strategies is their fidelity when transferring to uncontrolled environments due to the reactive skills required. For example, Saini et al. [66] found that destructive behaviour increased when FCT was transferred home. On the other hand, novel approaches effectively implemented functional behaviour [56] and communication training [70] at home via telehealth facilitation.

Overall, these studies continue to support the use of behaviour strategies. However, the success is heavily linked to the ability to personalise such strategies. For this reason, these interventions are limited in their impact due to the burden such methods of personalisation place on resources. Difficulties in wide scale application also follow from the dependency upon response consistency in less controllable environments by less experienced deliverers. Thus, going beyond a purely behavioural approach is clearly important.

Parent training
Ten studies involved parent-training interventions, all of which produced benefits in CB shown by children. Three of these involved training in strategies exclusively based on behavioural principles.

Two were delivered individually [22-23] and targeted a specific profile of contextualised CB. One delivered training to groups of parents [24] and showed evidence of context-specific effects. These studies highlight a further limitation of purely behavioural approaches – since skills are not imparted to children, without in depth generalisation training, gains are often limited to the specific settings subject to the intervention.

Addressing this limitation, the other parent training studies have drawn on the behavioural model alongside other models. Four of these involve parent-child interaction therapy (PCIT [25-28]). PCIT was developed for typically developing children and involves coaching parents to interact with children. It draws on attachment theory and social learning theory, which itself draws heavily on the behavioural model [29]. These four studies incorporate several important risks of bias, but they represent a growing application of PCIT to NDD populations. Indeed, a systematic review of PCIT between 2000 and 2016 identified 18 studies examining ADHD or ASD samples [30]. No quantitative synthesis was attempted but the studies all reported improvements in parent-rated child CB, alongside wider perceived benefits for parents. However, none of the studies were controlled, meaning that the efficacy of PCIT over and above any other form of regular contact with parents (or indeed over a purely behavioural approach), could not be demonstrated. This underscores an important gap in research on interventions that combine behavioural techniques with those based on other models – we know little about the relative contributions of the component parts to overall success, which makes it challenging to understand which approaches are likely to be most effective for which individuals, and at which time.

In a step towards filling this gap, Ollendick et al. [86] conducted a large randomised controlled trial with families with children with oppositional defiance disorder, comparing a behavioural based parent training programme with one in which parents are trained to teach children interpersonal problem-solving skills (CPS). Whilst both programmes were associated with reductions in CB, neither could be judged more effective. In this example however, since CPS does not include a behavioural component, we cannot determine whether the interpersonal problem solving would have additional benefit over and above the behavioural techniques. In future, we need more large randomised controlled trials that compare different parent training programmes – which have been mapped systematically to allow us to judge the relative benefits of specific components – and examine relationships with individual differences in family characteristics.

Meditation

Seven studies addressed interventions broadly classified as meditation, including mindfulness, yoga and deep breathing. Two of these studies [31-32] involved mindfulness training for parents of individuals with NDDs, both evaluated using pre-post uncontrolled designs. Whilst Jones et al. implemented purely mindfulness training, Singh et al. combined this with PBS training. Furthermore, the Singh et al. intervention was followed over 30 weeks – the Jones et al. only 8. Both studies report improvements in parent outcomes, for example perceived stress. However, only Singh et al. report improvements in CB shown by individuals with NDDs. Thus, whilst mindfulness training may have benefits for parents, potential positive effects on CB appear less clear. In this context, the combined training approach seems sensible. However, controlled trials are needed to evaluate the potentially additive benefit of the mindfulness component. Given the protracted course of improvements in the Singh et al. study, careful consideration must be given to intervention duration.

Mindfulness training (including yoga) for individuals with NDDs is assessed in four studies. Two moderately sized studies assessed the effects of such training in children with ADHD [33-34]. Although relatively high-quality studies in terms of sample size and inclusion of a randomised control procedure, risk of bias remained substantial given a lack of blinding and reliance on subjective informant report. Both however, reported apparent mindfulness mediated benefits on CB. On the other hand, two smaller studies examining adolescents/adults with a NDD including some level of ID [35-36] present a more mixed picture. Overall, there appeared to be more potentially beneficial effects of mindfulness on CB in individuals with greater intellectual functioning. Similarly, in a study applying deep breathing, on its own this was only effective in reducing CB of an adolescent without ID [37]. Thus, although meditation training for individuals with NDD appears to hold some promise in the treatment of CB, ensuring a strong match between the training and the individual's understanding may be of critical importance. At present the literature is missing a systematic approach to tailoring meditation-based interventions to individual needs. Careful application of an intervention mapping approach (e.g. [38]) may facilitate this.

Skill training

Six studies involved interventions that ultimately aimed to act by imparting skills to individuals with NDDs. One of these, one [39] is a randomised controlled trial that met our inclusion criteria because reduction in aggression was a secondary outcome – reductions in symptoms of depression were the primary outcome. We view the study as an important reminder that underlying mental health issues can precipitate CB in people with NDDs – assessment for and treatment of any mental health issues is an essential component of effective treatment for CB.

Another study (single case) [40] applying the assisted communication technique the Picture Exchange Communication System [41] serves to further emphasise the critical role for communication in the treatment of CB. Behavioural approaches place central importance on the maintaining role of impaired communication in CB. However, the focus is limited to communicating needs that are otherwise conveyed via CB. A systematic review of 56 studies published until 2016 [42] reported on use of touch screen speech generation devices used with individuals with NDDs. Only a tiny minority of the studies facilitated any form of communication outside one or two direct requests. With such devices, technology is not the limiting factor and we must be careful that in the pursuit of effective treatment for CB, we do not ignore the individual's wider social position. Indeed, providing an individual with the means to communicate a specific request can have a rapid impact on current CB, but maintenance of such impact may be much more problematic [43] – increasing the individual's wider communicative skill on the other hand may produce slower gains that are easier to maintain.

The remaining skill training studies lie at different positions in a continuum between targeting specifically identified underpinnings of CB and targeting general capacities which may ultimately support behaviour management. At the specific end, one study [44] that also drew heavily on behavioural principles, focused on CB precipitated by transitions and taught children different sets of rules to define expected classroom behaviour at different times relative to transitions. Not as close to the specific end, two studies trained children with ADHD in emotion recognition and regulation, since poor anger coping was conceptualised as being related to CB [45-46] (although both also included behavioural parent training). And at the general end, one study capacitated staff to train adults with ID in self-management [47]. Only this last study failed to show reductions in CB, but instead showed improvements in independence and self-reliance. We cannot make direct comparisons between these studies due to the range of designs and risk of biases. However, it

seems possible that models which describe more specific pathways to CB ultimately allow more efficient intervention development. This further emphasises the need for more modelling of CB with consideration of specific cognitive and emotional processes, which may ultimately constitute skill training targets.

Technology-assisted

Five studies employed a form of technology in the treatment of CB. However, all of these were small single case/ case series, including a total of 11 participants [48-52]. These studies were early stage research with, in general, concomitant low methodological rigour. Technology was applied in a range of ways, including delivery of behavioural programmes [48, 51] and scaffolding of cognitive and social skills [49, 50, 52], with associated reductions in CB in all cases. Given the benefits of digital technology in intervention settings and rapidly growing digitalisation of our society [53], this relatively small pool of technology-based interventions for CB is somewhat surprising. We have recently conducted a comprehensive systematic literature review of digital interventions for emotion regulation and social cognition skill training in children and adolescents [54]. More than 65% of the studies reported on the use of digital technology in an NDD population. This suggests a stark disconnect between the application of technology to interventions in NDD populations in general, and its application to interventions for CB. At least in part, this may reflect the heavy reliance on the behavioural model in CB treatment, and relative scarcity of complementary theoretical models that maintain the same level of acknowledgement of idiosyncrasy and environmental specificity, whilst also considering cognitive and emotional factors that contribute to the expression of CB. Our own work has illustrated that even when relatively unexplored models describe a role for cognitive/ emotional processes in pathways to CB [16], new possibilities for intervention can be identified, which provide the opportunity to develop technology that may provide effective treatment [55].

Conclusions

Several psychological interventions combine behavioural approaches with other components. More research is needed to better elucidate the contributions of the different components to intervention success, in a manner that is sensitive to individual differences. Indeed, in interventions based on non-behavioural approaches, greater attention is warranted on tailoring treatment to individual characteristics. Mental health and general social communication needs should be carefully considered within a CB intervention context. The development of more models that consider cognitive, emotional and behavioural processes in pathways to CB should be encouraged as these may result in efficient routes to psychological intervention development. Finally, technology should be considered as an important potential facilitator of intervention efforts.

Key points (3-5 bullet points):

- The recent psychological intervention for CB literature has been dominated by applied behaviour analysis but attempts to go beyond this have included parent training, meditation, skill training and technology assisted approaches.
- In approaches that go beyond a behavioural approach, elucidation of components drawn from different models; and systematic consideration of personalisation is required.

- Models that consider idiosyncratic cognitive and emotional factors in pathways to CB should be considered, as potentially efficient routes for psychological intervention development.
- Modern technology is an important potential facilitator of intervention efforts for CB, which appears to have been largely overlooked.

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Table 1: Summary of papers included. Abbreviations: ABA (applied behaviour analysis), DRO (differential reinforcement of other behaviour), FCT (functional communication training), hrs (hours), mins (minutes), NCR (non-contingent reinforcement), PBS (positive behavioural support), ss (sessions), yrs (years), ppt (participant), R+ (reinforcement), RCT (randomised controlled trial), non-RCT (non-randomised, controlled trial).

[Reference] author; year; country	Sample size; design	Gender; age range; mean age; diagnosis	Intervention strategy	Setting	Outcome measure summary	Results summary
[56] Monlux, 2019; US	n = 10; Case series	Males, 3-11 yrs, mean age=7.51 yrs, Fragile X syndrome	**ABA** (telehealth): Parent-led FCT and extinction training	Home; 25 ss over 25 hrs	Observation of CB by researchers - any behaviour that damages self/ others/ the environment	Reduction in CB by 78-95% after 12 weeks of treatments
[57] Briggs, 2017; US	n = 1; Case study	Female, 12 yrs, ASD, mild ID	**ABA**: FCT, multiple schedule FCT, chained schedule FCT	Clinic; 12 ss over 1 hr	Observation of the frequency of destructive, aggressive and self-injurious behaviour by 2 researchers	Both multiple schedule FCT and chained schedule FCT reduced CB to near-zero
[58] Kelley, 2018; US	n = 3; Case series	3 males, 3-5 yrs, mean age 4.3 yrs, ASD	**ABA**: Differential R+, extinction	Clinic; 35-90 ss over 2 hrs 55 mins - 7 hrs 30 mins	Observation of compliance and aggression by researchers	Aggression decreased to zero and compliance increased to 100%
[59] Mitteer, 2019; US	n = 2; Case series	Males, 7 yrs, ASD	**ABA**: FCT	Clinic; 120 ss over 10 hrs	Observations of CB by researchers; parent report questionnaire on CB	Reductions of CB to near zero
[60] Courtemanche, 2018	n = 3; Multiple baseline	Males, 28 yrs, 38 yrs, 46 yrs, mean age: 37.3 yrs. ASD, ADHD	**ABA**: Presentation order of preferred items	Home; 9ss over 1 hr 30 mins - 6 hrs	Researcher observation of self-injurious behaviour	For all 3 participants, antecedent manipulations decreased the rate of SIB; however, operant contingency values (measure of temporal distribution) did not change
[61] Planer, 2018; US	n = 3; Case series	Males, 9-12 yrs, mean age = 10.3 yrs, ASD	**ABA**: Pairing of low with high compliance demands	School; 40-65 ss over 10 hrs - 16 hrs 15 mins	Observation of compliance on low preference tasks by researchers	Compliance in low preference tasks increased
[62] Haq, 2018; US	n = 2; Case series	Males, 10 yrs, ASD	**ABA**: FCT, discrimination training	Home; 35 ss over 2 hrs 55 mins - 5 hrs 50 mins	Researcher observation of challenging behaviour	Reductions in CB with FCT; maintained following discrimination training

[63] Lugo, 2018; US	n = 1; Case study	Female, 4 yrs, ASD	**ABA**: Concurrent-chains arrangement (pre-ss pairing, free play or immediate onset of discrete trial instruction)	Clinic; 135 ss; duration unclear	Researcher observation of negative vocalizations - partial interval recording	Press pairing condition preferred across trials. Negative vocalisations decreased to zero after first concurrent-chain ss
[64] Stevenson, 2019; US	n = 3; Multiple baseline	Males 16-18 yrs, mean age = 16.66, ASD (1 ppt also has severe ID, hearing and visual impairment and seizure disorder)	**ABA**: Combination of function-based intervention and crisis intervention	School; 45 ss over 7 hrs 30 mins	Observation of CB by researchers	Reductions in CB for all participants, which were maintained over 6-12 weeks
[65] Bloom, 2018; US	n = 3; Case series	Males, 6-8 yrs, mean age = 7 yrs, ADHD, ODD, Asperger's	**ABA**: Delayed demands followed by escape extinction	Clinic; 36-50 ss over 9 hrs - 12 hrs 30 mins	Observation of CB and compliance by researchers	Delayed tasks caused more CB than tasks that were unavoidable from the onset, however, escape extinction decreased CB and increased compliance
[66] Saini, 2018; US	n = 4; Case series	75% male, 7-8 yrs, mean age = 7.75 yrs, ASD (1 also ADHD, 1 also Down syndrome)	**ABA**: therapist then parent-delivered FCT	Multiple - Home and Clinic; duration unclear	Researcher observations of destructive behaviour	Destructive behaviour decreased with functional communicative setting in clinic but 3 of 4 ppts showed increased CB when then retested in home setting
[67] Schreck, 2018; US	n = 1; Case study	Female, 12 yrs Genetic-Mucopolysaccharidosis type IIIA	**ABA**: Several techniques to coincide with the function of the various behaviours at different times	Multiple - Home and Clinic; 28 hrs per week for first year then 40 hrs per week for the remaining 8 yrs	Longitudinal case description - Researcher observation of pica, non-compliance, temper outbursts, and hand mouthing	Reduction in CB

[69] Lambert, 2018; US	n = 1; Case study	Female, 7 yrs, PWS	**ABA**: Differential R+	Clinic; ~ 51 ss over 112 hrs	Behavioural observation: Latency of food stealing behaviour by 2 therapists	Reduction in food stealing
[70] Benson, 2019; US	n = 2; Case series	Males aged 5 and 8 yrs, mean age = 6.5 yrs, Multiple-cerebral palsy, limited ambulation, ASD	**ABA:** telehealth	Home; 19 - 41 ss of FCT	Observation of self-injurious behaviour per mins by researchers	Levels of self-injurious behaviour decreased to near zero
[71] Sandberg, 2018; US	n =3; Case series	33% Male, 4-8 yrs mean age = 6.3 yrs, ASD	**ABA**: Bedtime fading with response cost	Home; Weekly ss for 8 weeks, duration unclear	Parent report questionnaires on bedtime resistance (including aggression); and on other sleep behaviours	Bedtime resistance improved, with no aggressive behaviour; sleep behaviour improved
[72] Fisher, 2018; US	n = 4; Case series	75% Males, 3-16 yrs, mean age = 8.5 yrs, ASD	**ABA**: FCT	Clinic; 60 ss over 5 hrs	Observation of CB & functional communication response researchers	Refinements in FCT were successful in decreasing the resurgence of destructive behaviour during an extinction challenge
[73] Cariveau, 2019; US	n = 1; Case study	Male, 8 yrs ASD	**ABA**: Differential R+, response cost	Clinic; 200 ss over 33.33 hrs	Observation of aggressive behaviour	Reduction of aggression and increased tolerance for interruptions
[74] Newcomb, 2018; US	n =1; Case study	Male, 13 yrs, ASD	**ABA**: NCR	Multiple - school, home; 55 school days over 330 hrs	Aggression	Rates of aggression reduced following intervention
[21] Herman, 2018, Ireland	n = 1; Case study	Male, 4 yrs ASD	**ABA**: most to least prompting; escape extinction; differential R+; high probability instruction sequence	School; 8 ss over 8 hrs	Observation of time spent dropping to floor by researchers	Reduced dropping to floor
[75] Muething, 2018; US	n = 4; Case series	75% males, 5- 14 yrs, mean age = 10.5 yrs, ASD	**ABA**: FCT, delayed R+ of mands	School; 15 ss over 1.25 hrs	Observation of behaviour by researchers	Reduction in CB with FCT; increase in response variability for 3 ppts with delay of R+
[76] Verriden, 2019; US	n = 4; Case series	75% males, 6-14 yrs, mean age = 9.75 yrs, ASD	**ABA**: NCR, DRA, response blocking	School; Alternating treatments design 40-60 ss	Observation of mouthing, hair manipulation, motor stereotypy	Rates of CB at lowest levels across ppts for NCR + DRA + response blocking

[23] Lin, 2018; US	n = 3; Case series	67% males, 4-6 yrs, mean age = 5 yrs, ASD	**ABA**: Parent-led manualised pivotal response training	Home; pp1: 2 ss over 20 hrs; pp2&3: 2 ss over 24 hrs	Researcher observations of child behaviour flexibility - engagement in non-restricted repetitive task without CB (various); and of indicators of parent-child interaction quality; Parent report questionnaires on behaviour flexibility	Child behaviour flexibility, and parent-child interaction improved; Parent rated behaviour flexibility improved
[77] Clay, 2018; US	n = 1; Case study	Female, 12 yrs, ASD	**ABA**: Non-contingent access to single; multiple or alternating competing stimuli	Clinic; 32 ss, 20 mins per ss	Observation of self-injurious behaviour; and item engagement by researchers	Non-contingent access to a single competing item most effective at reducing self-injurious behaviour
[78] Slocum, 2018; US	n = 3; Case series	Males, 3-12 yrs, mean age = 7.33 yrs, ASD	**ABA**: Signalled, continuous access to a functional reinforcer for aggression and slow increase in signalled unavailability of the reinforcer	Clinic; 1.87 hrs - 2.67 hrs	Observations of aggressive behaviour by researchers	Reduction in aggression across participants
[79] Randall, 2018; US	n = 1; Case study	Female, 11 yrs, ASD	**ABA**: Differential R+, punishment, visual schedule	Clinic; 2 ss over 48 hrs	Observation of aggression by researchers	Reduction in aggression
[91] Gerow, 2019; US	n = 3; Multiple baseline	Males, 3-7 yrs, mean age = 5.5 yrs, ASD	**ABA**: FBA, parent-led FCT	Multiple - Home and Clinic; 36 ss over 4.8 hrs	Researcher observation of CB	CB reduced with introduction of FCT in all 3 participants. 2 clear intervention effects; one less clear
[94] Delemere, 2018; Northern Ireland	n= 6; Multiple baseline	67% Males, 2.5- 6.5 yrs, mean age = 4.61 yrs, ASD	**ABA**: Bedtime fading and positive routines (stimulus control)	Multiple - Clinic, home; 168 hrs over 7 days	Researcher observations of CB (plus various sleep measures)	Reduction in CB alongside improvements in sleep behaviour
[98] Dowdy, 2019; US	n = 2; Case series	Males, 10 yrs, 17 yrs, ASD, ADHD	**ABA**: Differential R+	School; 17 -22 ss over 2.8 - 3.67 hrs	Latency of transitions and frequency of transition refusal behaviour observed by researchers	Transition refusal behaviour decreased with intervention; It remained low at follow up

[80] Iadarola, 2018; US	n = 150; non-RCT	Intervention: 85% male, mean age 7.1 yrs, Waitlist control: 88% male, mean age 7.1 yrs, ASD	**PBS** teacher training (STAT: schedules, tools and activities for transitions) on using ABA principles to facilitate transitions	School; 6-12 ss for 5-10 hrs	Researcher observation of behaviour; Teacher report questionnaire on CB; Teacher written descriptions of target behaviour problems rated by researchers for severity and intensity	Greater reduction in CB severity for students in the treatment versus waitlist group (no improvement in waitlist)
[81] McGill, 2018; UK	n = 21; RCT	52% males, age range 19-84 yrs, mean age unclear, ID	**PBS** Service model review and staff training support	Supported living; duration unclear	Staff report questionnaire of CB; observations of meaningful engagement in activities, and staff support	CB measured reduced significantly more in intervention group versus the control group CB reduced in all intervention group settings, but only in 7 of 12 control settings
[82] Lee, 2019; UK	n = 1; Case study	Female, 40 yrs, ID	**PBS** workshops and training for staff; modelling and coaching staff to provide better support	Supported residential accommodation; duration unclear	Observations of moaning and grabbing by staff Incident records by staff Interviews with staff	Reduction in moaning and grabbing, staff reported increase in ppts communicative ability and in staff ability to provide support
[83] Clarke, 2018; US	n = 1; Case study	Female, 13 yrs, ASD	**PBS** Peer-mediated	School; 7.5 hrs over 10 days	Peer buddy rated for social validation; Direct researcher observation of CB and engagement	CB such as aggression, self-injurious behaviour and screaming reduced
[84] Bowring, 2019; UK	n = 85; pre-post	32% Male, mean age = 25.38 yrs, ID, ASD, ID + ASD	**PBS** specialist PBS team	Community setting (supported living); duration unclear	Parent and health care professional completed rating scales, questionnaires, surveys on CB	Significant reduction in CB and increase in quality of life
[85] MacDonald, 2018; UK	n = 50; non-RCT	16% Males, 40 yrs, mean age = 41 yrs, 40 - 42 yrs ASD	**PBS** Staff training	Supported living; 8 ss, duration unclear	Caregiver report questionnaires on CB; and on several aspects of engagement and wellbeing; Behaviour recording forms	Reduction in CB

[92] Grey, 2018; UAE	n = 7; Multiple baseline	71% males, 8-13 yrs, mean age = 14 yrs ID	**PBS** Interim behavioural recommendations for waiting list patients	Supported living; 24 months, total duration unclear	Record for outcome measures in behaviour, impairment, symptoms and social functioning for participant; Frequency of target behaviours	An overall reduction in anxiety, depression, mania, ADHD and CBs between all participants following the intervention phase.
[99] Hassiotis, 2018; UK	n = 245; Cluster RCT	64% males, 25-51 yrs (mean age unclear)	**PBS** staff training + access to mentor for 1 year	Community ID support; 3 x 2 day workshops over 12.5 hrs	Caregiver completed questionnaire on CB (ABC) and others on various aspects of wellbeing	No treatment effects in any of outcomes
[86] Ollendick, 2018; US	n = 134; RCT	62% males, 7-14 yrs (mean age unclear), ODD	**Parent training:** Parent Management Training versus Collaborative and Proactive Solutions	Clinic; 12 ss over 15 hrs	Clinical global impression - severity and improvement Mother reports of children's aggression and conduct problems	Significant improvements in aggression and conduct problems in both intervention groups, but no significant difference between groups
[28] Briegel, 2018; Germany	n = 1; Case study	Male, 10 yrs, ADHD, ODD	**Parent training:** Parent-Child Interaction Therapy (PCIT)	Clinic; 13 ss, duration unclear	Clinician assessed diagnostic instrument Behavioural rating scales and questionnaires done by parents	After PCIT, child no longer met diagnostic criteria for ODD and conduct difficulties were within normal range 17 months post baseline
[87] Pennefather, 2018; US	n = 18; Pre-post	12 males, 4-8 yrs, mean age = 6 yrs, ASD	**Parent training:** Online, including principles of ABA and cognitive therapy (ACT & optimism training)	Home; 3 ss over 4 hrs 30 mins	Parent report questionnaire on behaviour problems; parent self-report questionnaires on stress	Improved hyperactive and prosocial behaviour in children; reduced stress in parents
[22] Fodstad, 2018; US	n = 11; Pre-post	70% male, 1-5 yrs, mean age unclear, ID	**Parent training:** SIB training	Clinic; 11 ss over 11 - 16.5 hrs	Observations of self-injurious behaviour during parent-child interactions; Clinician ratings of global problem; Rating scales and questionnaires completed by parents.	Decreases in self-injurious behaviour and decreases in negative parent-child interactions

[26] Zlomke, 2019; US	n = 28; Pre-post	75% male, age range 2-8 yrs, mean age = 4.29 yrs, ASD	**Parent training:** PCIT	Clinic; 16 ss over 16-24 hrs	Parent report questionnaires on child CB and parent stress	Significant reductions in parent rated disruptive behaviour; significant reductions in parent stress
[27] Hosogane, 2018; Japan	n = 2; Case series	50% Males, 3 yrs and 4 yrs 3 months, ADHD	**Parent training:** PCIT	Clinic; 23-30 ss over 30-45 hours	Parent completed questionnaire on CB	Reduction in CB
[24] Ciesielski, 2019; US	n = 159; Pre-post	47% Males, mean age = 8.09 yrs, 6 - 12 yrs, ADHD	**Parent training:** Behavioural parent training	Home; 8 ss, duration unclear	Parent scale for severity of problems with compliance for child; Parent-rated measure for frequency and degree of child behaviour-related stress experienced by parent/caregivers	1) Reductions in severity of child non-compliance; 2) Reductions in number of non-compliant contexts; effect size 1 > 2
[25] Cambric, 2019; US	n = 1 ; Case study	Male, 7 yrs, ASD	**Parent training**: Parent-Child Interaction Therapy (PCIT)	Clinic; 15 ss over 15 hrs	Parent report questionnaire on CB	Decrease in CB, increase in compliance
[35] Singh, 2018; US	n = 3; Multiple baseline	Males, age range 16-17 yrs, mean age 16.3 yrs, ASD, borderline intellectual function	**Meditation:** Mindfulness training (Surfing the Urge)	Home; 10 ss over 5 hrs	Parents incidents records of verbal and physical aggression	Significant decrease in CB
[33] Cohen, 2018; US	n = 23; RCT	65% male, 3-5 yrs, mean age = 4.08 yrs, ADHD, 12 received yoga first, 11 waitlist first	**Meditation:** Yoga	Multiple - School, home; 12 ss over 6 hrs	Parent and teacher ratings of problem behaviour	Yoga linked to improvements in parent rated hyperactivity; and teacher rated conduct problems and prosocial behaviour
[31] Singh, 2019; US	n = 93; Pre-post	65% males, 13-17 yrs, mean age = 15.15 (ASD), 15.56 (ID)	**Meditation:** Mindfulness + PBS training for parents	Home; 40 weeks, number of ss unclear	Mother report of child CB; mother self-report of stress	Decreases in child aggression and non-compliance; decreases in mother stress

[37] Phillips, 2019; US	n =3; Case series	3 males, 8-18 yrs, mean age = 11.33 Angelman syndrome, ASD, PDD-NOS	**Meditation:** Diaphragmatic breathing, DRO, FCT	Clinic; 3 hrs weekly with ss lasting 10 mins each, total duration unclear	Aggression measured by therapist	Aggression only reduced in one participant until extinction was included, then aggression reduced in all participants
[34] Huguet, 2019; Spain	n = 70; RCT	72.8% male, age range 7-12, mean age = 9 yrs (intervention group), 8.81yrs (control group), ASD	**Meditation:** Emotional self-regulation mindfulness	Clinic; 75 mins ss, amount of ss unclear	Clinician diagnostic interview and parent rating scales	Significant decrease in aggressive behaviour in mindfulness group but clear evidence of statistical difference from control group not reported
[36] Griffith, 2019; UK	n = 7; Pre-post	43% Male, mean age = 33.14 yrs, ID	**Meditation:** Mindfulness (Soles of the Feet)	Home; 6 ss over 6-10 hrs	Caregiver interview	Reports of reductions in aggressive behaviour for some ppts; Reports of approach being more effective for individuals with better understanding of programme
[32] Jones, 2018; UK	n = 21; Pre-post	62% males, 4-16 yrs, mean age = 10.53 yrs, ASD	**Meditation:** Mindfulness training for parents	Clinic; 8 weeks over 16 hrs	Self-report on general mindfulness and self-compassion; parent report questionnaire on child CB	Reduction in parent stress; increase in mindfulness and self-compassion; No change in child CB
[46] Gallego-Matellán, 2019; Spain	n = 1; Case study	Male, 11 yrs, ADHD	**Skill training:** Behavioural based psychoeducation with parents Emotion recognition, perspective taking and empathy training with child via discussion of emotional films/ photographs	Multiple - Clinic, home; 36 ss, duration unclear	Parent report questionnaire on prosocial behaviour; parent descriptions of behaviour	Improved prosocial behaviour; improved disruptive behaviour

[40] Hu, 2018; China	n = 1; Case study	Male, 4 yrs, ASD	**Skill training:** communication training using Picture Exchange Communication System (PECS)	Multiple - Therapy room, playground, home; 15 min ss; total duration unclear	Observation of behaviour by researchers	Increase in vocal mands and decrease in aggressive behaviour
[44] Aspiranti, 2018; US	n = 21; Multiple baseline	67% Males, ages 5-11, mean age of classroom A = 6.5, mean age of classroom B = 9.5, mean age of classroom C = 6.5, ASD	**Skill training:** Colour wheel training - red, yellow, green rules for different situations, so that children know what to expect at any given time (i.e. strong focus on facilitating transitions)	School; 22-23 ss over 7.3 -7.6 hrs	Observation of inappropriate behaviours by 2 observers per class	Decrease in disruptive behaviours across all 3 classrooms
[39] Jahoda, 2018; UK	n = 161; RCT	47% Male, mean age = 40 yrs, ID + depression	**Skill training:** Behavioural activation, guided self-help for depression	Clinic; BeatIt: Mean 10 ss over 10-20 hrs, Stepup: 8 ss over 8-12 hrs	Caregiver ratings of aggressiveness; Self- and caregiver report questionnaires on depression and other mental wellbeing measures	Decreases in symptoms of depression and level of aggressive behaviour. No statistically significant differences between groups for depressive symptom scores.
[47] Sandjojo, 2018; Netherlands	n = 26; Non-RCT	62% males, mean age = 33.45 yrs, ID	**Skill training:** Staff training in promoting self-management (On your own two feet)	Home - supported residential accommodation; 2 ss over 12 hrs	Staff completed questionnaires on service users' emotional and behaviour problems; independence and self-reliance; support needs	No significant group difference in CBs or support needs; Significant improvements in independence and self-reliance and
[45] Vanzin, 2018; Italy	n = 62; Non-RCT	68% males, 8-13 yrs, mean age not given, ADHD	**Skill training:** Parent training in behavioural strategies Child training in emotion recognition and coping	Clinic; 34 ss over 51 hrs	Parent (and other) questionnaires on behavioural and emotional problems of child; Clinician ratings on patents' symptoms of mental illness	Significant improvement in the children's global functioning, emotional and behavioural problems at the end of treatment. Treatment group were more likely to shift from a more severe functional impairment class to a less severe one.

[49] Wills, 2018; US	n = 1; Case study	Female, 30 yrs, ASD	**Technology:** self-monitoring application - provides regular prompts e.g. "are you on task?"; "are you being appropriate?"	Work; 32 ss for 5 hrs 20 mins	Observation of inappropriate vocal behaviours	Decrease in inappropriate vocalisation behaviours which were maintained
[48] Muharib, 2018; US	n = 2; Case series	50% male 5-6 yrs, ASD	**Technology:** FCT with an iPad app	School; 15 ss over 1 hr 45 mins	Observation of CB by researchers	Reduction in CBs, minimal prompting necessary for use of app
[50] Fachantidis, 2019; Greece	n = 1; Case study	Male, 9.75 yrs, ASD	**Technology:** Robot construction activity used in a classroom in group activities between child with ASD and classmates	School; 18 ss over 36 hrs; 9 ss over 9 hrs	Observational data forms for social and communication skills and undesirable behaviour completed by researchers	Reduction in CB (stereotypical movements, task avoidance, indifference to surroundings, excessive reactions to shouting, the desire to leave the classroom and the tendency to talk about unrelated issues)
[51] Harper, 2018; US	n = 1; Case study	Male, 21 yrs, ID	**Technology:** ambulatory support (Gait trainer)	School; 8 ss over 40 mins	Observation of aggression by researcher	Aggression immediately decreased when intervention was initiated
[52] Hoffman, 2019, South Africa	n = 6; Multiple-baseline	83% males, 27-56 yrs, mean age = 45.33 yrs, ID	**Technology:** Messaging app to teach person with NDD person permanence (target: separation anxiety)	University; Up to 21 weeks, duration unclear	Caregiver daily ratings of observed behaviour (distress, CB, clinging, anxiety); Standardised informant report questionnaires on CB and anxiety	Significant decrease in the frequency of CBs; and in anxiety
[88] Beh-Pajooh, 2018; Iran	n = 60; Non-RCT	Males, mean age = 12 yrs, ID	**Arts:** Painting therapy	School; 2 ss over 18 hrs	Parent report questionnaires on externalising behaviour problems; Test of drawing skill and IQ	Reduction in externalising behaviour after the painting therapy programme. Significantly greater reduction in intervention versus control group
[89] Tudor 2018; US	n = 1; Case study	Female, 9 yrs, Tourette's	**CBT:** 12 week family CBT	Clinic; 12 weekly ss over 12 hrs	Independent observer reported questionnaire on aggression	Decrease in aggression following intervention

[90] Brookman-Frazee, 2019; US	n = 202; RCT	84.2% male, mean age 9.1 yrs (age range unclear), ASD	**Multiple:** Individualised mental health intervention (AIM HI)	Clinic; AIM-HI mean 15.05 ss; usual care mean 13.59 ss	Behavioural rating scales conducted by research team	Intensity and severity of CBs in children with ASD decreased with publicly funded MH service therapists who were trained to do the intervention
[93] Muldoon, 2018; US	n = 3; Pre-post	Males, 3-5 yrs, mean age = 4 yrs, ASD	**Multiple:** Family centred mealtime intervention (Easing Anxiety Together with Understanding and Perseverance)	Clinic; 30-36 ss over 25.8-30 hrs	Reports on participant food behaviour and mealtime skills by researcher	Reduction in CB during mealtimes
[95] Delion, 2018; Italy	n = 41; RCT	78% males, mean age = 8.29 yrs, ASD	**Sensory reintegration**: Therapeutic body wraps	Clinic; 2 ss over 18 hrs - 24 hrs	Caregiver report of CB (focus on aberrant behaviour irritability)	Irritability scores decreased following intervention
[96] Neijmeijer, 2019; Netherlands	n = 604; Longitudinal	83% males, mean age = 33.5 yrs, ID	**Service delivery model:** Flexible assertive community treatment (ACT), Intensive assertive outreach	Clinic; 6 yrs, total duration unclear	Staff report questionnaires on several aspects of functioning, including CB	Level of social disturbance and the risk factors for challenging and criminal behaviour diminished. Significant reductions in CB over time.
[97] Loring, 2018; US	n = 19 Pre-post	63% male, 11-17 yrs, mean age 14.7 yrs, ASD	**Sleep:** Sleep education	Home; 2 ss over 2 hrs 50 mins	Multiple informant report questionnaires	Significant improvement in externalising behaviour

Table 2: Risk of biases present in included studies. Studies presented in alphabetical order based on the surname of the first author. Risk of bias domains are operationalised in the supplementary materials. In general, "Yes" indicates that the authors have appropriately guarded against the specified risk; "No" indicates they have not; "N/A" indicates that the risk is not applicable to the study; and "U/C" indicates that it is unclear whether the risk has been appropriately guarded against or not.

Author, year	RoB 1. Random sequence	RoB 2. Concealment	RoB 3. Representation	RoB 4. Blinding (ppts/raters/deliverer)	RoB 5. Aquiescence	RoB 6. Blinding (objective/outcome	RoB 7. Complete outcome data	RoB 8. No selective reporting	RoB 9. Baseline outcome	RoB 10. Validity/Reliablity of outcome measures	RoB 11. Study criteria transparency	RoB 12. Seasonality	RoB 13. Competing Interest
Aspiranti, 2018	N/A	N/A	No	No	Yes	U/C	Yes	Yes	Yes	No	No	Yes	Yes
Beh-Pajooh, 2018	No	No	Yes	No	N/A	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Benson, 2019	N/A	N/A	No	No	N/A	U/C	Yes	Yes	Yes	No	No	Yes	Yes
Bloom, 2018	N/A	N/A	No	No	No	No	Yes	Yes	Yes	Yes	U/C	Yes	No
Bowring, 2019	N/A	N/A	U/C	No	No	No	U/C	Yes	Yes	Yes	U/C	Yes	No
Briegel, 2018	Yes	Yes	Yes	Yes	N/A	No	Yes	Yes	Yes	Yes	N/A	Yes	Yes
Briggs, 2017	N/A	N/A	No	No	U/C	No	Yes	Yes	Yes	Yes	No	Yes	Yes
Brookman-Frazee, 2019	Yes	No	Yes	PT	No	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes
Cambric, 2019	N/A	N/A	N/A	No	No	No	N/A	Yes	N/A	No	No	Yes	Yes
Cariveau, 2019	No	No	No	None	N/A	No	Yes	Yes	U/C	Yes	Yes	Yes	Yes
Ciesielski, 2019	N/A	N/A	No	No	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Clarke, 2018	N/A	N/A	N/A	No	No	No	Yes	Yes	Yes	Yes	No	Yes	PT
Clay, 2018	N/A	N/A	No	No	Yes	No	N/A	Yes	Yes	Yes	U/C	Yes	Yes
Cohen, 2018	U/C	No	No	No	No	No	U/C	Yes	Yes	No	No	Yes	No
Courtemanche, 2018	N/A	N/A	No	Yes	No	No	Yes	Yes	Yes	No	Yes	Yes	Yes
Delemere, 2017	U/C	No	Yes	No	Yes	No	Yes	Yes	Yes	Yes	N/A	Yes	U/C
Delion, 2018	Yes	Yes	No	U/C	Yes	U/C	Yes	Yes	Yes	Yes	No	No	Yes
Dowdy, 2019	N/A	N/A	No	No	U/C	U/C	Yes	Yes	Yes	No	No	Yes	No
Fachantidis, 2019	N/A	N/A	U/C	No	U/C	U/C	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Fisher, 2018	N/A	N/A	No	No	Yes	U/C	Yes	Yes	Yes	No	No	Yes	No

Fodstad, 2018	N/A	No	No	No	No	No	Yes	Yes	Yes	Yes	No	Yes	Yes
Gallego-Matellán, 2019, Spain	N/A	N/A	No	No	No	No	Yes	Yes	N/A	No	No	Yes	PT
Gerow, 2019	N/A	N/A	No	No	U/C	U/C	No	No	N/A	No	No	Yes	Yes
Grey, 2018	N/A	N/A	No	No	Yes	No	N/A	Yes	Yes	No	U/C	No	Yes
Griffith, 2019	N/A	N/A	U/C	No	U/C	No	No	Yes	N/A	No	No	Yes	Yes
Haq, 2018	N/A	N/A	No	No	No	No	Yes	Yes	Yes	No	No	Yes	Yes
Harper, 2018	N/A	N/A	No	No	No	No	Yes	Yes	Yes	Yes	No	Yes	No
Hassiotis, 2018	Yes	Yes	No	No	Yes	U/C	Yes	Yes	Yes	No	No	Yes	Yes
Herman, 2018,	No	No	No	None	N/A	No	Yes	Yes	Yes	Yes	Yes	No	Yes
Hoffman, 2019,	N/A	N/A	No	No	Yes	No	N/A	Yes	Yes	No	U/C	Yes	PT
Hosogane, 2018,	N/A	N/A	N/A	No	No	No	Yes	Yes	No	No	Yes	Yes	Yes
Hu, 2018	N/A	N/A	N/A	No	No	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes
Huguet, 2019	Yes	No	Yes	No	Yes	No	No	Yes	Yes	No	U/C	Yes	Yes
Iadarola, 2018	N/A	N/A	No	No	No	No	Yes	Yes	N/A	Yes	No	Yes	Yes
Jahoda, 2018	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes
Jones, 2018	N/A	N/A	No	No	Yes	Yes	No	Yes	Yes	Yes	Yes	No	Yes
Kelley, 2018	N/A	N/A	No	No	U/C	U/C	Yes	Yes	Yes	Yes	N/A	Yes	Yes
Lambert, 2018	N/A	N/A	No	No	N/A	No	Yes	Yes	Yes	Yes	No	Yes	Yes
Lee, 2019	N/A	N/A	N/A	No	No	No	Yes	Yes	N/A	No	No	U/C	No
Lin, 2018	N/A	N/A	U/C	No	No	PT	Yes	Yes	Yes	Yes	No	Yes	Yes
Loring, 2018	N/A	N/A	U/C	No	U/C	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Lugo, 2018	N/A	N/A	N/A	No	No	No	Yes	Yes	Yes	Yes	No	Yes	Yes
MacDonald, 2018	No	No	Yes	No	U/C	U/C	Yes	Yes	Yes	U/C	N/A	Yes	No
McGill, 2018	Yes	No	No	No	No	No	Yes	Yes	Yes	Yes	N/A	Yes	Yes
Mitteer, 2019	N/A	N/A	No	No		No	Yes	Yes	Yes	Yes	N/A	Yes	No
Monlux, 2019	Yes	No	No	No	N/A	No	Yes	No	Yes	Yes	N/A	Yes	PT
Muething, 2018	N/A	N/A	No	No	Yes	No	Yes	Yes	Yes	Yes	No	Yes	No
Muharib, 2018	N/A	N/A	No	No	No	No	N/A	Yes	Yes	Yes	No	Yes	Yes
Muldoon, 2018	N/A	N/A	N.A	No	No	No	No	Yes	Yes	Yes	No	No	Yes
Neijmeijer, 2019	N/A	N/A	No	No	No	N/A	Yes	Yes	Yes	Yes	No	No	Yes
Newcomb, 2018	N/A	N/A	No	No	Yes	No	Yes	Yes	Yes	Yes	N/A	No	U/C
Ollendick, 2018	Yes	Yes	No	No	Yes	No	No	Yes	Yes	Yes	N/A	Yes	PT

Pennefather, 2018	No	No	No	No	No	No	Yes	Yes	Yes	No	No	Yes	Yes
Phillips, 2019	N/A	N/A	N/A	No	No	No	Yes	Yes	U/C	Yes	U/C	Yes	Yes
Planer, 2018	No	No	No	No	Yes	No	Yes	Yes	Yes	Yes	N/A	Yes	No
Randall, 2018	N/A	N/A	No	No	Yes	No	N/A	Yes	Yes	Yes	U/C	No	PT
Saini, 2018	N/A	N/A	No	U/C	No	No	Yes	Yes	Yes	Yes	No	Yes	No
Sandberg, 2018	N/A	N/A	No	No	Yes	U/C	No	Yes	Yes	Yes	Yes	Yes	PT
Sandjojo, 2018	N/A	No	No	No	U/C	No	Yes	Yes	Yes	Yes	N/A	U/C	Yes
Schreck, 2018	N/A	N/A	N/A	No	No	No	U/C	Yes	N/A	No	No	No	Yes
Singh, 2018	N/A	N/A	No	No	U/C	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Singh, 2019	No	No	No	No	U/C	No	Yes	Yes	Yes	Yes	No	Yes	Yes
Slocum, 2018	N/A	N/A	Yes	No	U/C	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Stevenson, 2019	N/A	N/A	No	No	No	U/C	Yes	Yes	Yes	No	N/A	Yes	Yes
Tudor 2018	Yes	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	PT
Vanzin, 2018	U/C	No	Yes	None	None	None	Yes	Yes	Yes	No	Yes	No	Yes
Verriden, 2019	N/A	N/A	Yes	No	U/C	Yes	Yes	Yes	Yes	Yes	N/A	Yes	No
Wills, 2018	N/A	N/A	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	PT
Zlomke, 2019	No	No	No	No	No	No	Yes	Yes	Yes	Yes	No	Yes	No

Supplementary materials

Search terms

Three groups of search terms were composed, pertaining to each of the three primary inclusion criteria – that it is an intervention study, addressing challenging behaviour, in a neurodevelopment disorder population. The groups of search terms were combined with OR operators between items within the same group, and with AND operators across groups. The syntax was adapted appropriately in line with the requirements of each database. Search terms were developed with reference to previously published reviews and recent papers in the area. We wanted to include genetic neurodevelopmental disorders, which may be referred to only by their genetic or syndromal name. Thus, we used the list of syndrome names published by the international research association, the Society for the Study of Behavioural Phenotypes (https://ssbp.org.uk/syndrome-sheets/). We recognise that this is by no means an exhaustive list of genetic neurodevelopmental disorders, but it is a reasonable compromise given the scope of this review. The groups of search terms were as follows:

(intervention* OR “behavioural management” OR “behavioral management” OR “behaviour management” OR “behavior management” OR strategies OR strategy OR treatment* OR support OR train* OR teach* OR tool* OR “behavi* suppression” OR “behavi* reduction” OR “applied behavi* analysis”)

AND

(Neurodiversity OR “neurodevelopmental disorder*” OR Neurodiverse OR neurodivergent OR "autism spectrum disorder*" OR "prader willi syndrome" OR “prader-willi syndrome” OR "williams syndrome" OR "fragile x syndrome" OR "attention deficit hyperactivity disorder" OR “attention disorder*” OR "down syndrome" OR "intellectual disabil*" OR “learning disabil*” OR “Angelman Syndrome” OR “CHARGE Syndrome” OR “Coffin-Lowry Syndrome” OR “Coffin Siris Syndrome” OR “Cornelia de Lange Syndrome” OR “Cri du Chat Syndrome” OR “Foetal Alcohol Syndrome” OR “Lesch-Nyhan Syndrome” OR “Mowat-Wilson Syndrome” OR “Neurofibromatosis Type 1” OR “Noonan Syndrome” OR “Rett Syndrome” OR “Rubinstein-Taybi Syndrome” OR “Triple-X Syndrome” OR “klinefelter syndrome” OR “XXY syndrome” OR “Tuberous Sclerosis Complex” OR “Turner Syndrome” OR “Wolf-Hirschhorn Syndrome” OR “XYY Syndrome” OR “22q11.2 Deletion Syndrome”)

AND

(aggress* OR “self injur*” OR “self-injur*” OR SIB OR destruct* OR tantrum* OR "temper outburst*” OR meltdown* OR blip* OR rage* OR “challenging behaviour*” OR “challenging behavior*” OR pica OR stereotypy OR noncompliance OR “inappropriate vocalization*” OR “inappropriate vocalisation*” OR screaming OR “off task behaviour” OR “off task behavior" OR yelling OR “inappropriate touching” OR pushing OR “not following directions” OR shouting OR arson OR regurgitation OR pinching OR scratching OR throwing OR biting

Inclusion criteria

We included studies with any design which were published or in press in a peer reviewed journal, met the inclusion criteria described in Supplementary table 1, and were not excluded based on the criteria described in Supplementary table 2.

Supplementary table 1: Inclusion criteria for abstract, title and full-text screening

Inclusion criteria
Describes any intervention which may prevent, manage or reduce any display of challenging behaviour through any method except pharmaceutical or medical (device or surgical procedure), including but not limited to: • Previously developed or tested interventions which have been now been tested with a different NDD or used to target a different mechanism (related to challenging behaviour) or a different challenging behaviour • Any combination of previously evidenced strategies in a novel intervention targeting challenging behaviour or underlying mechanisms • Caregiver training (psychoeducation or caregiver-led intervention) which facilitates the reduction of challenging behaviour in the NDD A non-biological mechanism targeted with demonstrated rationale of a link to a challenging behaviour
The subject of the intervention includes an NDD population or instructs carers of an NDD population • Including any NDD • Including any age • Including any co-morbidity
Challenging behaviour is measured such that the potential impact of the intervention on behaviour can be determined. Challenging behaviour is defined as behaviour which can be harmful to the individual or those around them, including but not limited to: • Externalised: refusals, self-injurious behaviours, temper outbursts, verbal or physical aggression, property destruction • Internalised: high levels of anxiety, insistence on sameness

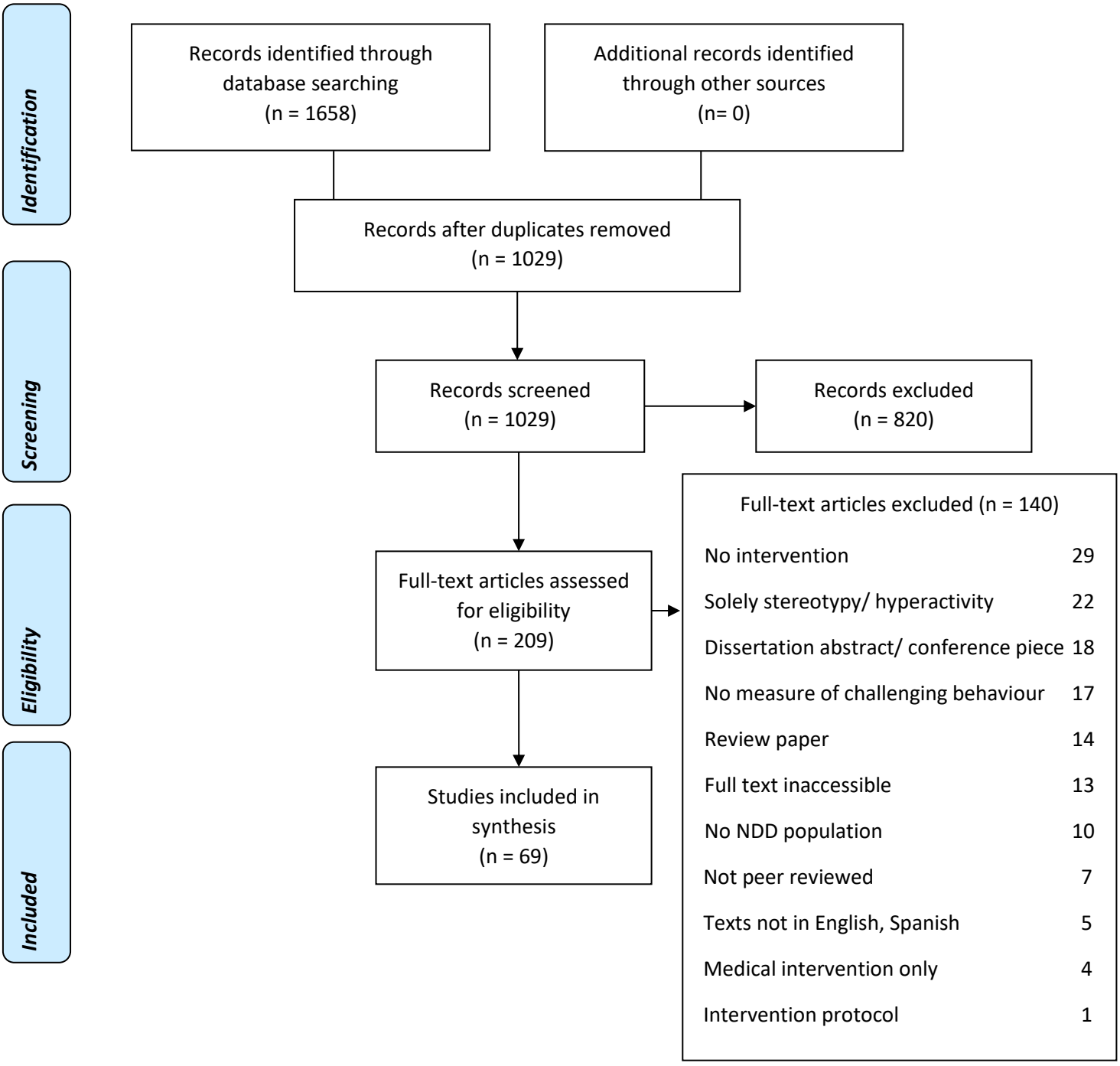
Supplementary table 2: Exclusion criteria for abstract, title and full-text screening

Exclusion criteria
Texts not in English or Spanish
Books, chapters, dissertations, conference abstracts or reports
Biological (including diet) or medical intervention only
Animal studies
Describes the development of assessments for challenging behaviours
Assesses challenging behaviours not further defined such, where it is unclear whether the behaviour meets our definition for challenging behaviour
Measures only behaviours that that are not always classified as challenging. Including and limited to: off-task behaviour, stereotypy, inappropriate vocalisations, hyperactivity, impulsivity, irritability
No measure of challenging behaviour, can include questionnaire, interview, direct observation or – if a qualitative study – reduction of challenging behaviour is not a theme

Screening

Covidence [100] was used to facilitate the screening of articles for inclusion by two independent researchers. Screening was conducted at title/abstract, and full text levels sequentially based on the PRISMA statement [101]. The screening process is described in Figure 1.

PRISMA 2009 Flow Diagram



Risk of bias rating

Risk of bias was assessed using bespoke criteria developed by our research team for a previous systematic review of mostly early and mid-stage intervention development research. The criteria were based on guidelines published by the Cochrane group, National Institute for Health Research Online Guidance for Feasibility and Pilot Intervention Studies and the Newcastle Ottowa Quality Assessment Scale for Assessing the Quality of Non-Randomised Studies [19-20]. Table 3 describes these criteria.

Table 3 Risk of bias assessment criteria

RoB domain	**RoB source**	**Low risk of bias definition for review**	**Yes, no, unclear or N/A with example of associated extraction table statement** *(see extraction table)*
Selection bias	1. Random sequence generation	Randomisation was employed to allocate participants to intervention ***and*** the random sequence generation method was clearly explained (where, using what method, with what software).	Yes: Randomisation was employed to allocate participants to intervention and the randomisation lists were obtained using x procedure (explain this clearly), at x location. No: Randomisation was not employed to allocate participants to intervention *or* randomisation was used BUT evidence for generation of a randomised sequence was not provided. Unclear: The randomisation lists were created at x but further details were not provided. N/A: The study design employed was not relevant to random sequence generation selection bias, e.g. single case study or feasibility study.
	2. Allocation concealment	Randomisation was employed to allocate participants to intervention ***and*** the method used to conceal the allocation sequence from the researchers was explained clearly.	Yes: Randomisation was employed to allocate participants to intervention and x method was used to conceal the allocation sequence (explain this clearly); this was implemented by x. No: Randomisation was not employed to allocate participants to intervention *or* the allocation to intervention was not concealed before intervention assignment. Unclear: The allocation sequence was concealed but further details were not provided. N/A: The study design employed was not relevant to allocation concealment selection bias, e.g. single group repeated measures study.

	3.	Population representation	It was clear from the recruitment method that participants recruited for the study were representative of the population from which they were drawn.	Yes: Participants recruited for the study were representative of the population from which they were drawn, (e.g. five randomly selected children's homes from a whole population of children's homes in Scotland were included in the study or stratified sampling or systematic sampling). No: Participants recruited for the study were not representative of the population from which they were drawn (e.g. opportunistic/convenience sampling at a youth wellbeing drop-in group in x city suburb used to recruit a looked-after children population or self-selecting sample). Unclear: Recruitment method is unclear, *or* participants are fairly typical of the average in the population from which they were drawn (e.g. looked-after children population). N/A: The study design employed was not relevant to population representation selection bias, e.g. RCT.
Performance bias	4.	Blinding of participants, raters and intervention deliverer*	Measures are used to blind participants, raters and intervention deliverer(s) from knowledge of which intervention participants received and these were explained; ***or*** (measures were used to blind participants from knowing that the authors wished to create a satisfactory intervention/assess part of an intervention **applicable to feasibility/acceptability type studies only*) ***and*** information relating to whether the intended blinding was effective was provided.	Yes: Participants, raters and intervention deliverer(s) taking part in the feasibility study were advised they would be taking part in research on x but full aim of the study (i.e. to find out if a part of an intervention was satisfactory) was not divulged (clearly explain the relevance of the type of study in relation to the definition). The efficacy data showed x. No: Participants and/or raters and/or intervention deliverer(s) were not blinded from knowledge of which intervention participants received in the RCT (clearly explain the relevance of the type of study in relation to the definition). Unclear: The blinding measures were unclear.
	5.	Acquiescence	In studies examining new interventions or components of interventions, methods taken to ensure that outcome assessments objectively seek opinions rather than suggesting that that one answer is desirable are described clearly ***and***	Yes: X procedure was used in the case study to ensure that participants did not feel pressured into giving certain responses (explain this clearly). The efficacy data showed x. No: A procedure was not put in place to ensure that participants did not feel pressured into giving certain responses in the acceptability single group study.

			Information pertaining to whether these measures were effective is also provided.	Unclear: It is not clear how effective the measures used to ensure that participants did not feel pressured into giving certain responses were as efficacy data was not provided. N/A: The study design employed was not relevant to acquiescence performance bias, e.g. RCT.
Detection bias	6.	Blinding/objectivity of outcome measures*	The person(s) interpreting the data was not aware of the hypotheses and aims; information was not accessible to them to allow them to be able to foresee the outcome (e.g. group affiliation data) ***and*** information concerning whether this was effective was provided *or* the outcomes were objective e.g. time taken to maintain an oscillatory frequency above a specified threshold.	Yes: The methods used to blind the person(s) interpreting the data from knowledge of the study hypotheses, aims and information pertaining to likely outcome of participants result were x (clearly explain this). The efficacy data showed x. No: The person(s) interpreting the data were not blinded from knowledge of the hypotheses and aims and which intervention participants received. Unclear: The blinding (and/or) objectivity of all outcome measures were unclear.
Attrition bias	7.	Incomplete outcome data*	Data was provided for all outcome variables. For each outcome measure, attrition (<15% total across all available data) and exclusions from analysis data was provided with reasons (including the numbers in each intervention group (compared with total participants), ***and*** any re-inclusions in analyses for the review; ***or*** the study design employed resulted in complete outcome data e.g. single case study.	Yes: Data was provided for all outcome variables *and* <15% attrition (give specific %). This was due to x. n = x lost in x group, n = x lost in x group; total participants = x. No: Data was not provided for all outcome variables *and/or* >15% attrition (give specific %). No information regarding exclusions provided and no information provided related to reasons, or breakdown for each intervention group. Unclear: The attrition data was not provided or was unclear. N/A: The study design employed was not relevant to attrition bias, e.g. a study examining a component part of an intervention.
Reporting bias	8.	Selective reporting*	Selective outcome reporting was documented ***and*** the findings were presented.	Yes: There are no discrepancies between measures used and outcome data; *or* any discrepancies between the measures and outcome data are clearly justified (document justification). No: There are discrepancies between measures used and outcome data *and* justification information in relation to selective outcome reporting was not provided.

9.	Baseline outcome measurements similar*	Performance or clinical outcomes were measured before the intervention in non-randomised trials, ***and*** there were no significant differences across groups, ***or*** there were differences across groups in randomised trials but this was taken into account in the analysis (e.g. ANCOVA).	Yes: Performance in x and x were measured at baseline in the non-randomised trial and there were no significant differences between groups; *or* performance in x and x were measured at baseline in the randomised trial and significant differences observed between groups was taken into account in the statistical analysis (report statistical method used). No: Important differences were found in baseline performance scores in the non-randomised trial; *or* there were differences between groups in the randomised trial and this was not taken into account in the analysis. Unclear: Baseline performance was measured, however data was not provided. N/A: The study design employed was not relevant to baseline outcome measurements similar reporting bias, e.g. single group repeated measures design.
10.	Validation and reliability of outcome measures*	All outcome measures were validated and/or reliable, as evidenced in the text or through further investigation into the outcome measure(s).	Yes: All outcomes measures were validated and/or reliable (report validity and reliability data for each outcome measure); for example: acceptable factor analysis loading values for validity and/or Cronbach's α values for reliability. No: Some, but not all outcome measures were validated and/or reliable (report available validity and reliability data for each outcome measure); for example: acceptable factor analysis loading values for validity and/or Cronbach's α values for reliability.
11.	Full-scale study criteria transparency	The criteria used in feasibility, pilot or single case studies to determine whether to conduct a full-scale study were provided (as well as results of all outcome measures) ***and*** the outcome and implications of this were clearly documented.	Yes: The criteria that was employed to determine whether to take the current study to a full-scale study were: x, x and x. The outcome of this was: x, the implication of this was: x. No: Criteria used to determine whether to take the current study to a full-scale study was not provided. Unclear: The criteria that was used to determine whether to take the current study to a full-scale study were: x, x and x, however the outcome of this was not provided *or* were unclear.

			N/A: The study design employed was not relevant to future research criteria transparency, e.g. RCT.
Other bias(s)	12. e.g. **Seasonality**, time of measurement, maturation, mortality, intervention setting differences, extreme high or low score at baseline (regression to mean effects), measurement differences (different outcome measure for different type of intervention).	There was no evidence of other sources of bias (i.e. caused by an extraneous variable) not accounted for by clearly described, specific methods, not previously covered in the other 5 domains.	Yes: There was no evidence of other sources of bias. No: A spurious effect may have been caused, e.g. by seasonal differences; the baseline measures were completed in January and the post intervention measures were completed in August. Unclear: There were potential spurious effects of x and x, however these were unclear.
	13. Competing interest and source of support	The author clearly stated that there were no competing interests **and** documented any sources of support (i.e. funding).	Yes: There were no competing interests and the source(s) of support are documented. Partial: Only the competing interest information *or* only the source of support was documented by the author. No: The competing interest and source of support was not documented by the author.