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The *Applied Behaviour Analysis* imperative in the management of autism

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Summary

ABA (applied behavioural analysis) interventions have the most robust body of evidence showing their efficacy in improving the outcome of autism. Comprehensive ABA interventions include the Lovaas model and a group of more recent naturalistic developmental behavioural interventions. Randomised clinical trials have shown developmental gains associated with those interventions in key domains (cognitive, language, play, social behaviour), although initial results have not been consistently replicated. There is no direct head-to-head comparison of comprehensive ABA approaches, and evidence for the need to deliver treatments with very high intensity or to intervene at a very early age is largely inconsistent. Implementation of ABA comprehensive interventions in community settings is associated with lesser effects. Pragmatic, low-cost interventions for low-resource countries and settings are currently being tested.

Keywords: *autism*, *applied behaviour analysis*, *treatment*, *efficacy*, *effectiveness*, *community implementation*, *nonverbal*, *intellectual disability*, *review*

When there is no cure, there are thousands of treatments.

The history of failed treatments of autism is impressive and equally long for pharmacological (LSD experiments, fenfluramine, naltrexone, secretin, haloperidol…), psychoanalytical (Bettelheim school, "refrigerator mother"), psychological (electric shocks, play and family therapy) and other treatment "philosophies" (megavitamins, gluten-free casein-free diet, facilitated communication, hyperbaric oxygen chamber…). Effective interventions developed in the 1970s with the adaptation of specialised education methods to autism as exemplified in Schopler's TEACCH model of services in North Carolina. In the 1980s, the publication by Lovaas of an apparently successful intervention relying purely on behavioural techniques [1] raised hopes for families and led to the development of new interventions collectively referred to as "ABA", short for applied behavioural analysis.

Like cognitive behavioural therapy (CBT) or behavioural modification, ABA for autism is a set of procedures rooted in validated experimental and learning theories (inspired by contributors such as Pavlov, Skinner, Eysenck...) that have in common: use of well-described and fully specified techniques, reliance on intensive data collection and analysis, articulation with behavioural theory as referential, a focus on practical and measurable change and a goal of effecting long-lasting changes across settings. These techniques are used in various combinations in psychiatric interventions, but also in domains unrelated to human health such as business, management, or behavioural phenotyping of rodents.

The ABA paradigm relies on the three-term contingency A-B-C (antecedent-behaviour-consequence), employs experimental functional analysis to identify factors that precipitate and maintain dysfunctional behaviours, tests hypotheses about the A-B-C linkages and uses a collection of methods (positive or differential reinforcement, fading, extinction, shaping…) to achieve change.

ABA approaches used in autism are heterogeneous and one must therefore specify what one means by "ABA" when using that terminology (table 1). *Focused* ABA interventions to remedy specific skills deficits or reduce interfering behaviours (table 1, right column) are widespread and well established methodologies that are not considered in this review. Rather, we refer to *comprehensive* ABA programmes that employ a broad curriculum targeting multiple skills such as imitation, cognition, communication, social interactions and compliance in sequences that are individualised for each child. Comprehensive ABA interventions for autism (table 1, left column) started with Lovaas' early intensive behavioural intervention (EIBI) program. EIBI usually starts with intense, highly structured one-on-one teaching in the home relying on discrete trial training (DTT) to build foundational skills. Thereafter, the intervention targets more functional skills (play, dressing...) in other settings (school) that include peers and other interaction partners.

Comprehensive ABA interventions were later enriched by a series of naturalistic developmental behavioural interventions (NDBIs) more suitable for preschool children and toddlers, and that focus on developmental appropriateness, child's choice of activities and on relationship building with the interventionist. Both DTT-EIBI and NDBIs have manuals, establish individualised treatment goals, require substantial therapist training and supervision, have tools to assess fidelity of implementation, and collect observation-

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al data to analyse the problem, evaluate treatment response and monitor progress over time. Unlike DTT-EIBI, therapists in NDBIs follow the child's lead and choice of activity or routine, teach in natural settings with real life materials, enhance social motivation using natural reinforcers, reward attempts as well as successes, and use as well classical behavioural techniques such as prompting, modelling, fading and imitation [2].

Much research on ABA was established using single-case experimental designs whereby a treatment is administered by the investigator in different phases to the same subject, who acts as their own control; measured outcomes are then related to treatment phases to draw inferences on their causal relationship. Although these n-of-1 studies generated useful findings, group comparison of treatment effects following randomisation remains the standard to evaluate treatment efficacy.

Lovaas' initial study [1] required 40 hours/week of one-on-one treatment in the home, with strong parent involvement on a small (n = 19) sample of 32-month-olds compared with a low intensity control group. A follow-up at school age concluded that the intensive treatment was superior and claimed recovery ("normal intellectual and educational functioning") in 47% of subjects vs 2% among controls. However, methodological shortcomings were serious: there was no randomisation, baseline characterisation of subjects was minimal, aversives were used (slaps on the thigh), fidelity and actual treatment time were not measured, and evaluation of recovery was not standardised. In 2000, a methodologically improved replication study [3] showed much more modest effects, with end-point IQs still in the impaired range, and limited progress of children with more severe autistic disorder at baseline. A recent meta-analysis of five studies (pooled n = 202) of Lovaas' EIBI showed average significant gains in IQ (15 points), in adaptive behaviour (9 points), in communication (11 points) and more modest gains in receptive and expressive language, but no effect on autism severity or behavioural problems [4]. Of note, sample sizes were small (<50 for the combined experimental and control groups) and post-treatment maintenance of gains was rarely examined.

The naturalistic interventions (NDBIs) have shown gains of the same magnitude. Thus, compared with community treatment as usual, the Early Start Denver Model (ESDM) initial randomised study documented significant IQ, expressive and receptive language, and adaptive behaviour gains of about one standard deviation in 18- to 30-month-old children (n = 24) treated for 2 years and compared with 21 community (treatment as usual) controls [5]. However, a replication study on 118 14- to 24-month-old toddlers enrolled in three separate treatment sites largely failed to replicate the initial results; a modest benefit on a composite language outcome was obtained at only two of the three sites, and after analyses that departed from the original plan [6]. As well, in a randomised controlled trial of 98 toddlers aged 14 to 24 months, a 12-week low-intensity parent-administered version of ESDM failed to show superior results when compared with community treatment as usual [7]. Another naturalistic intervention, JASPER (Joint Attention Symbolic Play Engagement Regulation), is a modular intervention that targets specific foundational skills known to predict improved language, cognitive, social and other outcomes in autism spectrum disorder (ASD). It has been now evaluated in several randomised controlled trials with a consistent demonstration of its efficacy. In a first randomised controlled trial of 58 preschool children with ASD, deficits in joint attention and play skills were significantly reduced by the intervention (trained therapists, 30 minutes daily for 6 weeks) [8], which also led to increased use of language 1 year later [9]. A companion randomised controlled trial of 38 toddlers aged 30 months showed large improvements in joint attention and functional play with an 8-week (three 1-hour sessions/week) caregiver-mediated intervention [10]. Gains in joint engagement were maintained 1 year later. In another randomised controlled trial, a 10-week parent-delivered intervention showed superiority (effect size 0.69) over parent psycho-education in 86 toddlers aged 22 to 36 months for both joint attention and symbolic play [11]. JASPER's efficacy has now been confirmed in several replications across different groups of investigators and different settings (delivered by teachers in schools, by trained therapists or parents, in underserved groups, with toddlers or school age participants, or with minimally verbal children [12–15]). Despite its lower duration and intensity compared with other comprehensive ABA packages, the JASPER intervention has currently the strongest data supporting immediate efficacy, as well as maintenance of gains post-treatment. Gains in language production of similar magnitude have been reported in randomised studies of Pivotal Response Training (PRT) administered in parent group and individual child-therapist sessions combinations either for 12 weeks [16] or 24 weeks [17] in samples of preschool children with ASD and language delay. A recent meta-analysis has summarised the results of both EIBI and NDBI programmes, alongside other interventions [18]. When all studies were considered, both EIBI and NDBI were as-

Table 1: ABA nomenclature

Comprehensive ABA interventions	Core skills ABA interventions
• **Lovaas/UCLA Early Intensive Behavioural Intervention** Discrete Trial Training (DTT) • **Naturalistic developmental behavioural interventions (NDBI)** Pivotal Response Training (PRT; Koegel and Koegel) Applied Verbal Behaviour Model Joint Attention Symbolic Play and Engagement Regulation (JASPER; Kasari) Early Start Denver Model (ESDM; Rogers and Dawson)	**Skill deficits** ***Communication*** Sign language PECS (Picture Exchange Communication System) Augmentative Communication ***Social impairment*** Joint attention, symbolic play, imitation (RIT: reciprocal imitation training) Social skills training RDI, peer mediated interventions Theory of mind, social stories **Behaviour reduction** ***Core ASD symptoms*** Repetitive behaviours ***Associated symptoms*** Aggression, self-injurious behaviour, irritability, etc.

sociated with significant gains of comparable magnitude in cognition, language, play and social communicative behaviour. However, when only randomised controlled trials were included, only NDBIs programmes could reliably demonstrate such positive gains.

Four issues require special consideration. First, most treatments were devised and initially tested with small, highly selected samples, in academic centres. As illustrated above, first successes were not always confirmed in replication trials. Moreover, effectiveness studies examining the impact of ABA interventions in community-based settings often show smaller effects when compared with university hospital-based studies [19]. Several factors may account for this difference, including highly selected samples in original randomised controlled trials, lesser therapist training and supervision in community settings, or lower fidelity of implementation and delivery. Second, randomised trials have compared active treatments with eclectic treatment as usual that varies across settings and over time. No randomised trial has yet compared head to head two different types of comprehensive ABA interventions; in the few non-randomised quasi-experimental studies that performed such comparisons, results have usually shown no superiority of any one programme [20]. Third, the evidence for the importance of dose (how many hours/week at least should an intervention be delivered?) or of age of treatment initiation (is very early intervention more beneficial? Is there a critical age beyond which efficacy is lessened?) is, at most, mixed. Gains associated with less intensive and costly interventions such as parent-mediated JASPER or PRT show equivalent or even better effect sizes than more intensive ABA programmes. As for optimal age at treatment initiation, use of a parent-mediated intervention with 7 to 10 months old infants at risk of ASD showed inconsistent effects in a randomised trial [21]. A more adequately powered randomised controlled trial of the same intervention (iBASIS) on 103 9- to 14-month-old infants with emerging signs of autism also failed to show efficacy [22]. Likewise, the low-intensity 12-week parent administered version of ESDM for toddlers at risk for ASD did not show efficacy in another randomised trial [7]. Finally, with few exceptions [23], research has been exclusively conducted in developed countries. Implementation of costly ABA programmes in low-income countries and/or with low-resources communities remains seriously challenging. Addressing this gap, the World Health Organization has developed the Caregiver Skills Training (CST) intervention (based on JASPER and other ABA methods), which is designed to empower families with practical skills, can be flexibly implemented in different low-resource communities and delivered by different professionals. CST is pilot tested in 17 countries and results should be available in 2020 [24].

The evidence base for evaluating the efficacy and effectiveness of comprehensive ABA interventions in autism is still thin (table 2) although there was a remarkable increase in the number of randomised, well-controlled clinical trials in the last 15 years. Among the many questions that remain to be addressed are: Do the statistically significant benefits reported in some trials translate into clinically meaningful and sustained long-term benefits? What are the critical ingredients of each comprehensive ABA intervention? In which sequence should they be employed? Which type of ABA programme works best for which child? What could be learnt from systematic cost-benefit analyses that have not yet been performed? Is response to ABA associated with long-term functional and structural positive changes in neuronal circuitry?

Thus, much remains to be learnt but ABA interventions are, today, the most evidence-based intervention available to improve the outcome of autism.

Key messages

- ABA is a short acronym loosely used to describe a family of teaching procedures to learn a range of skills; when applied to autism, it is important to specify which ABA technique or programme is envisaged.
- Comprehensive ABA interventions (which include Lovaas' and more naturalistic developmental behavioural interventions) target multiple skills and have been shown to improve developmental trajectories in children with autism with respect to cognitive, play, language and sociocommunicative behaviour.
- The less intensive and shorter naturalistic intervention JASPER has shown efficacy in several randomised controlled trials that replicated its effects across settings (clinic vs community), samples (underserved or not), and delivery type (professionals, parents, teachers).
- Evidence for improved outcomes with interventions starting at a very early age or with high levels of intensity (40 hours/week) is lacking.
- The effectiveness of ABA interventions is lessened in real-world, community settings; however, an encouraging low-cost intervention, applicable to low-resource countries and settings, is currently being tested.

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Table 2: Strengths and limitations of comprehensive ABA interventions.

Strengths	Limitations
• Has good and replicated evidence for efficacy • Is data driven, target behaviours and outcomes are measurable • Can be individually tailored • Applicable across settings and lifespan • Less intense/costly versions work (JASPER)	• Claims about effect sizes and universal response are often exaggerated • Paucity of rigorous randomised controlled trial • Unclear which "package" is better for whom? • Requirement of therapist training and intense supervision • Costs +++ • Uncertain generalisation and post-treatment maintenance of gains • Adolescent/adult studies needed • Long term beneficial effects? • Not easily scalable/portable or cross-culturally relevant • Ethical concerns (Lovaas EIBI)

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