

NZHTA TECH BRIEF SERIES

April 2004

Volume 3 Number 1

What is the evidence for the effectiveness of behavioural and skill-based early intervention in young children with Autism Spectrum Disorder (ASD)?

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This report should be referenced as follows:

Doughty, C. What is the evidence for the effectiveness of behavioural and skill-based early intervention in young children with Autism Spectrum Disorder (ASD)? *NZHTA Tech Brief Series 2004; 3(1)*

Titles in this Series can be found on the NZHTA website:
http://nzhta.chmeds.ac.nz/earlyintervention_autism.pdf

2004 New Zealand Health Technology Assessment (NZHTA)

ISBN 1-877235-60-1

ISSN 1175-7884

ACKNOWLEDGEMENTS

This Tech Brief was commissioned by the New Zealand Ministry of Health. The report was prepared by Dr Carolyn Doughty (Research Fellow) who selected and critically appraised the evidence. The literature search strategy was developed and undertaken by Ms Margaret Paterson (Information Specialist). Mrs Ally Reid (Administrative Secretary) provided document formatting. Internal peer review was provided by Dr Ray Kirk (Director) and Dr Claire Tochel (Research Fellow).

We are grateful to the following people who provided background information, valuable specialist advice and clinical input:

- Dr Matt Eggleston, Child and Adolescent Psychiatrist and Clinical Head of the Child and Family Specialty Service, Christchurch, New Zealand.
- Margaret Jenkins, Social Worker, Child and Family Specialty Service, Christchurch, New Zealand.
- Maryska Tawroszewicz, Clinical Psychologist, Child and Family Specialty Service, Christchurch, New Zealand.

Other people contacted by email included:

- Dr Connie Kasari, University of California, Los Angeles, United States of America.
- Dr Ole Lovaas, University of California, Los Angeles, United States of America.
- Dr Yashwant Sinha, Royal Children's Hospital, Melbourne, Australia.
- Dr Gregory Stores, Department of Psychiatry, University of Oxford, United Kingdom.
- Dr Tony Charman, Institute of Child Health, London, United Kingdom.

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LEVEL OF EVIDENCE CONSIDERED IN TECH BRIEFS

Tech Briefs are rapidly produced assessments of the *best available evidence* for a topic of highly limited scope. They are less rigorous than systematic reviews. Best evidence is indicated by research designs, which are least susceptible to bias according to the National Health and Medical Research Council's (NHMRC) criteria (see **Appendix 1**). Where methodologically acceptable and applicable, appraised evidence is limited to systematic reviews, meta-analyses, evidence-based clinical practice guidelines, health technology assessments and randomised controlled trials (RCTs). Where not available, poorer quality evidence may be considered.

CONFLICT OF INTEREST

None.

BACKGROUND

This Tech Brief was requested by Mr Lester Mundell, Chief Advisor, Disability Issues Directorate, Ministry of Health, New Zealand Government.

Autism is a life-long developmental disability, which can affect communication, social interaction, and behaviour. Its form and severity can vary from person to person, but people with autism share some difficulty in making sense of the world. First illustrated in a series of case histories by Leo Kanner in 1943, it was not until 1980 that autism was officially recognised as a disorder of development distinct from childhood schizophrenia (American Psychiatric Association 1994). People with features similar to classic, Kanner-type autism have been identified who do not fulfil the same specific criteria for autism. To recognise this, a broader range of pervasive developmental disorders including Asperger's Syndrome, Pervasive Developmental Disorder – Not Otherwise Specified (PDD NOS)/Atypical Autism are now considered to be part of the autistic spectrum (although classification is contentious). This is collectively known as Autism Spectrum Disorder, or ASD. For simplicity, this Tech Brief will use the term “autism” and ASD interchangeably to refer to people across the spectrum of ASD.

Core features of ASD are evident in three of the following behavioural areas:

1. social interaction
2. language and communication
3. restrictive interests and repetitive behaviour.

These three areas are sometimes referred to as the “triad of impairment”. Onset of abnormal or delayed functioning in at least one of these areas is expected before the age of three years. Associated features of ASD sometimes evident include unprovoked aggressive or violent behaviour to self and others, attention, concentration and sleep problems, unusual responses to sensory stimuli and special skills and interests. The triad of core features forms the basis for diagnostic criteria used by the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) (American Psychiatric Association 1994) and the International Classification of Diseases (ICD-10) (World Health Organization 1992) to classify ASD (see **Table 1**).

Table 1. Diagnoses relevant to Autism Spectrum Disorders

Classification System	Disorders/syndromes (Code)
DSM-IV	<ul style="list-style-type: none"> ▪ Autistic Disorder (299.00) ▪ Pervasive Developmental Disorder – Not Otherwise Specified (299.80) ▪ Asperger's Syndrome (299.80) ▪ Rett's Disorder (299.00) ▪ Childhood disintegrative disorder (299.10)
ICD-10	<ul style="list-style-type: none"> ▪ Childhood autism (F84.0) ▪ Atypical autism (F84.1) ▪ Rett's Syndrome (F84.2) ▪ Asperger's Syndrome (F84.5) ▪ Other Pervasive Developmental Disorders (F84.8) ▪ Pervasive Developmental Disorder – unspecified (F84.9)

Autism can be diagnosed accurately in many children by or before three years of age (Stone et al. 1999), (Filipek et al. 1999). Yet it often remains undiagnosed until or after late preschool age because appropriate tools for routine developmental screening and screening specifically for autism are minimally used based on lack of availability, accessibility or awareness by health professionals (McGahan 2001). Milder difficulties in the autistic spectrum, including Asperger's Syndrome and PDD NOS may not present until later, if at all and are often more difficult to recognise. Once any diagnosis is confirmed, prompt access to clinical and support services are required by the child and their caregiver/s.

Problem behaviours and specific skill deficits are a common concern for parents of young children with autism, as they can become major barriers to effective education and social development. Epidemiological studies suggest that up to one third of young children with autism engage in problem behaviours that warrant intervention. Typical problem behaviours may include physical aggression, self-injury, property destruction, pica, stereotypy, defiance and tantrums (Baird et al. 2003; Horner et al. 2002). For this reason, behavioural interventions are often used to promote both social and adaptive behaviour in children with autism. Behavioural therapy is essentially the application of basic psychological principles of learning to human behaviour. Experimental analysis of behaviour has led to the application of these principles to change behaviour and applied behavioural analysis (ABA) refers to the application of these principles to improve socially important behaviours (McGahan 2001). They can include specific approaches (for example, Lovaas Therapy/ABA) that are used to help children acquire or change behaviours (or skills) by reinforcing adaptive responses and suppressing non-adaptive responses or they may form part of a broader social-developmental treatment programme. Historically, treatment programmes for children with autism have had a much greater focus on developing or supporting the demonstration of specific skills rather than the promotion of relatedness and attachment. However intervention models which prioritise the developmental dimensions of social communication and emotional regulation are gaining recognition. Comprehensive or integrated treatment programmes may also seek to include components of speech and language therapy or children may be already receiving this as part of their usual care.

Assessing the efficacy of early intervention for autism is not straightforward. Researchers have only recently begun to systematically address the inherent methodological issues and problems that arise in attempting to conduct trials for treatment of a disorder that is rare and has a very early age of onset. One of the key issues arising within a clinical setting is that children with autism generally receive multiple interventions and it is difficult to determine what it is about a particular treatment that may result in change (Kasari 2002).

To date, experts have recommended early diagnosis and intensive therapy to improve a child's ability to communicate, adapt to change, and to develop social skills (Volkmar et al. 1999). Intense, specialised, integrated programmes purport to be among those that produce the greatest gains. These programmes build upon a child's interests, offer a predictable schedule with parental involvement, teach tasks in a series of simple steps, actively engage a child's attention in highly structured activities, provide regular reinforcement of appropriate behaviours and aim towards generalisation of skills across environments (McGahan 2001).

Therefore, the focus of this Tech Brief is to consider the most *recent* and *best* evidence for the effectiveness of behavioural and skill-based early interventions that are used to manage ASD in young children.¹

SELECTION CRITERIA

Study inclusion criteria

Publication type

Studies published between January 2000 and December 2003 inclusive in the English language, including primary (original) research (published as full original report) and secondary research (systematic reviews and meta analyses) appearing in the published literature.

Intervention

Any formal behavioural or skill-based intervention aimed at treating or managing symptoms and associated conditions of ASD in young children.

¹ Due to the nature of this topic the information presented in the Evidence Tables is not restricted to a single intervention but instead refers to a number of interventions with a similar theoretical basis and/or comprehensive programmes that utilise either an intensive behavioural or social-developmental approach.

Sample

Study population primarily those (at least 75% of sample) with Autism Spectrum Disorder (ASD) as classified by or consistent with DSM-IV and/or ICD-10 (see **Table 1, page 1**), or where results are reported separately for this group.

Sample mean under eight years of age, or within the age range of one to seven years.

Studies that were not restricted to participants within this age range, but where results were reported separately on a subgroup of participants within this age range.

Studies with sample sizes of five or more people receiving the early intervention or comprehensive programme.

Setting

Early intervention or comprehensive programme used by parents or mental health services to prevent, manage or treat features of autism in young children.

Outcomes

Studies using at least one standardised and/or quantitative outcome measure relating to effectiveness of relevant interventions in achieving improvement of core features of ASD; improvement of associated features of ASD; global impression rating scales; and quality of life questionnaires for individuals with ASD and/or family/carers. These will include but not necessarily be limited to, the following measures and diagnostic or screening tools:

- Wechsler Intelligence Scales for Children-Revised (WPPSI-R or WISC-R)²
- Autism Diagnostic Interview-Revised (ADI-R)
- Checklist for Autism in Toddlers (CHAT)
- Bayley Scales of Infant Development-Revised (BSID-R)
- Merrill-Palmer Scale of Mental Tests (MPSMT)
- Reynell Developmental Language Scales (RDLS)
- Vineland Adaptive Behavior Scales (VABS)
- Griffiths Scale of Mental Development (NVIQ),
- MacArthur Communicative Development Inventory (CDI)
- Early Social Communication Scales (ESCS)
- Pre-Verbal Communication schedule (PVCS)

Primary outcomes of interest are those relating to behavioural change and the development of reciprocal social interaction and/or communication skills.

Study design

Studies will be considered for this review if they used one of the following study designs:

- systematic review of studies eligible for inclusion in this review (this will include relevant Health Technology Assessment reports which may or may not critically appraise primary studies)
- randomised controlled trials (including cross-over trials)
- pseudorandomised controlled trials (alternate allocation or some other method)
- concurrent controls or cohort studies
- case-control studies.

² Wechsler Intelligence Scales for Children-Revised (WPPSI-R or WISC-R) are similar instruments that cover a range of ages from three to seven years and six to 16 years respectively.

Study exclusion criteria

The following criteria will be used to exclude studies from appraisal.

Studies reporting on:

- pharmacological or dietary interventions, including secretin and vitamin therapy
- any other treatments, which aim to have a physiological effect
- auditory and sensory integration³
- specific learning difficulties where associated behavioural problems are not reported separately
- surgical interventions, or physical therapy including massage.

Studies that:

- reported outcomes for a study population with a mean or median sample age of eight years or older, or with an age range outside one to seven years
- had samples of fewer than five participants in either treatment or control group
- did not use standardised and/or previously validated outcome measures, with the exception of assessment of specific child skills
- looked solely at language outcomes, socio-emotional abilities or functioning but without any attempt by investigators to use behavioural techniques to develop skills or address behavioural problems
- are non-published work
- were non-systematic narrative reviews, editorials or expert opinion
- were correspondence, commentaries, book chapters, articles published in abstract form
- were single case presentation/series/reports, this includes experimental studies that used multiple baseline, post-test or pretest/post-test (“before-and-after”) designs
- did not clearly describe methods and results or had significant discrepancies
- were not published in English.

Any identified unpublished or ‘grey’ literature will be included for New Zealand specific studies only where they meet selection criteria.

³ For further information refer to Tochel C. Sensory or auditory integration therapy for children with autistic spectrum disorders. In Bazian Ltd (ed) STEER: Succinct and Timely Evaluated Evidence Reviews 2003; 3(17). Wessex Institute for Health and Development, University of Southampton and Bazian Ltd. [WWW document] URL <http://www.signpoststeer.org/> or Sinha Y, Silove N, Wheeler D, Williams K. Auditory integration training and other sound therapies for autism spectrum disorders (Cochrane Review). In: The Cochrane Library, Issue 1, 2004. Chichester, UK: John Wiley & Sons, Ltd.

MAIN SEARCH TERMS

Details of the search strategy are presented in **Appendix 2**.

- **Index terms from Medline MeSH terms:** asperger syndrome, autistic disorder, rett syndrome, child development disorders pervasive, exp behavior therapy, cognitive therapy, program evaluation, early intervention, evaluation studies, exp psychotherapy, treatment outcome.
- **PsychInfo headings:** autism, pervasive developmental disorders, aspergers syndrome, autistic thinking, rett syndrome, autistic children, early infantile autism, behavior therapy, behavior modification, cognitive therapy, parent training, early intervention, program evaluation, educational program evaluation, social skills training, exp social integration.
- **Embase headings:** autism, asperger syndrome, rett syndrome, infantile autism, behavior therapy, cognitive therapy, health program, parent, evaluation, psychotherapy, health care quality.
- **Cinahl headings:** autism, child development disorders, rett syndrome, behavior modification, behavior therapy, cognitive therapy, early intervention, early childhood intervention, exp psychotherapy, program evaluation, exp evaluation research.
- **Other keywords:** therapy adj3 condition*, behavio?r* adj3 modif*, behavio?r* adj3 therap*, behavio?r* adj3 intervention*, cognit* adj3 (therap* or psychotherap*), autis*, kanner's, behavio?r, intervention*.

SEARCH SOURCES

The NZHTA CORE Search was employed. Characteristics of the Core search include: essential sources only, major databases and secondary sources, and mostly published and indexed literature. For more detail about the search sources refer to the NZHTA Search Protocol at <http://nzhta.chmeds.ac.nz/nzhtainfo/protocol.htm> Steps 1-9 (Core sections).

Principal sources of information were

Bibliographic databases

Medline

Embase

Cinahl

PsychInfo

Amed

ERIC

Current Contents

Web of Science

Index New Zealand

Review databases

Evidence-based medicine reviews

Cochrane Database of Systematic Reviews

DARE

NHS Economic Evaluation Database

Health Technology Assessment Database

Other sources

In the preliminary search undertaken for this review, additional sources searched included website sources such as evidence-based and guidelines sites, government health websites, related health professional association websites, and major websites.

For more detail about the search sources refer to the NZHTA Search Protocol at <http://nzhta.chmeds.ac.nz/nzhtainfor/protocol.htm> Steps 1-15 (Core and Standard sections), and steps 16-17, 19-20 (Ideal section).

Hand searching of journals, contacting of manufacturers, or contacting of authors for unpublished research was not undertaken in this Tech Brief.

Articles published in English language only were considered. Searches were limited to English language material published from 1990 onwards. The searches were completed 9 January 2004.

APPRAISAL METHODOLOGY

Summaries of appraisal results are shown in tabular form (known as *Evidence Tables*) which detail study design, study setting, sample, methods, results, reported conclusions and NZHTA reviewer conclusions/comments based on the limitations and validity of the study.

The evidence presented in the selected studies were assessed and classified according to the NHMRC's revised hierarchy of evidence (**Appendix 1**). This report uses New Zealand terminology throughout, except for within the Evidence Tables which use the terminology (including spelling) of each original study.

RESULTS

From the above search strategy we identified 487 relevant articles/abstracts published since 2000, of which 108 were retrieved. Of these, 98 did not fulfil the inclusion criteria and were excluded. These papers, annotated with the reason for exclusion, are presented in **Appendix 3**. Reasons for exclusion included the following:

- expert opinion or narrative review (n=20)
- descriptive study or intervention or programme description (n=20)
- experimental study or systematic review of experimental studies (n=13)
- correspondence or commentary or book review/chapter (n=13)
- outcomes not reported or measured (n=9)
- conference proceedings or abstracts (n=7)
- case report with sample size of fewer than 5 in intervention or control group (n=6)
- not relevant to the topic (n=5)
- not population of interest (n=4)
- subjects outside age range (n=1).

Ten retrieved articles were appraised (listed in **Appendix 4**). Included papers are presented in the evidence table below. Included primary studies were all level III.2 and above according to NHMRC's hierarchy of evidence, and a further five secondary articles (systematic reviews of randomised controlled trials or health technology reports) were also appraised. Another 15 original articles (published prior to 2000) were retrieved from citation searching because they were either summarised or critically appraised in the systematic reviews included in this Tech Brief.

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
<p>Bassett et al. (2000)</p> <p>British Columbia, Canada</p> <p>British Columbia Office of Health Technology Assessment (BCOHTA)</p> <p>Level of evidence: I</p>	<p>Systematic Review</p> <p>Aim To examine whether early, intensive behavioural therapy for children with autism results in normal functioning, or essentially a cure.</p> <p>Search method Electronic searches from Current Contents (1996-1999), Embase (1988-1999), HealthStar (1975-1999), Medline (1966-1999) and combined Science and Social Sciences Index (1989-1999). Citation searching was performed on article reference lists. Other web library catalogues, commercial and in-house databases and peer-reviewed internet sites were also searched.</p> <p>Search terms included Autistic disorder, asperger, kanner, lovaas, early intervention, intensive, discrete trial learning, applied behavioural analysis. Co-authors of Lovaas and authors citing Lovaas were searched.</p> <p>Participants Studies on human paediatric (pre-school) populations with autism.</p>	<p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ reports were included if interventions were described as early, applied behavioural analysis, behavioural therapy or intensive home-based programs ▪ all studies examining some form of overall autism treatment program ▪ early behavioural therapy, where initiation of therapy when diagnosis is made and prior to age 5 when kindergarten services are available ▪ if study measured overall function including intellectual functioning, language, social interaction and play, adaptive or self-care skills, maladaptive behaviour ▪ if study design included a treatment and control group ▪ no exclusion due to presence of co-morbidity. <p>Exclusion criteria</p> <ul style="list-style-type: none"> ▪ individual case reports and case series ▪ studies that were limited to training trials (short intensive efforts to alter a child's communicative or social skills in a particular domain). <p>Outcome measures Specific measures varied from study to study (refer to original study for further detail).</p>	<p>Two reviewers independently assessed and selected studies for inclusion. The search strategy identified 1200 abstracts and citations in total. Approximately 150 articles met minimum inclusion criteria and were retrieved. Criteria were then reapplied to full published reports to determine appropriateness for full critical appraisal.</p> <p>No studies comparing alternative behavioural programs or RCTs of behavioural intervention programs were identified as meeting inclusion criteria.</p> <p>Evaluations of efficacy by BCOHTA Primary data included four controlled studies of treatment</p> <ul style="list-style-type: none"> ▪ Lovaas (1987) and McEachin et al. (1993) A prospective, non-randomised study comparing active treatment group (reported as 40 or more hours of 1: 1 behavioural therapy for 2 or more years) to two control groups. Clinically and statistically significant different between groups on overall outcome variables, IQ and function in classroom without special support. ▪ Birnbauer and Leach (1993) A prospective study on 11 children (reports on n=9), included a non-treated, matched control group. They reported increases in non-verbal IQ and improvements in language in the treated group compared with controls. Did not validate success rate of Lovaas where half of subjects achieved 'normal functioning'. ▪ Sheinkopf and Siegel (1998) A retrospective, controlled study methodology. No prospective experimental design. Treatment group had a mean IQ 25 points higher than control group (p=0.01). No group differences on symptoms, treated group had decreased symptom severity (p=0.01). No information on language development, adaptive behaviour, academic or social functioning. No children reported as attaining 'normal' or 'near normal' functioning. ▪ Ozonoff and Cathcart (1998) A ten-week, home program service based on the Treatment and Education of Autistic and Communication-Handicapped Children (TEACCH) curriculum (an alternative to Lovaas). Described as structured teaching as opposed to behavioural therapy, intended to maximise existing skills. Prospective, controlled trial using overall outcomes. Results suggested that auxiliary home interventions increase developmental functioning in young children, above and beyond gains due to school-based services. No information on adaptive behaviour, behaviour problems or social functioning. 	<p>The BCOHTA report lists the following limitations</p> <ul style="list-style-type: none"> ▪ the Lovaas (1987) study does not compare two different therapies, but two levels of intensity in a single form of behavioural intervention; group assignment was not randomised to different treatment conditions; children studied were referred to program following diagnosis by affiliated but independent institution so they may not be a cross-section of children with autism ▪ the Birnbauer (1993) study did not compare treatment options, there was no randomisation, and furthermore the sample is so small it is more like an uncontrolled case series. The authors provide no details regarding diagnostic tests, criteria or reliability or any description of treatment compliance or consistent application. Although there were also a number of strengths this study was too small, too short in duration and too methodologically weak to provide strong evidence for or against Lovaas therapy.

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
Bassett et al. (2000) British Columbia, Canada British Columbia Office of Health Technology Assessment (BCOHTA) Level of evidence: I (Continued)			<p>Secondary data included five systematic reviews that included critical appraisal of the primary data Rogers (1998), Tregear et al. (2000), Howlin (1997), Green (1996) and Smith (1998) and the critical appraisal debates following Lovaas (1987) and McEachin et al. (1993). Author's conclusions below as reported by BCOHTA.</p> <ul style="list-style-type: none"> ▪ Rogers (1998) "Studies did not demonstrate the level of improvement in multiple areas of functioning or the sustained long-term effects of the treatment that Lovaas reported". ▪ Tregear et al. (2000) "Although it appears possible to improve some aspects of function in children with autism, it is not clear that any one program is more effective than another". ▪ Howlin (1997) "For the present, Lovaas program confirms the power of behavioural interventions. The true extent of the benefits, however requires greater exploration and longer term evaluations". ▪ Green (1996) "Applied behavioural analysis is the treatment of choice, at least 30 hours of 1:1 therapy per week by individuals with extensive training. BCOHTA comments that this systematic review was the one with the least critical appraisal". ▪ Smith (1998) "Some outcome studies indicate a major breakthrough may have occurred. Nevertheless studies require replications with improved research methodologies". <p>BCOHTA author's conclusions The effectiveness claim of Lovaas et al. (1987) that half the children achieved normal or near-normal development and placement in schools remains uncorroborated by independent research. The outcome evidence does not support adopting a single entire program, whether Lovaas or alternative methods, based on the likelihood of achieving normal children. Due to major methodological flaws, the findings of Lovaas and McEachin cannot be regarded as conclusive; more research is required before authoritative statements can be made about efficacy. In the absence of scientifically valid studies of overall autism programs, they suggest it may be prudent to revise existing autism services using incremental, not programmatic steps.</p>	<ul style="list-style-type: none"> ▪ the Sheinkopf and Siegel (1998) study used a very weak, partially retrospective, observational study design. Provides some support of benefit from the Lovaas method. Due to little description of any treatment effect or of central coordination of the intervention, impossible to determine any causal inference relating to limited gains reported for these children ▪ the Ozonoff and Cathcart (1998) study did not compare home-based treatment options, was not randomised and did not follow a specific protocol or manual. It relied exclusively on internal evaluators, aware of treatment allocation and did not report method of clinical diagnosis. Parents were not taught a pre-defined set of skills so it is hard to assess degree of consistency with which intervention was applied. <p>Reviewer's comment A thorough review that highlights the lack of well-conducted research in this area, however this review does not allow conclusions to be drawn about best practice but does point the way forward for monitoring the state of effectiveness evidence.</p>

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
<p>Diggle et al. (2003)</p> <p>Newcastle, UK</p> <p>Cochrane Developmental, Psychosocial and Learning Problems Group</p> <p>Level of evidence: I</p>	<p>Systematic Review</p> <p>Aim To review the effectiveness of parent-mediated early intervention for young children with autism spectrum disorder compared with no treatment, waiting list group or a different form of intervention.</p> <p>Search method Electronic searches from ERIC (1966-present), Cochrane CTR Issue 1 (2002), Medline (1966-present), EMBASE (1980-2002), PsycINFO (1887-2002), CINAHL (1982-2002), Dissertation Abstracts International (1861-2002), Social Sciences Abstracts (1980-2002), Sociological Abstracts (1963-2002), Linguistics and Language Behavior Abstracts (1973-2002), National Research Register Issue 1 (2002), LILACS (1982-2002).</p> <p>Key journals (Journal of Autism and Developmental Disorders and the Journal of Child Psychology and Psychiatry and Allied Disciplines) were hand searched to identify studies not electronically catalogued.</p> <p>Search terms included Autism, pervasive developmental disorder, PDD, language delay, communication or speech disorder, childhood schizophrenia, Kanner, Aspergers, parent, caregiver, family, treatment, therapy, intervention, programme, training, rehabilitation.</p> <p>Participants Studies on children aged 1 year to 6 years 11 months diagnosed with Autism, Asperger syndrome, pervasive developmental disorder (PDD), PDD not otherwise specified (PDD-NOS).</p>	<p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ randomised or quasi-randomised studies where the interventions focus were on parent-implemented early intervention ▪ any intervention with a significant focus on parents as mediators, where training was provided on a group or individual basis. <p>Exclusion criteria Studies, which include drug treatments, or treatments that aim to have a physiological effect (such as dietary intervention) and surgical interventions.</p> <p>Outcome measures Studies with standardised or previously validated child related outcome measures, including assessment of specific child skills.</p> <p>Primary outcomes</p> <ul style="list-style-type: none"> ▪ child language progress ▪ child positive behavioural change ▪ parent interaction style. <p>Secondary outcomes</p> <ul style="list-style-type: none"> ▪ parent confidence ▪ reduction in levels of parental stress. 	<p>Two reviewers independently assessed and selected studies for inclusion. There was 100% concordance over studies selected.</p> <p>The search strategy located over 15,000 articles, including duplications and articles not relevant to the current topic. From this initial number, 68 articles were retrieved and formally reviewed.</p> <p>Of the 68 studies, five were published in a language other than English. Sixty-six of the studies obtained in full for review were excluded from the review, primarily on methodological grounds (n=55). Few studies involved a control group. Some were excluded as participants did not have a diagnosis of ASD (n=8), or the intervention did not focus on parent-mediated early intervention (n=1).</p> <p>Two RCTs were included in the final review.</p> <p>Jocelyn et al. (1998) carried out a RCT comparing parent training to community day care (usual service). There was one dropout from the parent-training group. Parent mediated early intervention was found to have a significant effect on child language but the reasons for this were unclear.</p> <p>Smith et al. (2000) carried out a RCT to compare intensive treatment (from student therapists) to parent training, both were based on the Lovaas treatment manual. No drop-outs were reported. Significant change was found on the two ability measures in favour of the intensive group but the long-term effects of intensive treatment did not score any higher than parent training for children's everyday living skills and co-operative behaviour.</p> <p>Author's conclusions The two studies are not directly comparable. Both studies raised more questions than they answered. The authors suggest the results should be viewed with great caution due to the small number of included studies and inherent differences between the two studies. Although they both used an intervention based broadly on behavioural principles, the two studies differed in theoretical approach, intensity, duration, context and the type of intervention comparison.</p>	<ul style="list-style-type: none"> ▪ clearly defined and specific selection criteria ▪ comprehensive search strategy that involved database searching and handsearching ▪ inclusion criteria established in advance although this was altered slightly after the protocol was first published ▪ non-English articles were translated to the degree necessary to be able to understand the essentials of the study ▪ it is not possible to blind either treatment facilitators or parents to the type of treatment being implemented or received but both studies used blind assessors ▪ no intent-to-treat analysis in Jocelyn et al. (1998) study ▪ neither study reported any power calculations and both had limited sample size, although these were above those often reported for autism research ▪ insufficient detail about recruitment so no judgement possible about representativeness of samples ▪ too few studies included to assess any publication bias. <p>Reviewer's comment A thorough review that highlights the lack of well-conducted research in this area, however this review does not allow conclusions to be drawn about best practice.</p>

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
Finch and Raffaele (2000) Ontario, Canada McMaster University Level of Evidence: III.2	<p>Systematic Review</p> <p>Aim To review whether Intensive Behavioral Intervention (IBI) results in positive gains for children with autism.</p> <p>Search method Electronic databases MEDLINE, CINAHL, AMED, Cochrane Reviews and occupational therapy specific databases were searched.</p> <p>Search terms included Autism, pervasive developmental disorder (PDD), early IBI and efficacy/effectiveness. Relevant books, community clinicians were consulted for background information and sources were reviewed for current statistical and legislative information. No handsearching was reported.</p> <p>Participants Studies on children in early childhood (under the age of eight) who were diagnosed with PDD or autism.</p>	<p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ those published in journals and involving experimental trials ▪ studies looking at IBI with children in early childhood. <p>Exclusion criteria</p> <ul style="list-style-type: none"> ▪ descriptive studies. <p>Outcome measures WISC-R, Stanford-Binet Intelligence Scale.</p> <p>No other measures specifically mentioned.</p>	<p>The literature search resulted in seven articles, which fitted the inclusion criteria. Critical appraisal of each study was completed using a previously developed review form.</p> <p>The articles reviewed described seven different studies including: three quasi-experimental matched pairs designs comparing an IBI group to a less intensive treatment group and a no treatment group (Anderson et al. 1987, Birnbauer and Leach 1993, Lovaas 1987), one matched-pairs design follow-up study comparing an intensive treatment group and a no treatment group (McEachin et al. 1993), one RCT comparing an IBI group to parent training (Smith 2000), one cohort study comparing IBI to a no treatment group (Sheinkopf and Seigel) and one outcome survey consisting of retrospective case reviews and parent questionnaires (Boyd and Cowley 2001)⁴.</p> <p>In all of the studies reviewed, positive gains in IQ (as measured by standardised instruments) and class placement were reported for the majority of children who received IBI. The extent of these gains varied considerably between subjects, within each study and across studies. Findings were inconsistent for changes in social and adaptive functioning. Studies looking at IBI in community or home setting have found fewer gains than those in university centre. Children receiving less intensive treatment (<25 hours) compared to Lovaas (40 hours) attained less significant results. Children receiving IBI from less experienced instructors made smaller gains.</p> <p>Author's conclusions Research indicates some positive gains in IQ, class placement and adaptive behaviour for most children receiving IBI. The authors of this review suggest it is difficult to conclude strongly that IBI is effective for all children with autism based on limitations in the evidence because:</p> <ul style="list-style-type: none"> ▪ not all of the children included in the studies had a diagnosis of autism ▪ diagnostic criteria for autism has changed since the older studies ▪ measures varied greatly between studies ▪ outcomes varied greatly between studies ▪ there was a lack of comparison of IBI to other treatment approaches similar in intensity and duration ▪ only one study included long-term follow-up. 	<p>Strengths</p> <ul style="list-style-type: none"> ▪ used a systematic approach to searching, selecting and appraising of articles. <p>Limitations</p> <ul style="list-style-type: none"> ▪ inclusion criteria was extended (post selection) to studies involving children with a diagnosis of autism or PDD, as per DSM-IV which includes ASD within the category of PDD ▪ only one study (McEachin et al. 1993) looked at long-term gain, however this study lacked random allocation, used potentially biased selection criteria, used different measures at post-test and follow-up and an outcome measure with no established psychometric properties. Therefore results from this study need to be interpreted with caution. <p>Reviewer's comment Brief report but succinctly highlights a number of the limitations associated with studies looking at the effectiveness of IBI in young children with autism.</p>

⁴ The study by Boyd and Cowley (2001) would be graded level IV which differs from the other comparative studies included in this systematic review.

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
<p>Ludwig and Harstall (2001)</p> <p>Alberta, Canada</p> <p>Health Technology Unit of Alberta Heritage Foundation for Medical Research (AHFMR)</p> <p>Level of evidence: III</p>	<p>Systematic review (of secondary reviews)</p> <p>Aim To review the published research on the effectiveness of intensive behavioural intervention (IBI) programs for children with an autistic spectrum disorder (ASD). IBI refers to behavioural interventions that are intensive and comprehensive.</p> <p>Search method Electronic databases searched from 1985 to 2000, where applicable included Medline, EMBASE, Best Evidence, HTA, EED, DARE, Cochrane, ISTAHC, HealthSTAR, PsychInfo, CINAHL, ERIC, Dissertation Abstracts, CMA practice guideline, US National guideline clearinghouse. No limits were placed on publication type or age.</p> <p>The following HTA databases were searched for reports/assessments CCOHTA, BCOHTA, Cochrane Database of Systematic Reviews, NHS Centre for Reviews and Dissemination, INAHTA, ISTAHC, NCCHTA, US Congress, NZHTA, AHRQ, HSURC, MCHPE, ICES, NIMH (refer to Appendix 5 for further details).</p> <p>Search terms included Autism, education, treatment, intervention, therapy, service, Lovaas, TEACCH, Denver, Leap, Rutgers, preschool program.</p> <p>Participants Young children with ASD.</p>	<p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ critical reviews of IBI for young children with autism published between 1985 and 2000, where 'critical' refers to reviews that appraised the scientific validity of the primary research studies according to a set of criteria. <p>Exclusion criteria</p> <ul style="list-style-type: none"> ▪ non-English reviews ▪ reviews where the focus was on mental retardation rather than pervasive developmental disorder. <p>Definitions It can be defined as "early intervention that is carried out all or most of the child's waking hours. Addressing all significant behaviours in all of the child's environments by all significant persons for many years".</p> <p>Outcome measures Achenbach Child Behavior Checklist (ACBC), Autism Treatment of Evaluation Checklist, Bayley Scales of Infant Development, Columbia Mental Maturity Scale (CMMS-III), Leiter International Performance Scale (LIPS), Merrill Palmer Scale of Mental Tests (MPSMT), Parenting Stress Index, Reynell Developmental Language Scales.</p>	<p>The abstracts were reviewed and only critical reviews were chosen. Three critical reviews of intensive programs were found by the literature. These were conducted by:</p> <p>ECRI (1999) Reviewed comprehensive treatment programs including Lovaas therapy, the Rutgers Autism Program, the TEACCH Program, the Denver Model, LEAP Program, and the Autism Pre-school Program.</p> <p>BCOHTA (2000), included in the present evidence tables Reviewed Lovaas therapy for children with autism.</p> <p>Smith (1999) Reviewed nine studies on behavioural therapy, one on the TEACCH program and two on the Denver Model.</p> <p>No additional critical reviews were discovered. From the critical reviews, the original study articles were identified but not all were retrieved before the AHFMR report deadline.</p> <p>Author's conclusions Three critical reviews evaluated a number of comprehensive treatment programs for young children with ASD. These included programs ranging from strict operant learning (Lovaas therapy) to broader applied behavioural analysis (Rutgers) to more developmentally oriented programs (Denver model and TEACCH).</p> <p>Most of the intensive interventions were shown to be effective in producing developmental gains, increases in IQ and less restrictive school placement. Because of methodological limitations and weaknesses of existing research evidence remains limited on the efficacy and effectiveness of one intervention in comparison to another. It does appear that children improve in functioning (as measured by a variety of measures) with behavioural programs. However, it remains to be determined if any one program is more effective than another program. Generally, researchers used standardised, reliable and valid measures of intellectual functioning, adaptive behavior, language performance and development, and various measures of personality, autism rating scales and stress measures. The most common measure used in studies was that of IQ. Further well-designed research studies using multiple independent measures are required. These should be used for all children enrolled in the study, administered by the same assessors blinded to the intervention, and then used again for all children at the end of the study. Optimal intensity and duration of treatment for children with ASD is yet to be determined.</p>	<p>Strengths</p> <ul style="list-style-type: none"> ▪ the appendix of this report provides a summary table of diagnostic and assessment appropriate for use with children/research. <p>Limitations</p> <ul style="list-style-type: none"> ▪ based on the quality of studies reviewed by ECRI, the question "is some improvement occurring?," but not the question, "can any or all of this improvement be attributed to Lovaas treatment?" ▪ this review did not critically appraise any new primary studies and reviewers did not sight all of the primary articles commented on by reviews included in this report ▪ to truly demonstrate efficacy, outcome measures should demonstrate improvements in a range of areas, including social interaction, communication, and overall functioning rather than rely on IQ. <p>Reviewer's comment This review was not a critical appraisal of primary studies but a systematic review of the literature, which summarises recent reviews regarding IBI.</p>

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
<p>McGahan (2001)</p> <p>Ontario, Canada</p> <p>Canadian Coordinating Office for Health Technology Assessment, Ottawa (CCOHTA)</p> <p>Level of evidence: III</p>	<p>Systematic Review (of secondary reviews).</p> <p>Aim To systematically review the literature which summarises recent reviews and expert opinions regarding behavioural therapy for preschool children with autism.</p> <p>Search method Electronic databases searched from January 1995 and November 2000 included Medline, Embase, HealthStar, ERIC, Psycinfo. CINAHL was searched (no date limits), the Cochrane Library (up to issue 2, 2001). HTA websites, Google and other specialised sites were also searched.</p> <p>Some handsearching of selected journals and CCOHTA library and citations was conducted.</p> <p>Search terms included Autism, autistic, asperger, kanner, behaviour therapy, behavioural modification, applied behaviour analysis (ABA), intensive behavioural intervention (IBI), early intervention, psychotherapy, sensory or auditory integration, social integration, social skills training, infants, children, preschool children, toddlers.</p> <p>Participants Studies on preschool children with autism or PDD.</p>	<p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ inclusion of subjects with an identifiable diagnosis of autism or a related PDD, or the presence of "autistic-like-disorder" ▪ any intervention accepted as a valid behavioural procedure by professionals in the field of behaviour modification or applied behaviour analysis , and publication in a refereed professional journal, professionally reviewed book, or a paper presented at a conference. <p>Exclusion Criteria</p> <ul style="list-style-type: none"> ▪ none specifically stated. <p>However, it was noted that studies with methodological flaws were not excluded and retrieval was limited primarily to secondary reviews.</p>	<p>One reviewer evaluated the search results. Irrelevant studies were discarded based on the title of article and information in the abstracts.</p> <p>Recommendations by working groups/guideline panels</p> <ul style="list-style-type: none"> ▪ The New York (New York State Department of Health, Early Intervention Program) group reviewed four studies of efficacy on IBI. In all four studies groups that received IBI showed consistent significant functional improvements in comparison with control groups. They also looked at TEACCH and describe findings as consistent with results indicating that parental involvement in the child's education improves generalisation skills. ▪ The California (the Collaborative Work Group on Autistic Spectrum Disorder) group reviewed five studies and recommended the use of ABA to assist a child in gaining skills and reducing negative or undesirable behaviours. Parent training and ongoing training of personnel was also advised. ▪ The Maine (Maine Administrators of Services for Children with Disabilities Autism Task Force) group reviewed information on eight interventions. They concluded there are peer-reviewed studies supporting the efficacy of ABA methods to improve socially significant behaviours in individuals with autism. <p>Evaluations of efficacy Five previously conducted assessments or critical reviews involving behavioural interventions for children with autism were identified. A brief synopsis of each agencies/authors findings is presented below.</p> <p>Authors's conclusions</p> <ul style="list-style-type: none"> ▪ L'Agence Nationale d'Accreditation d'Evaluation en Sante (ANAES): authors of this review that looked at four studies of behavioural therapies suggested that symptoms associated with autism are ameliorated by intensive behavioural intervention (IBI). 	<p>Strengths</p> <ul style="list-style-type: none"> ▪ specific selection criteria ▪ comprehensive search strategy that involved database searching and handsearching. <p>Limitations</p> <ul style="list-style-type: none"> ▪ only one reviewer ▪ studies examined for the New York guideline studies did not employ random assignment though guideline authors state that there did not appear to be any evidence of bias in assignment, and subject characteristics were equivalent based on baseline data collected by study authors ▪ TEACCH did not use random assignment to groups or assign independent assessors for intervention group ▪ BCOHTA excluded the Sheinkopf and Seigel (1998) retrospective study due to poor design ▪ the Lovaas (1987) study was not randomised so positive outcome in functioning may be due to selection bias ▪ IQ is commonly used as an outcome measure for children with autism, however this may be inadvisable given that this group do not exhibit consistent patterns of performance on intelligence tests.

Table 2. Evidence table of secondary research appraised relating to behavioural and skill-based early intervention for young children with autism (continued)

Authors Country	Aim and search method	Criteria for inclusion and exclusion	Results and authors conclusions	Limitations and comments
McGahan (2001) Ontario, Canada Canadian Coordinating Office for Health Technology Assessment, Ottawa (CCOHTA) Level of evidence: III (Continued)			<ul style="list-style-type: none"> <li data-bbox="1066 344 1733 687">▪ Emergency Care Research Institute (ECRI): this report reviewed comprehensive treatment approaches including Lovaas, Rutgers Autism Program, LEAP, and the Autism Preschool Program (APP). Seven studies were identified, four of which were on Lovaas therapy. ECRI stated the available evidence on Lovaas therapy was only suggestive of treatment effectiveness. The results from the Rutgers study were not evaluated as they did not use a control group or consider confounding factors. Evaluation of the Autism Preschool Program was based on one randomised controlled trial. ECRI noted that only language development in the treatment group showed significant improvement compared to the control group. According to ECRI, while results on TEACCH were statistically significant regarding rates of improvement in treated and untreated children, the differences may not be of clinical significance. ECRI excluded study on PCDI due to unknown number of drop-outs and potential for bias. <li data-bbox="1066 711 1733 847">▪ British Columbia Office of Health Technology Assessment (BCOHTA): authors of this review identified four studies of IBI (same as ECRI). They concluded that while many forms of behaviour therapy clearly benefit children with autism, there is insufficiently scientifically valid evidence to establish a causal relationship between a particular regimen of IBI and the achievement of normal functioning. <li data-bbox="1066 871 1733 983">▪ Smith (1999): The author examined 12 outcome studies including nine on behavioural analytic programmes. The review concluded that the most favourable outcomes occurred in programmes that provide the most intensive services; otherwise, no clear correlations were evident among treatment intensity, treatment model and outcome. <li data-bbox="1066 1007 1733 1184">▪ Alberta Heritage Foundation for Medical Research (AHFMR): An information paper on IBI programs for children with ASD. After summarising the analyses of ECRI, BCOHTA and Smith they state that due to methodological limitations and weaknesses of existing primary research, evidence regarding efficacy and effectiveness of any single treatment program is not available. While children who receive behavioural intervention exhibit functional improvement, it is not clear that any single program is more effective than another. 	Reviewer's comment This review was not a critical appraisal of primary studies but a systematic review of the literature, which summarises recent reviews and expert opinions regarding behavioural therapy. It highlights the lack of methodologically sound research on effectiveness and few conclusions can be made.

Table 3. Evidence table of a primary research study relating to behavioural and skill-based early intervention for young children with ASD

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Bibby et al. (2002) London, UK Autism and Developmental Disorders Education Research Level of evidence: III.2	Study design Cohort Intervention Parent- managed intensive behavioral intervention (IBI)	Study setting Community. Participants Parents of 75 out of 92 children agreed to participate. Six children dropped out or were excluded by parental request (n=4), one ceased treatment before assessments were conducted, one was excluded as an assessment could not be arranged, two had comorbid cerebral palsy and one girl was diagnosed with Rett's disorder. The final sample included 66 children served by 25 different early intervention consultants. All but two, who had a diagnosis of PDD had received independent diagnosis of autism or autistic spectrum disorder (ASD). Sex: 83.3% male Mean age: 45.0 months. Mean intervention time: 31.6 months. Inclusion criteria ▪ diagnosis of autism or ASD. Exclusion criteria ▪ Rett's Disorder.	This study is described as a variant of an accelerated multicohort longitudinal design ⁵ . It is longitudinal over 12 months for individual children and cross-sectional with respect to time into treatment as children were first assessed at different times after the start of treatment. The purposes of this study were to assess whether intervention reproduced Lovaas's clinic based results with respect to outcome; to measure progress of children receiving intervention and to determine variables that may predict children's progress and outcomes. Parent-managed IBI group Parent initiated approximation to Lovaas therapy. A child's programme commenced with workshop provided by consultant. A mixture of theory, modelling and practice was provided to the family and novice therapists. The consultant was hired for further 1-day follow-up workshops. A child's progress and curriculum was reviewed at these follow- ups, as was therapist performance.	Results for IQ and Vineland scales are reported for early data (i.e., score from assessment closest to start of behavioral intervention) and on all measures at T1 (i.e., time of first assessment for this study) and at T2 (i.e., time of second assessment 12 months after T1). All statistical tests were one-tailed and significant at p<0.01, unless otherwise stated. The group mean IQ did not change across 31.6 months of intervention (n=22). Vineland adaptive behavior scores had increased significantly by 8.9 points (n=21). No children aged >72 months attained normal functioning, i.e., IQ>85 and unassisted mainstream school placement (n=42). Progress for 60 children across 12 months was found for mental age (5.4 months), adaptive behavior (9.7 months), and language (5.1 months). At follow-up Mean IQ=53.1 (n=42, SD=20), 95% CI 46.8, 59.4 For all measures at T2-T1 on the Vineland (months; standard scores) ▪ Composite: 9.7, p<0.01 ; 2.2 ns ▪ Communication:12.3, p<0.01 ; 3.5, p<0.05 ▪ Daily living: 7.6, p<0.01; -2.1, ns ▪ Socialisation: 9.8, p<0.01 ; 3.1, ns ▪ Maladaptive (raw score only): -1.1, ns Merrill-Palmer (mental age, months): 6.8, p<0.01 Reynell (months) ▪ Comprehension: 5.7, p<0.1 ▪ Expressive: 4.6, p<0.01	Limitations ▪ the extent to which parents directed some important aspects (eg. teaching methods and curriculum) was not consistent or obvious ▪ treatment was not provided directly as part of the study ▪ the majority of children (81%) in the study received a variety of supplementary treatments whilst in the study, the extent to which these additional "interventions" may confound results is unknown ▪ the study was terminated one- year earlier than planned for ethical reasons ▪ five of the children with IQ>85 at follow-up were already in the normal intellectual functioning range before the start of the behavior intervention by some measure ▪ variations in treatment may account for much of the difference in outcome between parent-managed and centre- based studies ▪ failure to replicate Lovaas's results could be due to variations in the group of children, their treatment or a combination of these factors ▪ no direct measures of the quality of treatment.

⁵ An accelerated, multicohort longitudinal design is a prospective, longitudinal study in which multiple groups (two or more cohorts) are studied. The key feature of the design is the inclusion of cohorts who vary in age when they enter the study. The study is referred to as accelerated because the period of interest is studied in a way that requires less time than if a single group was followed over time.

Table 3. Evidence table of a primary research study relating to behavioural and skill-based early intervention for young children with ASD (continued)

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Bibby et al. (2002) London, UK Autism and Developmental Disorders Education Research Level of evidence: III.2 (Continued)			Outcome measures Assessments were composed of standardised tests of intelligence, visual- spatial skills, language and adaptive functioning. Specific scales utilised included WPPSI-R, WISC-R, ADI-R, the Bayley Scales of Infant Development- Revised, the Merrill-Palmer Scale of Mental Tests, the Reynell Developmental Language Scales and the Vineland Adaptive Behavior Scales. Follow-up interval 12 months.	Outcome was examined for 42 children who were at least 72 months of age and had received 2 years or more of intervention. None of these children fulfilled Lovaas's criteria for best outcome. Of 66 children, 10 (17%) were in the range of normal at T2, and three (5%) also achieved educational normal functioning.	Author's conclusions The parent-managed intensive behavioural interventions did not reproduce results from clinic-based professionally directed programmes. Reviewer's comments This study did not find any evidence to support the effectiveness of the intervention.

Table 3. Evidence table of a primary research study relating to behavioural and skill-based early intervention for young children with ASD (continued)

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Drew et al. (2002) London, UK Scottish Centre for Autism Level of evidence: II	Study design Randomised controlled trial Intervention Parent training group Comparison Local services only	Study setting This study was a pilot randomised controlled trial of parent training intervention for pre-school children with autism. It took place in an outpatient hospital setting in conjunction with local services group. Participants Forty-six were seen for a full clinical assessment. Of these 31 had childhood autism, five atypical autism or PDD-NOS, six a receptive-expressive language disorder, two global developmental delay and one attention deficit hyperactivity disorder. Only one child was clinically normal. A total of 31 eligible children were identified via screening. n=24 agreed to participate (77% parents) Sex: 80% male Mean age: 23 months. n=12 Parent training group (11 boys, 1 girl) n=12 Local services group (8 boys, 4 girls). Inclusion criteria ICD-10 diagnosis of childhood autism (F84.0). Exclusion criteria None stated.	Groups were matched for age at the pre-intervention assessment. Allocation to intervention was via random number table. Parent training programme Parents were given advice about behavioural management and promoting compliance. This included principles of reinforcement, interrupting unwanted behaviours, teaching alternative behaviours (using joint action routines). For example, teaching joint attention behaviours such as index finger pointing, gaze switching, and the use of visual supports for spoken language. Local services group These children received a mixture of services including speech and language therapy sessions, occupational therapy, physiotherapy, home worker input and Lovaas/Applied Behavioral Analysis (ABA) therapy. Outcome measures Non-verbal IQ from subscales of Griffiths Scale of Mental Development (NVIQ), MacArthur Communicative Development Inventory (CDI), Autism Diagnostic Interview-Revised (ADI-R) domains including reciprocal social interaction (RSI), nonverbal communication (NVC), repetitive and stereotyped behaviour (RSB), Parent Stress Inventory (PSI). Follow-up interval 12 months.	From activity checklists, the groups did not differ on number of hours per week in playgroup/nursery, hours per month in speech and language therapy, time parents spent in one-to-one structured activities. Local services group received more other intervention (n=3 ABA), $F[1,23]=3.6$, $p=0.07$. ANOVA was used to examine group differences at baseline. At baseline The groups did not differ in language ability (number of words understood, number of words used, number of gestures produced) as measured by the CDI. The parent training group did have a higher NVIQ $F[1,23]=14.8$, $p<0.001$. 11 of 12 children in both groups were nonverbal (< 5 words). No differences in symptom severity (as per RSI, NVC and RSB) or parental stress. ANCOVA was used to examine group differences at follow-up, initial scores on each variable were entered as covariates. At follow-up No differences in age or NVIQ A trend towards a difference was observed on CDI $F[1,16]=3.1$, $p=0.09$ No group differences on initial words or gestures produced but there were significant covariate effects for these and parental stress (i.e., time 1 scores were associated with time 2 scores). More children in the parent training group moved from being nonverbal to having single word/phrase speech (n=7) than local services group (n=2 plus one child went from single words to less than five), fisher exact test, $p<0.05$). There were no differences in symptom severity or parent self-report of stress. Author's conclusions A modest improvement in communication skills was observed for the parent training group compared with usual care.	Limitations <ul style="list-style-type: none">▪ two groups not matched at initial assessment in terms of NVIQ▪ parents of three children allocated to Local services group opted to start home-based behavioural programmes (although this sort of choice reflected the real-life clinical situation)▪ not clear who administered the random allocation procedure▪ it is impossible to maintain blindness to group intervention in this type of trial for either parents of participants or assessors▪ most of the measures relied on parental report▪ small sample size/low statistical power▪ little or no data on implementation of the parent training intervention. Reviewer's comments Provides some evidence of the positive effect of parent training on specific areas however it is not possible to rule out that the language gains of the parent training group were not due to differences in initial characteristics.

Table 3. Evidence table of a primary research study relating to behavioural and skill-based early intervention for young children with ASD (continued)

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Eikeseth et al. (2002) Akershus, Norway Level of evidence: III.2	Study design Comparative study with concurrent controls but no random allocation Intervention Intensive behavioral treatment (IBT) Comparison Eclectic treatment (usual care)	Study setting This study was a comparison- controlled study of intensive behavioral treatment at school for four to seven year old children with autism. Both treatments took place in public kindergartens and elementary schools for typically developing children. Participants A total of 25 children participated, plus there were 2 further drop-outs. n=13 IBT group Sex: 62% male Mean treatment: 28.0 hours n=12 Eclectic group Sex: 92% male Mean treatment: 29.08 hours Inclusion criteria <ul style="list-style-type: none"> ICD-10 diagnosis of childhood autism (F84.0) from Autism Diagnostic Interview-Revised and an independent child clinical psychologist chronological age (CA) between 4 and 7 years at the time of intake deviation IQ of 50 or above on the Weschler Preschool and Primary Scale of Intelligence- Revised or ratio IQ of 50 or above on the Bayley Scales of Infant Development absence of major medical conditions other than autism. Exclusion criteria <ul style="list-style-type: none"> children with IQs under 50. 	All children were assessed at intake and 1 year after treatment began. At follow-up, therapists were asked to report the number of hours per week of one-to-one treatment, the goals in each child's individualised education plan (including vocal language, alternative/augmentative communication, academics, play, social skills, imitation, motor skills, daily living skills, and behavior management. IBT group Behavioral treatment was based on Lovaas manual and videotapes with some modification. IBT began with simple tasks, such as responding to basic requests made by an adult. If then progressed to more complex tasks, such as imitating verbal and non-verbal behaviors, labelling objects, identifying actions and understanding abstract concepts. Advanced social skills such as answering questions, conversing and making friends were also addressed. Eclectic treatment group This treatment incorporated elements from a variety of interventions, such as project TEACCH, sensory-motor therapies, applied behavioural analysis and methods derived from personal experience. The combination of interventions was individually selected for the child based on recommendations from a multidisciplinary team of school personnel. Outcome measures Assessments were composed of standardised tests of intelligence, visual-spatial skills, language and adaptive functioning. Specific scales utilised included WPPSI-R, WISC-R, the Bayley Scales of Infant Development-Revised, the Merrill-Palmer Scale of Mental Tests, the Reynell Developmental Language Scales and the Vineland Adaptive Behavior Scales. Follow-up interval 12 months.	Mean change at follow-up, p value given where significant difference between treatment groups. IBT group CA (months) =12.19 IQ=17.15, p<0.01 Performance IQ=17.46 Language Comprehension=12.70, p<0.05 Expressive=22.57, p<0.05 Total=27.00, p<0.05 Vineland Scales Communication=15.69, p<0.01 Daily Living=9.23 Socialisation=10.00 Composite=11.23, p<0.05 Eclectic treatment group CA (months) =13.58 IQ=4.33 Performance IQ=8.33 Language Comprehension=-0.70 Expressive=-2.23 Total=1.08 Vineland Scales Communication=-1.58 Daily Living=5.50 Socialisation=8.50 Composite=0.17 Author's conclusions At a 1-year evaluation, children who received IBT made significantly larger improvements on measures of IQ, language expression and comprehension and communication. Results also suggest that IBT can be successfully adapted to school settings for some children with autism.	Strengths <ul style="list-style-type: none"> comprehensive, uniform assessment protocols administered by blind examiners manualised, research-based interventions for the behavioral treatment group treatment supervision by experienced personnel measures of the amount of treatment children received, skills addressed in treatment, and education of therapists group assignment by professional independent of study. Limitations <ul style="list-style-type: none"> potential for bias in non- random assignment to groups although authors assert that the only factor influencing group assignment was staff availability no intent to treat analysis small sample size measures that focussed more on cognitive than social development a lower treatment intensity than has been recommended by the developers of the behavioral treatment model was used in the study. Reviewer's comment This study provides some weak evidence that 4 to 7 year old children with autism may make specific gains with intensive behavioral intervention.

Table 3. Evidence table of a primary research study relating to behavioural and skill based early intervention for young children with ASD (continued)

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Salt et al. (2002) Glasgow, Scotland. Level of evidence: III.2.	Study design Comparative study with concurrent controls but no random allocation Intervention Scottish Centre for Autism (SCA) Early intervention programme Comparison Waiting list	Study setting Developmentally based early intervention programme. Participants Twenty children and their families. SCA programme n=14 Mean age: 42.4 months Mental age: 17 months Sex: 92% male IQ: 39.4 Pre-post interval: 10.0 months. Waiting list n=5. Mean age: 37.7 months Mental age: 20.8 months Sex: 60% male IQ: 55.7 Pre-post interval: 10.3 months. Inclusion criteria. ICD-10 diagnosis of childhood autism (F84.0). Exclusion criteria. None stated. Three participants dropped out. Two twin boys discontinued after being taken into social work care. One control family moved out of the country,	This study evaluates a Scottish early intervention programme with a social-developmental approach. A two group (treatment, control) pre- test post-test design was used. Treatments received across the assessment period: SCA programme group n=12 ▪ SCA therapy (8 hours) ▪ non-SCA intervention (total of 30.4 hours) including nursery (27.9 hours) and other 1:1 therapy (2.8 hours). Waiting list group n=5 ▪ non-SCA intervention (total of 40.9 hours) including nursery (38.7 hours) and other 1:1 therapy (2.2 hours). The treatment group received the SCA therapy but the controls did not. Group performance was compared using a range of assessment measures listed below. Outcome measures Bayley Scales of Infant Development (BSID) 2 nd ed., British Picture Vocabulary Scale (BPVS), Vineland Adaptive Behaviour Scale (VABS), Pre-Verbal Communication Schedule (PVCS), MacArthur Communication Development Inventories (MCDI), Symbolic Play Test 2 nd ed, Early Social Communication Scales (ESCS) and the Parenting Stress Index 3 rd ed (PSI, short form). Follow-up interval 10 months.	Pre-treatment Experimental and control group were comparable for age, mental age, socioeconomic background and number or hours of non-experimental therapy. The control group has a significantly higher pre-treatment IQ, $t(15)=-2.78$, $p=0.01$. Parametric statistical procedures were used. Interaction effects between time and group were examined. The test statistics and p-values are only presented for group by time interaction effects only. Treatment pre-test/post-test mean (SD) MCDI words understood: 62.6 (44.7)/91.2 (68.5) MCDI words produced: 4.7 (7.5)/17.8 (40.9) PVCS imitation: 1.9 (2.2)/7.7 (5.4) Symbolic Play Test: 1.9 (2.2)/7.7 (5.4) ESCS joint attention: 5.44 (3.17)/9.11 (5.58) ESCS requesting behaviour: 3.67 (2.12)/10.33 (6.96) ESCS social Interaction: 3.33 (2.00)/5.22 (2.73) Control n=5 pre/post mean (SD) MCDI words understood: 55.6 (42.75)/79.8 (50.16) MCDI words produced: 11.6 (15.08)/31.20 (22.61) PVCS imitation: 1.6 (0.55)/3.8 (0.84) Symbolic Play Test: 12.8 (4.82)/19.6 (3.78) ESCS joint attention: 6.00 (3.39)/4.6 (2.97) ESCS requesting behaviour: 6.6 (5.59)/6.2 (2.77) ESCS social Interaction : 2.86 (3.2)/0.84 (5.66) Treatment versus control MCDI words understood: $F[1, 15]$, ns MCDI words produced: $F[1, 15]$, ns PVCS imitation: $F[1, 15]=4.6$, $p<0.05$ Symbolic Play Test: $F[1, 15]=0.86$, ns ESCS joint attention: $F[1, 12]=5.07$, $p<0.05$ ESCS requesting behaviour: $F[1, 12]=4.45$, $p<0.06$ ESCS social Interaction : $F[1, 12]=5.66$, $p<0.05$ Group differences, including group by time effects for scores on Parenting Stress Index were non significant on all subscales. Author's conclusions Children in the treatment group improved significantly more than those in the control group on measures of joint attention, social interaction, imitation, daily living skills, motor skills and an adaptive behaviour composite. Results support the efficacy of this approach.	Strengths ▪ use of 'naturalistic' study design as opposed to discrete trial format of intervention ▪ early intervention treatment that uses social- developmental format ▪ adds to the debate about treatment intensity at lower intensity than typical ABA programmes. Limitations ▪ selection/recruitment methods not clear "usual clinical manner" ▪ non-parametric distribution free methods (for small samples) were not adopted although reasons for this are justified by the authors, including that they could not account for group and maturation effects over time ▪ small sample size, particularly in the control arm ▪ children were not randomly assigned to groups, leading to potential bias in the control group that had a significantly higher mean IQ at treatment onset ▪ the study was designed with a waiting list control group which focused on immediate outcome of treatment and could not address the issue of long-term follow-up. Reviewer's comment Provides weak evidence of gains in domains known to enhance social and communicative development.

Table 3. Evidence table of a primary research study relating to behavioural and skill based early intervention for young children with ASD (continued)

Authors Country	Study design	Sample and interventions	Methods	Results	Limitations and conclusions
Smith et al. (2000) The UCLA Young Autism Project Washington State, USA. Level of evidence: II	Randomised trial comparing two treatments. Intervention Intensive treatment Comparison Parent training	<p>Study setting University.</p> <p>Participants Twenty-eight children participated in the study including 14 with autism and 14 with PDD-NOS. There were no drop-outs among this group of children however one child's family declined participation at intake, and 8 other children were excluded (4 because they did not have diagnosis of autism or PDD-NOS; 2 scored below IQ cut-off; and 2 because they were in foster care).</p> <p>Inclusion criteria</p> <ul style="list-style-type: none"> ▪ chronological age (CA) between 18 and 42 months at the time of referral ▪ residence within a one-hour drive of the research/treatment site ▪ IQ ratio between 35 and 75 ▪ diagnosis of autism or pervasive developmental disorder NOS. <p>Exclusion criteria</p> <ul style="list-style-type: none"> ▪ major medical problems other than autism or mental retardation (e.g., cerebral palsy, blindness or deafness, known genetic disorders such as Down syndrome or neurological conditions such as uncontrolled seizure disorders) ▪ without a permanent residence able to provide home-based services offered to children in this study. 	<p>Children were assigned to intensive treatment or parent training based on a matched-pair, random assignment procedure. An independent statistician paired children in each cohort (autism or PDD-NOS) on the basis of IQ. A random numbers table was used to assign one member of each pair to each intervention. The unequal size of groups resulted from a design change outlined in limitations.</p> <p>Intensive treatment group</p> <ul style="list-style-type: none"> ▪ the aim was to maximise a child's intellectual, adaptive and socio-emotional functioning and, thereby, reduce their need for special education services ▪ for this study it was defined as 30 hours per week of intervention for each child for 2 to 3 years ▪ children received treatment from teams of 4 to 6 student therapists working under supervision ▪ each child's primary caregiver was also asked to conduct 5 hours per week of treatment working alongside a student therapist for the first three months of treatment. <p>Parent training group</p> <ul style="list-style-type: none"> ▪ the aim was to teach parents to use treatment approaches described by the Lovaas manual and assist them in using these approaches to help their children acquire skills ▪ the child's families received two sessions per week of parent training, totalling 5 hours per week, in their homes for 3 to 9 months. <p>Outcome measures</p> <p>Standardised instruments to assess skills in both typically and atypically developing children. These included Stanford-Binet Intelligence Scale, Bayley Scales of Infant Development Index, Merrill-Palmer Scale of Mental Tests, Reynell Developmental Language Scales (DLS), Vineland Adaptive Behavior Scales, Achenbach Child Behaviour Checklist and Teacher Report Form, Wechsler Individualized Achievement Test, Early Learning Measure and Family Satisfaction Questionnaire.</p> <p>Follow-up interval</p> <p>Subjects were reassessed at a CA of 7-8 years.</p>	<p>To test for between group differences two-tailed t-tests were performed. Bonferroni corrections were applied to each set of tests. Means and SDs of children's standardised test scores are presented only for the combined group and at follow-up. Significant differences at follow-up are indicated.</p> <p>Intensive treatment mean(SD) IQ=66.49 (24.08) Merrill-Palmer= 64.33 (18.74) Reynell Language scales</p> <ul style="list-style-type: none"> ▪ comprehension=42.87 (22.29) ▪ expressive=44.53 (23.48) ▪ total=87.40 (46.21) <p>Vineland</p> <ul style="list-style-type: none"> ▪ communication=67.87 (30.08) ▪ language=62.33 (25.76) ▪ socialisation=66.33 (24.78) ▪ composite=61.19 (29.72) <p>Parent training mean(SD) IQ=49.67 (19.74), p<0.05 Merrill-Palmer=49.17 (21.43), p<0.05 Reynell Language scales</p> <ul style="list-style-type: none"> ▪ comprehension=33.00 (16.86) ▪ expressive=36.23 (21.19) ▪ total=61.33 (31.88), p<0.05 <p>Vineland</p> <ul style="list-style-type: none"> ▪ communication=60.77 (17.26) ▪ language=63.00 (16.97) ▪ socialisation=68.92 (16.94) ▪ composite=58.50 (16.58) <p>Author's conclusions</p> <p>The groups appeared similar at intake on all measures; however, at follow-up the intensive treatment group outperformed the parent training group on measures of intelligence, visual-spatial skills, language, and academics, though not adaptive functioning or behavior problems. Children with PDD-NOS may have made more gains than those with autism.</p>	<p>Strengths</p> <ul style="list-style-type: none"> ▪ manualised treatments based on research and supervised by experienced personnel ▪ blind examiners ▪ long-term follow-up ▪ use of an experimental design ▪ comprehensive battery of assessment tools including standardised measures. <p>Limitations</p> <ul style="list-style-type: none"> ▪ small overall sample size means estimates lack precision and confidence intervals not presented although enough information is given so that these can be calculated (see Diggle et al. 2003) ▪ in general, low power to detect predictors of treatment response ▪ the authors intention was to also study children who had mental retardation but without PDD however sample size was too small (n=3) to draw any reliable inferences so data on these subjects was removed from the study, post completion (1 from intensive treatment group and 3 from parent training) ▪ no standardised diagnostic tool used ▪ only one measure of social skills. <p>Reviewer's comment</p> <p>Some evidence that intensive treatment may be more effective than parent training but multisite studies are still required to overcome key methodological limitations.</p>

OVERVIEW

Overall quality of evidence

Of the 10 articles appraised, five were secondary studies (see **Table 2, pages 7-13**) and five were primary studies (see **Table 3, pages 14-19**). The latter included one cohort study and four controlled studies. Of the controlled studies two were randomised controlled trials (Level II) and two were comparative studies with concurrent controls but no random allocation (Level III.2) according to the NHMRC hierarchy of evidence, see **Appendix 1**. The main comparisons and characteristics of types of intervention or treatment programmes considered in the primary studies are briefly summarised in **Table 4**.

Table 4. Design, levels of evidence and interventions for primary studies

Primary study	Design and level of evidence	Characteristics of interventions
Bibby et al. (2002)	Cohort study Level II	Parent managed intensive behavioural intervention <ul style="list-style-type: none"> parent initiated approximations to Lovaas therapy
Drew et al. (2002)	Randomised controlled trial Level II	Parent training <ul style="list-style-type: none"> advice about behavioural management and promoting compliance Local services (usual care) <ul style="list-style-type: none"> a mixture of services including speech and language therapy sessions, occupational therapy, physiotherapy, home worker input and Lovaas/Applied Behavioural Analysis (ABA) therapy
Eikeseth et al. (2002)	Concurrent controls, non randomised Level III.2	Intensive behavioural treatment <ul style="list-style-type: none"> behavioural treatment based on Lovaas manual and videotapes with some modification Eclectic treatment (usual care) <ul style="list-style-type: none"> a combination of elements from a variety of interventions, such as project TEACCH, sensory-motor therapies, applied behavioural analysis and methods derived from personal experience
Salt et al. (2002)	Concurrent controls, non randomised Level III.2	Early intervention programme <ul style="list-style-type: none"> Scottish Centre for Autism (SCA) therapy (8 hours) with a social-developmental approach plus non-SCA intervention (total of 30.4 hours) including nursery (27.9 hours) and other 1:1 therapy (2.8 hours) Waiting list <ul style="list-style-type: none"> non-SCA intervention (total of 40.9 hours) including nursery (38.7 hours) and other 1:1 therapy (2.2 hours)
Smith et al. (2000)	Randomised controlled trial Level II	Intensive behavioural treatment <ul style="list-style-type: none"> 30 hours per week of intervention for each child for 2 to 3 years from teams of 4 to 6 student therapists Parent training <ul style="list-style-type: none"> parent-managed Lovaas therapy to help their children acquire skills where a child's family received two sessions per week of parent training, totalling 5 hours per week, in their homes for 3 to 9 months

The five primary studies were conducted in a variety of settings and included young children with autism receiving care within an outpatient or community service, or enrolled in preschool/primary school or an intervention delivered as part of research project. Three of the studies were based in the United Kingdom, one in the United States of America and one in Norway. The secondary research included reviews which either summarised or critically appraised studies of the highest quality (Bassett et al. 2000; Diggle et al. 2003; Ludwig and Harstall 2001; McGahan 2001), with the exception of one review which included a mix of study designs (Finch and Raffaele 2003).

Summary of evidence for the effectiveness of early behavioural intervention

Secondary studies

The five secondary studies that met our selection criteria are summarised below.

Bassett et al. (2000) concluded that the effectiveness claim of Lovaas (1987) that half the children achieved normal or near-normal development and placement in schools remains uncorroborated by independent research. The outcome evidence does not support adopting a single entire programme, whether Lovaas or alternative methods, based on the likelihood that children will achieve normal function. They suggest that due to major methodological flaws, the findings of Lovaas (1987) and McEachin et al. (1993) cannot be regarded as conclusive; more research is required before authoritative statements can be made about efficacy. In the absence of scientifically valid studies of overall autism programmes, Bassett et al. further suggests “it may be prudent to revise existing autism services using incremental, not programmatic steps”.

The two studies included in the review by Diggle et al. (2003) on parent-mediated intervention were not directly comparable as they compared different interventions. The methodology of the studies also raised more questions than they answered. Although some evidence is presented in favour of the effectiveness of early intervention, the authors suggest the results should be viewed with great caution due to the small number of included studies and inherent differences between the two studies. They both used an intervention based broadly on behavioural principles however the two studies differed in theoretical approach, intensity, duration, context and the type of intervention comparison.

Finch and Raffaele (2003) suggest that research indicates some positive gains in IQ, class placement and adaptive behaviour for most children receiving intensive behavioural intervention (IBI). The authors of this review stated that it is difficult to conclude strongly that IBI is effective for all children with autism. This was a succinct review with relatively broad inclusion criteria but it highlights a number of the limitations associated with studies looking at the effectiveness of IBI in young children with autism.

The report by Ludwig and Harstall (2001) was not a critical appraisal of primary studies but a systematic review of the literature which summarises recent reviews regarding IBI. It included three critical reviews that evaluated a number of comprehensive treatment programmes for young children with autism. These included programmes ranging from strict operant learning (Lovaas therapy) to broader applied behavioural analysis (Rutgers) to more developmentally oriented programmes (Denver model and TEACCH). Most of the intensive interventions were shown to be effective in producing developmental gains, increases in IQ and less restrictive school placement. Ludwig and Harstall conclude that because of methodological limitations and weaknesses of existing research, evidence remains limited on the efficacy and effectiveness of one intervention in comparison to another. It does appear that children improve in functioning (as measured by a variety of measures) with behavioural programmes. However, it remains to be determined if any one programme is more effective than another programme.

McGahan (2001) systematically reviewed the literature which summarises recent reviews and expert opinions regarding behavioural therapy for preschool children with autism. They identified five previously conducted assessments or critical reviews involving behavioural interventions for children with autism. The author reaches the same conclusion as Ludwig and Harstall (2001) and states that due to methodological limitations and weaknesses of existing primary research, evidence regarding efficacy and effectiveness of any single treatment programme is not available. While children who receive behavioural intervention exhibit functional improvement, it is not clear that any single programme is more effective than another.

All five secondary studies appraised draw attention to the lack of well-conducted research on early intervention for autism in young children. All five reviews summarised here reached the same conclusion, that to date there is insufficient evidence to allow conclusions to be drawn about best practice. Furthermore, researchers have yet to establish a relationship between the amount (per day and total duration) of any form of early comprehensive treatment programme and overall outcome (Bassett et al. 2000).

Primary studies

Despite the relatively large volume of studies published (Trevarthen et al. 1998) and extent of interest of a variety of “stakeholders” in the effectiveness of interventions for young children with autism (Jacobson 2000) only five primary studies published since 2000 met selection criteria for relevance and methodological quality. The specific studies are summarised below.

One multi-cohort study was identified and this was the only longitudinal study appraised. Bibby et al. (2002) found that parent-managed intensive behavioural interventions did not reproduce results from clinic-based professionally directed programmes, such as Lovaas therapy.

Two randomised controlled trials were identified. The Smith et al. (2000) study is also included in the reviews by Diggle et al. (2003) and Finch and Raffaele (2003). This trial found that at follow-up the intensive treatment group outperformed the parent training group on measures of intelligence, visual-spatial skills, language, and academics, though not adaptive functioning or behaviour problems. The groups appeared similar at intake on all measures and children with PDD-NOS may have made more gains than those with autism. This study provides some evidence that intensive treatment may be more effective than parent training but multisite studies are still required to overcome key methodological limitations. More recently, Drew et al. (2002) conducted a pilot randomised controlled trial of parent training intervention for pre-school children with autism. A modest improvement in communication skills was observed for the parent training group compared with usual care. It was not possible to rule out that the language gains of the parent training group were not due to differences in initial characteristics.

A further two comparative studies were identified. Eikeseth et al. (2002) found at a one-year evaluation, children who received intensive behavioural therapy made significantly larger improvements on measures of IQ, language expression and comprehension and communication. Results also suggest that intensive behavioural therapy can be successfully adapted to school settings for some children with autism. This study provides some weak evidence that four to seven year old children with autism may make specific gains with intensive behavioural intervention. Salt et al. (2002) evaluated a Scottish early intervention programme that describes itself as having a social-developmental approach. A two group (treatment, concurrent controls) pre-test post-test design was used in this study. Children in the treatment group improved significantly more than those in the control group on measures of joint attention, social interaction, imitation, daily living skills, motor skills and an adaptive behaviour composite. Initial results provide some support for the efficacy of this approach.

Three of the studies concerned parent-managed intervention strategies and the remaining studies describe respectively, a preschool/primary school-based programme and a clinic-based early intervention programme. Preliminary evidence from these studies suggest that early intervention (note this includes different types of behavioural intervention, across different settings) may lead to selected gains in a number of specific domains; however, further research is required to address the methodological limitations of existing studies and replicate their findings. In particular studies with larger sample sizes (from multisite collaborations using identical methods and outcome measures) are required to provide greater statistical power and more precise estimates of effectiveness.

Methodological issues and future direction

A number of common themes emerged in relation to assessing change in early intervention programmes for children with autism (Charman and Howlin 2003; Kasari 2002). There is a need within autism research for more studies that incorporate the following features:

- treatments that follow a specific protocol or manual (replicability of treatment)
- studies that use random assignment to treatment group or matching procedures
- blind examiners administering assessment
- a variety of assessment tools that includes standardised outcome measures and these should demonstrate improvements in a range of areas, including social interaction, communication, and overall functioning

- measures of the amount or intensity of the treatment provided
- further studies that compare one treatment approach to another
- larger sample sizes from multisite collaborations.

Ongoing studies

There are several trials currently in progress or recently completed that appear to be relevant but their results are yet to be published:

- Dr Connie Kasari and colleagues (USA) are conducting a randomised controlled trial looking at the efficacy of teaching caregivers how to initiate and maintain episodes of joint attention with their children (<http://www.controlled-trials.com/mrct/trial/kasari/1059/40841.html>).
- Professor Gregory Stores (UK) is the principal investigator for a randomised trial assessing the efficacy of behavioural treatment for severe sleep disorders in children with autism (<http://www.controlled-trials.com/mrct/trial/gregory%7cautism/1046/26773.html>).
- Dr Tony Charman (UK) is conducting a follow-up evaluation (randomised controlled trial) of a social-communication parent training intervention for children with autism (<http://www.controlled-trials.com/mrct/trial/charman/1060/41983.html> and <http://www.controlled-trials.com/mrct/trial/charman/1046/37277.html>).
- Dr Ole Lovaas (USA) is listed on the clinical trials register as the study chair for a completed NIMH randomised trial of one-to-one behavioural treatment in the home and neighbourhood compared with at home, individualised parent training in preschool children. Eleven sites are also listed on the website for the Lovaas Institute for Early Intervention (<http://www.lovaas.com>) as being National Institute for Mental Health (NIMH) replication sites, six of these are in the USA and additional centres can be found in Iceland, Japan, Norway, Spain and the United Kingdom. It is not clear if this is the same trial or another NIMH funded multisite study.

Future research

In a recently published report on identifying priority areas for future research in the United Kingdom, Charman and Clare (2004) suggest that there is increasing evidence that appropriately targeted intervention improves outcomes in children with autism. This evidence includes a large number of individual case and small case series studies, providing support for behavioural and some psychosocial interventions, targeting language, behaviour, play and social skills. They also note that in contrast there is a relative paucity of well-controlled empirical group evaluations of intervention programmes for children with autism and fewer randomised controlled trials. The strongest evidence is for early, intensive behavioural interventions that have led to gains in IQ and language ability. Preliminary findings from ongoing studies using education-based approaches have also demonstrated positive outcomes in terms of IQ gains and reductions in symptom severity. Intervention approaches that place an emphasis on the development of non-verbal communicative skills have also provided promising data (Charman and Clare 2004; Koegel 2000; Lord 2000; Ozonoff and Cathcart 1998).

Further treatment studies that incorporate all the elements of scientific rigour are required. Comparative studies of behavioural and skill-based early intervention should be designed in a way that will allow some judgement to be made over whether one treatment approach is superior to another. Moreover, current treatment programmes need to evaluate the active ingredients of their treatment. A number of programmes use similar strategies and techniques but they also differ in critical ways – e.g., whether they employ one to one versus group treatment and home-based versus school or clinic based treatment (Kasari 2002). Kasari (2002) also notes that there are a number of autism-specific behaviours that may improve as a result of particular components of a comprehensive treatment package or in direct teaching of these skills deficits.

Limitations of this report

This Tech Brief was limited in scope; it focuses on the “best” evidence from group designs and ignores single case experiments however the utility of “single” case research designs for evaluating the effect of specific interventions is also acknowledged. Studies employing smaller sample sizes (fewer than

required for a randomised trial) with multiple data points may provide important information on key intervention targets (Kasari 2002). Experimental evidence already strongly suggests that focal treatments directed either at reducing specific behavioural problems associated with autism; or at increasing behavioural successes are effective in reducing symptom frequency and severity as well as increasing adaptive skills (Bassett et al. 2000; Matson et al. 1996; Rogers 1996).

Three recent systematic reviews were outside the scope of this Tech Brief as they focused solely on experimental studies that did not include control groups (Campbell 2003; Horner et al. 2002; Hwang and Hughes 2000). Horner et al. (2002) summarised research on behavioural interventions for children with autism eight years of age or younger. Horner et al. (2002) suggest behavioural interventions may reduce problem behaviours by up to 80 to 90 percent. This review did not identify any type of intervention that is uniquely effective with young children with autism. In a quantitative synthesis of single subject research, Campbell (2003) suggests that behavioural treatments are effective but that the type of target behaviour and type of treatment do not moderate the average effect of treatment. Hwang and Hughes (2000) reviewed studies that investigated the effects of social interactive interventions designed to increase early social communicative skills. Although they found increases for social and affective behaviours, nonverbal and verbal communication, eye contact, joint attention and imitative play, limited generalisation or maintenance of target behaviours was reported.

No studies were found that were set in New Zealand but several recently published guidelines may be of interest to families/carers, clinicians, and service funders and providers in New Zealand (Connecticut Birth to Three System 2002; Filipek et al. 2000; Le Couteur 2003; Maine Administrators of Services for Children with Disabilities (MADSEC) 2000; New York State Department of Health 1999).

Conclusions

It does appear that children improve in functioning (as measured by a variety of outcome measures) with behavioural intervention (Campbell 2003; Horner et al. 2002). The majority of recent primary studies reviewed here document some improvement associated with intervention (Drew et al. 2002; Eikeseth et al. 2002; Salt et al. 2002; Smith et al. 2000), however it remains to be determined if any one early and/or intensive intervention programme is more effective than another programme (Bassett et al. 2000; Diggle et al. 2003; Finch and Raffaele 2003; Ludwig and Harstall 2001; McGahan 2001). The primary studies included in this Tech Brief cover a range of interventions and comparisons and it was not clear that the definition of intensive behavioural treatment, parent training or parent-managed behavioural therapy were uniform across individual studies evaluating similar approaches to early intervention. The intensity and duration of the intervention offered was only documented in some of the studies and sample sizes were mostly small. Given these and other limitations, the primary studies appraised generally provide only very preliminary evidence about the effectiveness of behavioural and skill-based early intervention. Nevertheless, two of these studies were graded as Level II according to the NHMRC hierarchy which reflects high level evidence (Drew et al. 2002; Smith et al. 2000).

- Parent-managed intensive behavioural interventions were not found to be as effective as clinic-based professionally directed programmes by Bibby et al. (2002), however a parent training intervention was more effective than usual care for improving communication (Drew et al. 2002).
- Intensive behavioural treatment may have a positive effect on measures of IQ, language expression and comprehension and communication (Eikeseth et al. 2002) and one recent study has suggested intensive behavioural intervention may be more effective than parent training (Smith et al. 2000).
- Early intervention that utilises a social-developmental approach may be an effective treatment model (Salt et al. 2002).

Direct empirical evidence that early compared to later intervention has a specific positive benefit is not yet available, at best there appears to be consensus among experts in the field that developmental principles support the notion of early intervention (Charman and Clare 2004). More research in this area is needed and the conclusions of this Tech Brief should be revisited as the results, particularly of current and ongoing trials become available.

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Volkmar, F., Cook, E. H., Jr., Pomeroy, J., Realmuto, G., & Tanguay, P. (1999). Practice parameters for the assessment and treatment of children, adolescents, and adults with autism and other pervasive developmental disorders. American Academy of Child and Adolescent Psychiatry Working Group on Quality Issues. *Journal of the American Academy of Child & Adolescent Psychiatry*, 38, 32S-54S.

World Health Organization (1992). *The ICD-10 classification of mental and behavioural disorders: clinical descriptions and diagnostic guidelines*. Geneva: World Health Organization.

APPENDIX 1: LEVELS OF EVIDENCE*

- Level I Evidence obtained from a systematic review (or meta-analysis) of relevant randomised controlled trials.
- Level II Evidence obtained from at least one randomised controlled trial.
- Level III. 1 Evidence obtained from pseudo randomised controlled trials (alternate allocation or some other method).
- 2 Evidence obtained from comparative studies (including a systematic reviews of such studies) with concurrent controls and allocation not randomised, cohort studies, case control studies or interrupted time series with a control group).
- 3 Evidence obtained from comparative studies with historical control, two or more single-arm studies or interrupted time series without a parallel control group.
- Level IV Evidence obtained from case series, either post-test or pretest/post-test.

*National Health and Medical Research Council (1999)

APPENDIX 2: SEARCH STRATEGY

Medline

- 1 asperger syndrome/ or autistic disorder/ (6975)
- 2 Rett Syndrome/ (842)
- 3 autis\$.mp. (7830)
- 4 kanner's.tw. (40)
- 5 Child Development Disorders, Pervasive/ (634)
- 6 or/1-5 (8885)
- 7 limit 6 to ((all infant <birth to 23 months> or preschool child <2 to 5 years> or child <6 to 12 years>) and yr=1990-2004) (4095)
- 8 exp Behavior Therapy/ (28421)
- 9 (therapy adj3 condition\$.tw. (3553)
- 10 (behavio?r\$ adj3 modif\$.tw. (5453)
- 11 (behavio?r\$ adj3 therap\$.tw. (6349)
- 12 (behavio?r adj3 intervention\$.tw. (1115)
- 13 Cognitive Therapy/ (4257)
- 14 (cognit\$ adj3 (therap\$ or psychotherap\$).tw. (3269)
- 15 or/8-14 (40085)
- 16 15 and 7 (218)
- 17 limit 16 to english language (207)
- 18 from 17 keep [SELECTED REFERENCES] (109)
- 19 program evaluation/ (19016)
- 20 early intervention/ (445)
- 21 evaluation studies/ (115898)
- 22 exp psychotherapy/ (92087)
- 23 or/19-22 (224707)
- 24 7 and 23 (352)
- 25 24 not 16 (158)
- 26 25 (158)
- 27 limit 26 to english language (116)
- 28 from 27 [SELECTED REFERENCES] (51)
- 29 treatment outcome/ (172710)
- 30 (7 and 29) not (16 or 25) (142)
- 31 dt.fs. (971727)
- 32 30 not 31 (51)
- 33 from 32 keep [SELECTED REFERENCES]

PsychInfo

- 1 rett syndrome/ (187)
- 2 autis\$.tw. (9804)
- 3 kanner's.tw. (74)
- 4 autism/ or pervasive developmental disorders/ or aspergers syndrome/ or autistic thinking/ (5930)
- 5 autistic children/ (3022)
- 6 early infantile autism/ (479)
- 7 or/1-6 (10221)
- 8 behavior therapy/ or behavior modification/ (15486)
- 9 cognitive therapy/ (8679)
- 10 (therap\$ adj3 condition\$.tw. (1189)
- 11 (behavio?r\$ adj3 modif\$.tw. (6283)
- 12 (behavio?r\$ adj3 therap\$.tw. (14449)
- 13 (behavio?r\$ adj3 intervention\$.tw. (7616)
- 14 parent training/ (3623)
- 15 or/8-14 (41229)
- 16 early intervention/ (3933)
- 17 intervention.ti. (11975)
- 18 16 or 17 (14279)

- 19 7 and 15 (984)
- 20 18 and 7 (329)
- 21 20 not 19 (203)
- 22 limit 21 to (english language and (100 childhood <birth to age 12 yrs> or 120 neonatal <birth to age 1 mo> or 140 infancy <age 2 to 23 mo> or 160 preschool age <age 2 to 5 yrs> or 180 school age <age 6 to 12 yrs>) and yr=1990-2004) (142)
- 23 from 22 keep [SELECTED REFERENCES] (70)
- 24 from 23 keep 1-70 (70)
- 25 program evaluation/ (3514)
- 26 educational program evaluation/ (3259)
- 27 social skills training/ (2728)
- 28 exp Social Integration/ (1714)
- 29 or/25-28 (11106)
- 30 7 and 29 (163)
- 31 30 not (19 or 20) (115)
- 32 limit 31 to (english language and 100 childhood <birth to age 12 yrs> and yr=1990-2004) (51)
- 33 from 32 keep [SELECTED REFERENCES] (30)

Embase

- 1 autism/ or asperger syndrome/ (4545)
- 2 Rett Syndrome/ (968)
- 3 autis\$.tw. (4493)
- 4 kanner's.tw. (12)
- 5 Infantile Autism/ (803)
- 6 behavior therapy/ or cognitive therapy/ (12307)
- 7 (therap\$ adj3 condit\$.tw. (4059)
- 8 (behavio?r\$ adj3 modif\$.tw. (3239)
- 9 (behavio?r\$ adj3 therap\$.tw. (2601)
- 10 (behavio?r\$ adj3 intervention\$.tw. (881)
- 11 health program/ (21545)
- 12 "parent"/ (6477)
- 13 or/6-12 (48267)
- 14 or/1-5 (6454)
- 15 13 and 14 (420)
- 16 limit 15 to (english language and yr=1990-2004 and (infant <to one year> or child <unspecified age> or preschool child <1 to 6 years> or school child <7 to 12 years>)) (204)
- 17 from 16 keep [SELECTED REFERENCES] (57)
- 18 evaluation/ (20445)
- 19 psychotherapy/ (19195)
- 20 health care quality/ (26414)
- 21 or/18-20 (65148)
- 22 14 and 21 (169)
- 23 22 not 15 (138)
- 24 limit 23 to (english language and yr=1990-2004 and (child <unspecified age> or preschool child <1 to 6 years> or school child <7 to 12 years>)) (49)
- 25 from 24 keep [SELECTED REFERENCES] (6)

Cinahl

- 1 Rett Syndrome/ (57)
- 2 autis\$.tw. (723)
- 3 kanner's.tw. (0)
- 4 Autism/ (858)
- 5 Child Development Disorders/ (607)
- 6 or/1-5 (1536)
- 7 behavior modification/ or behavior therapy/ or cognitive therapy/ (2847)
- 8 early intervention/ or early childhood intervention/ (1286)
- 9 (behavio?r adj3 (therap\$ or modificat\$ or condition\$ or intervent\$).tw. (973)
- 10 behavio?r.ti. (4245)

- 11 intervention\$.ti. (8671)
- 12 or/7-11 (16263)
- 13 6 and 12 (245)
- 14 13 (245)
- 15 limit 14 to (english and yr=1990-2003) (218)
- 16 limit 15 to (infant <1 to 23 months> or preschool child <2 to 5 years> or child <6 to 12 years>) (154)
- 17 from 16 keep [SELECTED REFERENCES] (46)
- 18 exp Psychotherapy/ (25391)
- 19 Program Evaluation/ (5708)
- 20 exp Evaluation Research/ (6711)
- 21 or/18-20 (36429)
- 22 6 and 21 (164)
- 23 limit 22 to (english and (infant <1 to 23 months> or preschool child <2 to 5 years> or child <6 to 12 years>) and yr=1990-2003) (113)
- 24 23 not 16 (52)
- 25 from 24 keep [SELECTED REFERENCES] (11)

Other sources

Combinations of the index terms and keywords from the above strategies were used to search other sources of information without controlled vocabulary, or where source did not permit sophisticated searching.

APPENDIX 3: EXCLUDED RETRIEVED PAPERS

Aldred, C., Pollard, C., Phillips, R., & Adams, C. (2001). Multidisciplinary social communication intervention for children with autism and pervasive developmental disorder: the Child's Talk project. *Educational & Child Psychology*, 18, 76-87.

Outcomes not reported. Describes intervention and a randomised-controlled evaluation of the effectiveness of this project is reported as ongoing.

Attwood, T. (2003). Frameworks for behavioral interventions. *Child & Adolescent Psychiatric Clinics of North America*, 12, 65-86.

Narrative review.

Baird, G., Cass, H., & Slonims, V. (2003). Diagnosis of autism. *BMJ*, 327, 488-493.

Narrative review.

Baker, B. L., & Feinfield, K. A. (2003). Early intervention. *Current Opinion in Psychiatry*, 16, 503-509.

Narrative review.

Bauminger, N. (2002). The facilitation of social-emotional understanding and social interaction in high-functioning children with autism: intervention outcomes. *Journal of Autism & Developmental Disorders*, 32, 283-298.

Not population of interest (n=15, all study participants aged 8 to 17 years).

Bargerhuff, M. E. (2003). Exploring the spectrum of autism and pervasive developmental disorders: Intervention strategies. *Remedial and special education*, 24, 255-256.

Book review.

Blackwell, J., & Niederhauser, C. (2003). Diagnose and manage autistic children. *Nurse Practitioner*, 28, 36-43; quiz 44-35.

Background.

Boyd, R. D., & Corley, M. J. (2001). Outcome survey of early intensive behavioral intervention for young children with autism in a community setting. *Autism*, 5, 430-441.

Case series (n=22).

Buschbacher, P. W., & Fox, L. (2003). Clinical forum. Understanding and intervening with the challenging behavior of young children with autism spectrum disorder. *Language, Speech & Hearing Services in the Schools*, 34, 217-227.

Case study.

Butter, E. M., Wynn, J., & Mulick, J. A. (2003). Early intervention critical to autism treatment. *Pediatric Annals*, 32, 677-684.

Narrative review.

Campbell, J. M. (2003). Efficacy of behavioral interventions for reducing problem behavior in persons with autism: a quantitative synthesis of single-subject research. *Research in Developmental Disabilities*, 24, 120-138.

Systematic review of case studies/series that have examined behavioral interventions for problem behaviour.

Carr, D. (2001). Parents' education as autism therapists: applied behaviour analysis in context. *Journal of Applied Research in Intellectual Disabilities*, 14, 157-160.

Book review.

Chandler, S., Christie, P., Newson, E., & Prevezer, W. (2002). Developing a diagnostic and intervention package for 2- to 3-year-olds with autism: outcomes of the frameworks for communication approach. *Autism*, 6, 47-69.

Descriptive study (n=10).

Charman, T. (2003). Why is joint attention a pivotal skill in autism? *Philosophical Transactions of the Royal Society of London - Series B: Biological Sciences*, 358, 315-324.

Experimental study measuring the concurrent and longitudinal associations between joint attention and other social communication abilities in a sample of young children with autism (n=9) or atypical autism/PDD-NOS (n=9). Same sample as Charman et al. (2003).

Charman, T., & Howlin, P. (2003). Research into early intervention for children with autism and related disorders: methodological and design issues. Report on a workshop funded by the Wellcome Trust, Institute of Child Health, London, UK, November 2001. *Autism*, 7, 217-225.

Conference proceedings. Discusses recommendations for improving quality of autism intervention research in the UK.

Charman, T., Baron-Cohen, S., Swettenham, J., Baird, G., Drew, A., & Cox, A. (2003). Predicting language outcome in infants with autism and pervasive developmental disorder. *International Journal of Language & Communication Disorders*, 38, 265-285.

Experimental study measuring the concurrent and longitudinal associations between joint attention and other social communication abilities in a sample of young children with autism (n=9) or atypical autism/PDD-NOS (n=9).

Cohen, S. (2001). Do-watch-listen-say: social and communication intervention for children with autism. *Journal of the Association for Persons with Severe Handicaps*, 26, 127-128.

Book review.

Cohen, I. L., Schmidt-Lackner, S., Romanczyk, R., & Sudhalter, V. (2003). The PDD Behavior inventory: a rating scale for assessing response to intervention in children with pervasive developmental disorder. *Journal of Autism and Developmental Disorders*, 33, 31-45.

Background.

Connecticut Birth to Three System (2002). Autistic spectrum disorder: intervention guidance for service providers and families of young children with autistic spectrum disorder. Hartford, CT: Connecticut Birth to Three System.

Narrative review/Expert opinion. This document is a service guideline for families and service providers.

Coq, J. M., des Noettes, A. L., Sarfaty, J., Thomas, N., & Bracha, Z. (2000). Prevention and early intervention in autistic disorders: a research action project. *Infant Mental Health Journal*, 21, 379.

Conference abstract.

Couper, J. J., & Sampson, A. J. (2003). Children with autism deserve evidence-based intervention. *Medical Journal of Australia*, 178, 424-425.

Editorial.

Cuccaro, M. L., Shao, Y. J., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., Abramson, R.K., et al. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry & Human Development*, 34, 3-17.

Experimental study on behaviour but no intervention described.

Dawson, G. W. R. (2000). Interventions to facilitate auditory, visual, and motor integration in autism: a review of the evidence. *Journal of Autism & Developmental Disorders*, 30, 415-422.

Narrative review. These interventions were not included in the scope of this report.

Delprato, D. J. (2001). Comparisons of discrete-trial and normalized behavioral language intervention for young children with autism. *Journal of Autism and Developmental Disorders*, 31, 315-325.

Systematic review of experimental studies. This 'critical' review examined a series of 10 controlled studies, 8 of 10 with multiple baseline or reversal designs. This review does provide explicit inclusion and exclusion criteria but gives no details of the search strategy used. In all eight studies with language criterion responses, normalised language training was more effective than discrete trial training.

Dillenburger, K. (2002). Raising a child with autism: a guide to applied behavior analysis for parents. *Autism*, 6, 438-440.

Book review.

Dunn, L. M. (2000). Innovation in practice. Using "learning stories" to assess and design programs for young children with special needs in New Zealand. *Infants & Young Children*, 13, 73-82.

Not relevant to the topic. Not a controlled study of intervention for autism in young children.

Eikeseth, S. (2001). Recent critiques of the UCLA young autism project. *Behavioral Interventions*, 16, 249-264.

Background (Lovaas Therapy).

Erba, H. W. (2000). Early intervention programs for children with autism: conceptual frameworks for implementation. *American Journal of Orthopsychiatry*, 70, 82-94.

Outcomes not reported. Describes four early intervention programmes.

Ferraro, F. R. (2001). Survey of treatments for childhood autism. *Psychology & Education: an Interdisciplinary Journal*, 38, 29-41.

Narrative review. Eclectic summary of a variety of interventions including applied behavioral analysis.

Field, T., Sanders, C., & Nadel, J. (2001). Children with autism display more social behaviors after repeated imitation sessions. *Autism*, 5, 317-323.

Comparative study with random assignment to an imitation or contingently responsive playgroup, however no validated outcome measures were used. Frequency of behaviours was assessed using videotape recordings.

Gabriels, R. L., Hill, D. E., Pierce, R. A., Rogers, S. J., & Wehner, B. (2001). Predictors of treatment outcome in young children with autism - a retrospective study. *Autism*, 5, 407-429.

Case series (n=17), post-test. Pre-test measures obtained retrospectively from record review.

Goldstein, H. (2002). Communication intervention for children with autism: a review of treatment efficacy. *Journal of Autism and Developmental Disorders*, 32, 373-396.

Narrative review. Appears to be a thorough summary of the literature but no details pertaining to the use of a systematic search strategy or selection criteria for included and excluded articles.

Goldstein, H. (2003). Interventions to facilitate auditory, visual, and motor integration: "show me the data" - response. *Journal of Autism and Developmental Disorders*, 33, 553-555.

Commentary.

Harris, S. L., & Handleman, J. S. (2000). Age and IQ at intake as predictors of placement for young children with autism: a four- to six-year follow-up. *Journal of Autism & Developmental Disorders*, 30, 137-142.

Background.

Harris, S. L., & Delmolino, L. (2002). Applied Behavior analysis: its application in the treatment of autism and related disorders in young children. *Infants and Young Children*, 14, 11-17.

Background.

Harrower, J. K., & Dunlap, G. (2001). Including children with autism in general education classrooms - a review of effective strategies. *Behavior Modification*, 25, 762-784.

Narrative review. Discusses peer intervention and multicomponent interventions.

Hastings, R. P., & Symes, M. D. (2002). Early intensive behavioral intervention for children with autism: parental therapeutic self-efficacy. *Research in Developmental Disabilities*, 23, 332-341.

Not population of interest. Descriptive study.

Hastings, R. P., & Johnson, E. (2001). Stress in UK families conducting intensive home-based behavioral intervention for their young child with autism. *Journal of Autism & Developmental Disorders*, 31, 327-336.

Outcomes of interest not reported. Study looks primarily on parental stress.

Horner, R. H., Carr, E. G., Strain, P. S., Todd, A. W., & Reed, H. K. (2002). Problem behavior interventions for young children with autism: a research synthesis. *Journal of Autism & Developmental Disorders*, 32, 423-446.

Systematic review of uncontrolled experimental studies. For studies included in this review, a single subject design was used (n=3 or more for eight out of nine of the studies).

Howlin, P. (2000). Autism and intellectual disability: diagnostic and treatment issues. *Journal of the Royal Society of Medicine*, 93, 351-355.

Narrative review. Discusses principal components of effective treatments.

Howlin, P. (2002). The research basis for autism intervention. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 43, 549-549.

Book review.

Hwang, B., & Hughes, C. (2000). The effects of social interactive training on early social communicative skills of children with autism. *Journal of Autism & Developmental Disorders*, 30, 331-343.

Systematic review of uncontrolled experimental studies. For studies included in review, methodological design was primarily multiple-baseline or probe across subjects and or reversal (n=14 studies) or pretest-posttest (n=1 study) and pretest-posttest (n=1 study).

Jacobson, J. W. (2000). Early intensive behavioral intervention: emergence of a consumer-driven service model. *Behavior Analyst*, 23, 149-171.

Background.

Jahr, E., & Eldevik, S. (2002). Teaching cooperative play to typical children utilizing a behavior modeling approach: a systematic replication. *Behavioral Interventions*, 17, 145-157.

Case series (n=3).

Jensen, V. K., & Sinclair, L. V. (2002). Treatment of autism in young children: behavioral intervention and applied behavior analysis. *Infants and Young Children*, 14, 42-52.

Background.

Kabot, S., Masi, W., & Segal, M. (2003). Advances in the diagnosis and treatment of autism spectrum disorders. *Professional Psychology-Research and Practice*, 34, 26-33.

Background.

Kasari, C. (2002). Assessing change in early intervention programs for children with autism. *Journal of Autism and Developmental Disorders*, 32, 447-461.

Background. Intervention studies based on comprehensive interventions for young children with autism are reviewed in respect to well-designed treatment studies.

Kasari, C., Freeman, S. F. N., & Paparella, T. (2001). Early intervention in autism: joint attention and symbolic play. In L. M. Glidden (Ed.), *International review of research in mental retardation: autism* (pp. 207-237). San Diego: Academic Press.

Book Chapter.

Kobayashi, R. (2000). Early intervention for infants with autistic spectrum disorders in Japan. *Infant Mental Health Journal*, 21, 372.

Conference abstract.

Koegel, R. L., Koegel, L. K., & McNeerney, E. K. (2001). Pivotal areas in intervention for autism. *Journal of Clinical Child Psychology*, 30, 19-32.

Narrative review.

Koppenhaver, D. A., Erickson, K. A., Harris, B., McLellan, J., Skotko, B. G., & Newton, R. A. (2001). Storybook-based communication intervention for girls with Rett syndrome and their mothers. *Disability & Rehabilitation*, 23, 149-159.

Not population of interest (n=6).

Krasny, L., Williams, B. J., Provencal, S., & Ozonoff, S. (2003). Social skills interventions for the autism spectrum: essential ingredients and a model curriculum. *Child & Adolescent Psychiatric Clinics of North America*, 12, 107-122.

No outcomes reported. Describes social skills interventions.

Kubina, R. M., Morrison, R., & Lee, D. L. (2002). Benefits of adding precision teaching to behavioral interventions for students with autism. *Behavioral Interventions*, 17, 233-246.

Narrative review. Describes precision teaching as an adjunct to behavioral intervention.

Laushey, K. M., & Heflin, L. J. (2000). Enhancing social skills of kindergarten children with autism through the training of multiple peers as tutors. *Journal of Autism and Developmental Disorders*, 30, 183-193.

Case study (n=2).

Le Couteur, A. (2003). National Autism Plan for Children (NAPC): plan for the identification, assessment, diagnosis and access to early interventions for pre-school and primary school aged children with autism spectrum disorders (ASD). London: The National Autistic Society for NIASA.

Narrative Review/Expert opinion. This document is a service guideline for carers/families, teachers, advocates, health professionals and service providers. Broad focus on vision of what a good service for the child with suspected and proven ASD would look like. Working party recommendations are allocated grades of evidence.

Lord, C. (2000). Commentary: achievements and future directions for intervention research in communication and autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 30, 393-398.

Commentary.

Lovaas, O. I., & Smith, T. (2003). Early and intensive behavioral intervention in autism. In A. E. Kazdin (Ed.), *Evidence-based psychotherapies for children and adolescents* (pp. 325-340). New York: Guilford Press.

Book chapter.

Luiselli, J. K., O'Malley Cannon, B., Ellis, J. T., & Sisson, R. W. (2000). Home-based behavioral intervention for young children with autism/pervasive developmental disorder: a preliminary evaluation of outcome in relation to child age and intensity of service delivery. *Autism*, 4, 426-438.

Descriptive study of home-based services for young children (n=16) with autism or PDD.

Magerotte, G. (2000). From quality of services to quality of life of persons with autism - contributions to research, training, and community services of the University of Mons-Hainaut. *International Journal of Mental Health*, 29, 60-77.

Not relevant to the topic as not a study of behavioural intervention.

Magiati, I., & Howlin, P. (2003). A pilot evaluation study of the Picture Exchange Communication System (PECS) for children with autistic spectrum disorders. *Autism*, 7, 297-320.

Descriptive study (n=24). Note the data for this study was from a subset of a larger, ongoing longitudinal study of early intervention.

Mahoney, G., & Perales, F. (2003). Using relationship-focused intervention to enhance the social-emotional functioning of young children with autism spectrum disorders. *Topics in Early Childhood Special Education*, 23, 77-89.

Experimental study, pre-test/post-test analysis of young children (n=20) with autism.

Maine Administrators of Services for Children with Disabilities (MADSEC) (2000). Report of the MADSEC autism task force. Manchester, ME: MADSEC.

Narrative review/Expert opinion. This document is a service guideline for families, health professionals and service providers. The autism task force characterised the research pertaining to eight interventions routinely used for children with autism as meeting one of four evidence-based criteria. Applied behaviour analysis (ABA) was graded as “substantiated as effective based upon the scope and quality of research” although it is not clear from the executive summary on what basis this was determined. This report also suggests that ABA evaluative procedures are effective not only for behaviourally-based interventions, but also for the systematic evaluation of the efficacy of any intervention intended to affect learning and behaviour.

Marcus, L. M., Garfinkle, A., & Wolery, M. (2001). Issues in early diagnosis and intervention with young children with autism. In E. Schopler & N. Yirmiya (Eds.), *The research basis for autism intervention* (pp. 171-185). New York: Kluwer Academic/Plenum Publishers.

Book chapter.

Mastergeorge, A. M., Rogers, S. J., Corbett, B. A., & Solomon, M. (2003). Non-medical interventions for autism spectrum disorders. In S. Ozonoff & S. Rogers (Eds.), *Autism spectrum disorders: A research review for practitioners* (pp. 133-160). Washington: American Psychiatric Publishing, Inc.

Book chapter.

Maurice, C., Mannion, K., Letso, S., & Perry, L. (2001). Parent voices: difficulty in accessing behavioral intervention for autism; working toward solutions. *Behavioral Interventions*, 16, 147-165.

Background.

McConnell, S. R. (2002). Interventions to facilitate social interaction for young children with autism: review of available research and recommendations for educational intervention and future research. *Journal of Autism and Developmental Disorders*, 32, 351-372.

Narrative review. Appears to be a thorough summary of the literature but no details pertaining to the use of a systematic search strategy or selection criteria for included and excluded articles.

Moes, D. R., & Frea, W. D. (2002). Contextualized behavioral support in early intervention for children for autism and their families. *Journal of Autism & Developmental Disorders*, 32, 519-533.

Experimental study, multiple baseline design (n=3) of functional assessment and functional communication training.

Moore, V., & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism*, 7, 47-63.

Not relevant to topic as not a study of a behavioral intervention or comprehensive programme. This paper discusses the development of an early diagnostic service and the reliability of early diagnosis in young children.

Mudford, O. C., Martin, N. T., Eikeseth, S., & Bibby, P. (2001). Parent-managed behavioral treatment for preschool children with autism: some characteristics of UK programs. *Research in Developmental Disabilities*, 22, 173-182.

Descriptive study looking at children (n=75) who had participated in early, intensive behavioral intervention in the UK.

Mulick, J. A., & Butter, E. M. (2002). Educational advocacy for children with autism. *Behavioral Interventions*, 17, 57-74.

Narrative review.

Murray-Slutsky, C., & Paris, B. A. (2003). Exploring the spectrum of autism and pervasive developmental disorders: intervention strategies. *Remedial and Special Education*, 24, 255-256.

Book Review.

Paul, R. (2003). Promoting social communication in high functioning individuals with autistic spectrum disorders. *Child & Adolescent Psychiatric Clinics of North America*, 12, 87-106.

Describes interventions but no outcomes reported.

Pelios, L. V., & Lund, S. K. (2001). A selective overview of issues on classification, causation, and early intensive behavioral intervention for autism. *Behavior Modification*, 25, 678-697.

Background.

Prizant, B. M., Wetherby, A. M., Rubin, E., & Laurent, A. C. (2003). The SCERTS model - a transactional, family-centered approach to enhancing communication and socioemotional abilities of children with autism spectrum disorder. *Infants and Young Children*, 16, 296-316.

No outcomes reported. Describes model for intervention.

Roberts, C., Mazzucchelli, T., Taylor, K., & Reid, R. (2003). Early intervention for behaviour problems in young children with developmental disabilities. *International Journal of Disability, Development & Education*, 50, 275-292.

Not restricted to population of interest.

Salt, J., Sellars, V., Shemilt, J., Boyd, S., Coulson, T., & McCool, S. (2001). The Scottish Centre for Autism preschool treatment programme. I: A developmental approach to early intervention. *Autism*, 5, 362-373.

Describes National Health Service (NHS) programme developed in Scotland.

Sasaki, M. (2000). Aspects of autism in Japan before and after the introduction of TEACCH. *International Journal of Mental Health*, 29, 3-18.

Descriptive study.

Schreck, K. A. (2000). It can be done: an example of a behavioral Individualized Education Program (IEP) for a child with autism. *Behavioral Interventions*, 15, 279-300.

Case study of individualised education programme (IEP).

Schreck, K. A. (2001). Behavioral treatments for sleep problems in autism: empirically supported or just universally accepted? *Behavioral Interventions*, 16, 265-278.

Narrative review. Few details of review methodology provided (e.g., computer search of all relevant literature was conducted). Concludes that there is a lack of published research on the effectiveness of ABA treatment of sleep problems for children with autism.

Schreck, K. A. (2002). Autism Academy CourseWare: behavioral programming for children with autism. *Behavioral Interventions*, 17, 107-109.

Not a study.

Schweigert, P., & Rowland, C. (2000). Tangible symbols, tangible outcomes in communication intervention for children with autism. *Journal of Intellectual Disability Research*, 44, 456-456.

Conference abstract.

Shields, J. (2000). The NAS EarlyBird Programme: autism-specific early intervention for parents. *Professional Care of Mother & Child*, 10, 53-54.

Describes UK based early intervention programme for parents. Not a study however a doctoral dissertation for an evaluation of this programme is referenced.

Shields, J. (2001). The NAS EarlyBird Programme: partnership with parents in early intervention. National Autistic Society. *Autism*, 5, 49-56.

Describes programme for parents and refers to an efficacy study/further monitoring but no outcomes of interest reported.

Siller, M., & Sigman, M. (2002). The behaviors of parents of children with autism predict the subsequent development of their children's communication. *Journal of Autism & Developmental Disorders*, 32, 77-89.

Not relevant to the topic. The focus of this study was not on evaluating the efficacy of a specific intervention or comprehensive programme. Standardised measures were used to report outcomes for children with autism compared with typically developing children over time.

Smith, T., Lovaas, N. W., & Lovaas, O. I. (2002). Behaviors of children with high-functioning autism when paired with typically developing versus delayed peers: a preliminary study. *Behavioral Interventions*, 17, 129-143.

Experimental study, within-subjects, crossover design examining the effect of placement with typically developing or delayed peer on nine children with high-functioning autistic disorder in a comprehensive behavioral treatment program. Did not use standardised outcome measures.

Sofronoff, K., & Farbotko, M. (2002). The effectiveness of parent management training to increase self-efficacy in parents of children with Asperger syndrome. *Autism*, 6, 271-286.

Comparative study with wait-list control group of parents who had originally consented to the study but declined intervention before commencement. The mean age of children involved was 8 years, 3 months.

Solomon, R., Necheles, J., Ferch, C., & Crawford, K. (2003). The University of Michigan PLAY Project Home Consultation (PPHC) program for young children with autism: initial program evaluation. *Pediatric Research*, 53, 79A.

Conference abstract.

Solomon, R., Necheles, J., Ferch, C., & Crawford, K. (2003). Program evaluation of a statewide autism training and early intervention project: The University of Michigan PLAY Project. *Pediatric Research*, 53, 79A-80A.

Conference abstract.

Stahl, L., & Pry, R. (2002). Joint attention and set-shifting in young children with autism. *Autism*, 6, 383-396.

Case-control study of children with autism (n=15) and normally developing, concurrent controls (n=21). Assessment of skill on tasks of set-shifting and joint-attention but no intervention evaluated.

Strain, P. S. (2001). Empirically based social skill intervention: a case for quality-of-life improvement. *Behavioral Disorders*, 27, 30-36.

Narrative review.

Taubman, M., Brierley, S., Wishner, J., Baker, D., McEachin, J., & Leaf, R. B. (2001). The effectiveness of a group discrete trial instructional approach for preschoolers with developmental disabilities. *Research in Developmental Disabilities*, 22, 205-219.

Experimental study, multiple-baseline design across tasks. Mixed sample of eight subjects (n=2 with autism).

Tregear, S. (2000). Comprehensive programs for the treatment of children with autism, *16th Annual Meeting of the International Society of Technology Assessment in Health Care: June 18-21, The Hague, The Netherlands*. Montreal: ISTAHC.

Conference Abstract.

Volkmar, F. (2003). Autism. *Lancet*, 362, 1133-1141.

Background.

Webster, A., Webster, V., & Feiler, A. (2002). Research evidence, polemic and evangelism: how decisions are made on early intervention on autistic spectrum disorder. *Educational & Child Psychology*, 19, 54-67.

Narrative review.

Whalen, C., & Schreibman, L. (2003). Joint attention training for children with autism using behavior modification procedures. *Journal of Child Psychology & Psychiatry & Allied Disciplines*, 44, 456-468.

Experimental study using single subject, multiple-baseline design. Typical children did not participate in the intervention phases of this investigation and were not considered a 'control' group, rather they were assessed to identify 'normal' levels of social behaviours during the preschool years.

Williams, K. R., & Wishart, J. G. (2003). The Son-Rise Program intervention for autism: an investigation into family experiences. *Journal of Intellectual Disability Research*, 47, 291-299.

No outcomes of interest reported.

Wolery, M., & Garfinkle, A. N. (2002). Measures in intervention research with young children who have autism. *Journal of Autism and Developmental Disorders*, 32, 463-478.

Systematic review but focus on measurement practice rather than intervention.

Woods, J. J., & Wetherby, A. M. (2003). Early identification of and intervention for infants and toddlers who are at risk for autism spectrum disorder. *Language, Speech & Hearing Services in the Schools*, 34, 180-193.

Narrative review.

APPENDIX 4: APPRAISED RETRIEVED PAPERS

Secondary studies

Bassett, K., Green, G., & Kazanjian, A. (2000). *Autism and Lovaas treatment: a systematic review of effectiveness evidence*. Vancouver: BC Office of Health Technology Assessment.

Diggle, T., McConachie, H. R., & Randle, V. R. L. (2003). Parent-mediated early intervention for young children with autism spectrum disorder. *Cochrane Database of Systematic Reviews*, 2.

Finch, L., & Raffaele, C. (2003). Developing expert practice. Intensive behavioural intervention for children with autism: a review of the evidence. *Occupational Therapy Now*, 5, 20-23.

Ludwig, S., & Harstall, C. (2001). Intensive intervention programs for children with autism. Edmonton: Alberta Heritage Foundation for Medical Research.

McGahan, L. (2001). Behavioural interventions for preschool children with autism. Ottawa: Canadian Coordinating Office for Health Technology Assessment.

Primary studies

Bibby, P., Eikeseth, S., Martin, N. T., Mudford, O. C., & Reeves, D. (2001). Progress and outcomes for children with autism receiving parent-managed intensive interventions. *Research in Developmental Disabilities*, 22, 425-447.

Bibby, P., Eikeseth, S., Martin, N. T., Mudford, O. C., & Reeves, D. (2002). Progress and outcomes for children with autism receiving parent-managed intensive interventions. *Research in Developmental Disabilities*, 23, 81-104.⁶

Drew, A., Baird, G., Baron-Cohen, S., Cox, A., Slonims, V., Wheelwright, S., et al. (2002). A pilot randomised control trial of a parent training intervention for pre-school children with autism - preliminary findings and methodological challenges. *European Child & Adolescent Psychiatry*, 11, 266-272.

Eikeseth, S., Smith, T., Jahr, E., & Eldevik, S. (2002). Intensive behavioral treatment at school for 4- to 7-year-old children with autism. A 1-year comparison controlled study. *Behavior Modification*, 26, 49-68.

Salt, J., Shemilt, J., Sellars, V., Boyd, S., Coulson, T., & McCool, S. (2002). The Scottish centre for Autism Preschool Treatment Programme. II: The results of a controlled treatment outcome study. *Autism*, 6, 33-46.

Smith, T., Groen, A. D., & Wynn, J. W. (2000). Randomized trial of intensive early intervention for children with pervasive developmental disorder. *American Journal on Mental Retardation*, 105, 269-285.

Smith, T., Groen, A. D., & Wynn, J. W. (2000). Randomized trial of intensive early intervention for children with pervasive developmental disorder. *American Journal on Mental Retardation*, 105, 269-285.

⁶ This study by Bibby et al. appears to have been published twice. The article appears in the same journal, in exactly the same format in consecutive years (2001 and 2002). The more recent citation has been used in this report.

APPENDIX 5: WEBSITE SOURCES OF INFORMATION

HTA Organisations	Website URL
Alberta Heritage Foundation for Medical Research (AHFMR)	http://www.ahfmr.ab.ca/
Agency for Health Research Quality (AHRQ)	http://www.ahrq.gov
L'Agence nationale d'Accréditation et d'Evaluation en Santé	http://www.anaes.fr
British Columbia Office of Health Technology Assessment (BCOHTA)	http://www.chspr.ubc.edu.ca/bcohta
Canadian Coordinating Office for Health Technology Assessment (CCOHTA)	http://www.ccohta.ca
ECRI (USA)	http://www.ecri.org
Institute for Clinical Systems Improvement (ICSI)	http://www.icsi.org
Institute of Technology Assessment of the Austrian Academy of Science (ITA)	http://www.oeaw.ac.at/ita/hta/
International Network of Agencies for Health Technology Assessment (INAHTA)	http://www.inahta.org
International Society of Technology Assessment in Health Care	http://www.istahc.org
National Coordinating Centre for Health Technology Assessment (NCCHTA)	http://www.soton.ac.uk/~hta
New Zealand Health Technology Assessment (NZHTA)	http://nzhta.chmeds.ac.nz
Other Organisations	
Australian National Health & Medical Research Council	http://www.health.gov.au/nhmrc/index.htm
National Institute for Mental Health (NIMH)	http://www.nimh.nih.gov/
Professional Associations/Societies	
Controlled Clinical Trials	http://www.controlled-trials.com/
Clinicaltrials.gov	http://www.clinicaltrials.gov