Parent implemented early intervention for young children with autism spectrum disorder: a systematic review

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Abstract

Background Recent estimates concerning the prevalence of autism spectrum disorder (ASD) suggest that at least one in 200 children is affected. This group of children and families have important service needs. The involvement of parents in implementing intervention strategies designed to help their autistic children has long been accepted as helpful. The potential benefits are increased skills and reduced stress for parents as well as children. **Methods** This research review focused on interventions for children aged 1–6 years, and was carried out using systematic methodology: a comprehensive search of psychological, educational and biomedical databases, as well as bibliographies and reference lists of key articles, contact with experts in the field, and hand search of key journals. Only studies which involved a concurrent element of control were included. **Results** The review found very few studies that had adequate research design from which to draw conclusions about the effectiveness of parent-implemented early intervention. Both randomized and controlled studies tended to suggest that parent training leads to improved child communicative behaviour, increased maternal knowledge of autism, enhanced maternal communication style and parent child interaction, and reduced maternal depression. **Conclusion** It seems that parent training can successfully contribute to intervention for young children with ASD. However, the review highlights the need for improved research in this area.

Introduction

Definitions and prevalence

Autism is the core disorder of the pervasive developmental disorders (PDDs) (ICD-10, WHO 1993; DSM IV, APA 1994), and is evident before the age of 3 years. Autism is, however, a dimension (or dimensions) rather than a distinct category and is generally understood as a spectrum, along which children experience varying degrees of difficulty in the areas of communication, social interaction, and a tendency toward repetitive behaviours and lack of imagination. Children with autism spectrum disorder (ASD) lack understanding of how to initiate and respond to joint attention with another person, have difficulties in social timing of communication, and may not understand other people's intentions as expressed through language and gestures, even though they may appear affectionate and want to be with other people socially. They have difficulty with organizing their responses, and with inhibition of repetitive behaviours and interests. Children with core autism have more profound difficulties, and are more likely

to have associated learning difficulties, than those who have other diagnoses within the PDDs (e.g. Asperger syndrome).

Estimates of the prevalence of ASD are continually being revised. Fombonne *et al.* (2001) reported the prevalence of PDDs in the British national survey of mental health in children aged 5–15 years as 26.1/10 000 (with a 95% confidence interval of 16.2–36.0/10 000). This represents more than one individual in 400, with a male-to-female ratio of 4.8 : 1. A prevalence of 26.1 per 10 000 children is lower than other recent best estimates, from surveys of young children with focused case-finding and diagnosis e.g. Baird *et al.* (2000) at 57.9 children per 10 000 and Chakrabarti & Fombonne (2001) at 62.6 children per 10 000). The estimated prevalence for core autism has been revised upwards also, from 5 per 10 000 (Fombonne 1999) to 16.8 per 10 000 (Chakrabarti & Fombonne 2001) and 30.8 per 10 000 (Baird *et al.* 2000). Thus, although in the past autism was considered to be a rare disorder, now that the underlying characteristics and varying severity of the disorder are more widely recognized by health professionals, teachers and parents, detection rates for autism and ASD have increased considerably.

Impact and early intervention

Many aspects of the children's difficulties gradually come to the notice of parents during the first 2 years of the child's life, undermining their confidence in their ability as parents, as well as causing concern about what the difficulties mean. Children with ASD frequently pose considerable behavioural challenges to their parents and other family members.

The children need help to develop early skills in establishing joint attention, imitation of others, communicating interest and meaning as well as immediate wants, understanding the language of others, getting on with and enjoying the company of other people, tolerating change, and so on. This broad agenda has spawned a broad range of approaches to early intervention, with controversial claims for their efficacy (Green 1996a; Harris 1998; Rogers 1998a,b). Some intensive programmes, involving up to 40 hours of structured input to the child every week, have claimed to restore 'normal functioning' (Kaufman 1981; McEachin *et al.* 1993). All compre-

hensive programmes for young children with ASD (see review by the National Research Council 2001) explicitly involve parents in implementing the strategies, to a greater or lesser extent. Other programmes are based in special education nurseries, with additional training of parents in specific skills (e.g. Ozonoff & Cathcart 1998) and a range of supports offered to families (Prizant *et al.* 2003). Finally, there are intervention approaches involving parents in behaviour management and promotion of communication skills which are non-intensive, utilizing teaching within everyday situations (e.g. Howlin *et al.* 1987; Sussman 1999; Shields 2001).

Training parents

The involvement of parents in implementing intervention strategies designed to help their children with autism has a history stretching back at least three decades (e.g. Schopler & Reichler 1971). There is some evidence to suggest that earlier intervention for children with ASD is better (Rogers 1996), which logically implies involvement of parents. Increased parental skills allow for continual opportunities for children's learning in a range of situations. Training parents as 'co-therapists' allows consistent handling, and ensures that intervention is appropriate in enhancing children's earliest social relationships. The potential benefits of parent training are increased skills, renewed confidence and reduced stress for parents as well as for children. Group training for parents in new skills has been demonstrated to facilitate mutual support (e.g. Baxendale *et al.* 2001; Symon 2001). Measurement of the efficacy and effectiveness of the involvement of parents in programmes to help their children's development should include a range of outcomes: child developmental progress, parent–child interaction patterns, parents' knowledge, attitudes and stress levels, family functioning, and cost-benefit analysis.

What evidence do we have that parent-implemented early intervention for children with ASDs is beneficial? Most previous reviews of early intervention in this area (e.g. Green 1996b; Dawson & Osterling 1997; Connor 1998; Harris 1998; Gresham *et al.* 1999; Probst 2001) have lacked a systematic approach, minimizing their comprehensiveness and validity. Three other reviews have been

more systematic (Smith 1999; Delprato 2001; National Research Council 2001), using a search strategy to locate studies comprehensively and assessing their quality, but they either did not exclude uncontrolled studies or relied mostly on single-case designs. By including studies that lack scientific rigour or generalizability and which contain methodological weaknesses, the reviews have drawn conclusions from evidence which may be confounded. In addition, Smith (1999) used a very narrow basis of outcome comparison, reporting chiefly on children's intellectual functioning; although a large proportion of children with autism have delays in intellectual functioning, dependence on intelligence tests as the sole outcome does not allow for the accurate evaluation of different intervention approaches. The most satisfactory scientific review of early intervention in autism was conducted for the New York State Department of Health (1999) in order to develop clinical practice guidelines; however, it did not include a specific focus on the effectiveness of parental involvement in implementing intervention. Diggle *et al.* (2003) systematically reviewed evidence from randomized controlled trials of parent-mediated early intervention, but reported only outcome measures for children.

Methods

This review of research into parent-implemented intervention for pre-school children with autism has taken a broad approach in terms of intervention type. It has documented and compared both direct and indirect effects of intervention, encompassing outcomes that relate to the child, to the parents and to the family as a whole.

Search strategy

The authors conducted a computer search of the following databases; ERIC, The Cochrane Library, MEDLINE, EMBASE, PsycInfo, CINAHL, Dissertation Abstracts International, Social Sciences Abstracts, Sociological Abstracts, Linguistics and Language Behavior Abstracts, National Research Register, LILACS. No date limit was introduced in the search strategy. The search strategy was designed to combine four groups of key terms relating to the

child, parents, parent training and ASD. In all searches appropriate truncations and possible misspellings were included and the search terms were adapted for different databases. The strategy was designed in consultation with the Cochrane Collaboration Developmental, Psychosocial and Learning Problems group. Other sources of information were examined including the bibliographies of systematic and non-systematic reviews and reference lists of key articles identified through the search strategy. Experts in the field were contacted via email in order to identify unpublished studies. Key journals were hand searched to identify studies that have not been electronically catalogued in databases.

Inclusion criteria

Studies were included where parents/carers were the main mediators of the intervention and where the study intervention had a significant focus on parent-implemented intervention. All included studies had a concurrent control or comparison group of children. Single case experimental designs were not included, on the basis of publication bias and uncertain generalizability. Published and unpublished studies were considered, with no language or time limit restrictions. Studies included in this review involved intervention for children aged 1 years to 6 years 11 months, diagnosed with any of the following: autism, ASD, Asperger syndrome, PDD, PDD not otherwise specified.

Critical appraisal

Critical appraisal of included studies considered the following: (1) degree of allocation blindness in those studies which used random assignment to group, independent diagnosis and assessments; (2) multiple intake and follow-up measures to assess functioning across different domains (intelligence, adaptive functioning, etc.), (3) multiple measures to assess both child and parental or family outcomes, (4) length of follow-up assessments; and (5) the use of standardized tests and diagnostic instruments. Reporting the representativeness of the sample was considered including how the sample was obtained and who was excluded, the outcomes of participants who withdrew, data on individual difference and demographic

information. Appraisal of the included studies was carried out by two reviewers (TD who had specific training in conducting systematic reviews, and HM an experienced clinician and researcher in child disability including autism).

Analysis

Continuous data were analysed on the basis of post-test means and standard deviations. Standardized mean differences (MDs), and for meta-analysis weighted mean difference (WMD), 95% confidence intervals and P-values were calculated on the post-intervention outcome measures between groups using Hedges adjusted g (similar to Cohen's d) within the RevMan programme (Cochrane Collaboration).

Due to excessive levels of heterogeneity between study interventions and outcomes it was possible to compare data directly only from two included studies in the form of a meta-analysis.

Results

The search strategy located over 15 000 articles; there were many duplications, and irrelevant articles located through the use of a wide search strategy. From the initial yield of citations, 439 articles qualified for further inspection based on the abstract, 369 of which were excluded and not formally reviewed. Reasons for exclusions included: discussion article only, children with speech and language impairment only, etc. Thus, 70 studies appeared possibly to meet the inclusion criteria and qualified for formal review. The senior authors were approached (via email:) to find out if they were aware of any published or unpublished studies that had not been identified through the database searches, yielding one study which was added to the list for formal review (McClannahan et al. 1982).

Seventy-one studies were obtained in full for formal review. Five studies were published in a language other than English: two from Italy (Micheli 1999; Panerai et al. 2000), two from Japan (Ikeda et al. 1974; Ono 1994), and one from Turkey (Sucuoglu et al. 1994). These studies were translated to a degree necessary to understand the essentials of the research design. Nine studies were unpublished doctoral dissertations, and one was an unpublished conference paper (Chambliss & Doughty 1994). Fifty-nine of these studies were excluded from further review, primarily on methodological grounds ($n = 46$), age of the children, or the degree to which parents were the main mediators of intervention.

Thus, 12 studies met the inclusion requirements for review. These have been reported according to their methodological design in a hierarchy of evidence, with randomized controlled trials (RCTs) presented in Table 1 and controlled group studies presented in Table 2. (Three of the controlled group studies are related papers and so will be reported hereafter as in Howlin et al. 1987).

It is important to note that all statistics have been calculated for this review on post-intervention scores. This is based on the assumption that the samples in each arm of the studies were equivalent, which assumption holds in the case of RCTs. However, in controlled studies a systematic difference may exist between comparison groups. Therefore, possible pre-treatment differences between groups were investigated and no statistically significant group differences in demographic or outcome measures were reported pre-intervention.

Methodological quality of included studies

Randomized controlled trials

Four studies met full Cochrane criteria for review in terms of randomization, that is, Aldred et al. (2004), Drew et al. (2002), Jocelyn et al. (1998), and Smith et al. (2000a). Three other studies described as randomized did not report the method of randomization used (Sherman et al. 1988; Schreibman et al. 1991; Koegel et al. 1996), nor if this process was concealed from both clinicians and participants. Two randomized studies did not specify the diagnostic tools used (Sherman et al. 1988; Smith et al. 2000a). One study did not carry out blind or independent assessment (Sherman et al. 1988). Only Smith et al. (2000a) carried out a long-term follow-up. Thus the basic design of available studies described as RCTs had many shortcomings.

Four randomized studies mentioned how representative the sample was in terms of socio-economic status, although these data were not compared to the general population. However, the method by which

Table 1 Randomized controlled studies of parent training for young children with autism spectrum disorder

Study	Participants	Design	Outcome measures and results
Aldred et al. (2004)	Intervention n = 14 Control n = 14 Age 24–71 months Diagnosis confirmed by Autism Diagnostic Interview and Autism Diagnostic Observation Schedule	**Social communication training for parents vs. routine care.** Randomization stratified for age and severity. Pre- and post-measures at 12 month interval. Intervention: individual parent psycho-educational workshops, then individual monthly clinic sessions for 6 months, then less frequent (SCT). Parents asked to do 30 minutes practice per day. Also received routine local care. Control: ongoing local educational and therapy provision (RC)	**Child outcomes:** Autism Diagnostic Observation Schedule social-communication algorithm score improved (SCT > RC); MacArthur Communication Development Inventory words understood (NS), words said (NS); Vineland Adaptive Behavior Scales (NS) **Interaction outcomes:** child communication acts (SCT > RC), parent synchrony (SCT > RC), shared attention (NS) **Parental outcome:** Parenting Stress Index (NS)
Drew et al. (2002)	Intervention n = 12 Control n = 12 Mean age 22 months ICD10 diagnosis of autism confirmed by Autism Diagnostic Interview	**Joint attention focussed parent training vs. routine care.** Randomization – seven refused. Pre- and post-measures at 12.3 month interval. Intervention: individual home sessions every 6 weeks (JAFPT). Parents then asked to do 30–60 minutes per day. Also received routine local care. Control: ongoing local education and therapy provision (3 started ABA) (RC)	**Child outcomes:** MacArthur Communication Development Inventory words understood (NS), words said (NS) **Parental outcome:** Parenting Stress Index (NS)
Jocelyn et al. (1998)	Intervention n = 16 Control n = 19 Age 24–72 (mean 43 months) DSM III diagnosis of autism confirmed by Childhood Autism Rating Scale	**Day care plus parent training vs. day care alone.** Randomization stratified according to severity. Pre- and post-measures at 12 week interval. Intervention: Autism pre-school programme (APP) parents were trained for 15 hours over 10 weeks in functional analysis. Children attended a day care centre during this time. Control: Community Day Care (DC) children received 15 hours of day care over 10 weeks plus social worker support	**Child outcomes:** Autism Behaviour Checklist (independent assessor and parental) (NS); Early intervention/pre-school developmental profile (NS) **Parental outcomes:** Stress-Arousal Checklist (NS), Family Assessment Measure (NS), TRE-ADD Autism Quiz (APP > DC)
Koegel et al. (1996)	PRT n = 7 ITB n = 10 Age 3–9 (mean 6 years) DSM III R diagnosis of autism	**Pivotal response training vs. Individual target behaviour training of parents.** Pre- and post-measures were used (post- training measures were taken when parents reached 80% accuracy on the procedures). Pivotal response training (PRT) is based on a behaviour modification approach that encourages motivation and response to multiple cues. Individual target behaviour (ITB) is based on a series of discrete trials and the use of behaviour shaping and prompting. A manual was used to aid parents	**Interaction outcomes:** Observational measures of Happiness (PRT > ITB), Interest (PRT > ITB), low Stress (PRT > ITB), Communication style (PRT > ITB)

Table 1 *Continued*

Study	Participants	Design	Outcome measures and results
Schreibman *et al.* (1991)	PRT *n* = 12 ITB *n* = 12 Age 2.8–12.7 (mean 7.2 years)	Interventions as above. Only post-measures used. 19 parents participated. Five of the 19 parents took part in both conditions, thus 12 samples were available for each training group	**Parental outcomes:** Observer rating of affect: enthusiasm, interest, happiness – no useable data
Sherman *et al.* (1988)	Home-based *n* = 5 Residential *n* = 5 Outpatient *n* = 5 Age 2.6–7 (mean 5.1 years) Diagnostic measures unspecified	**A comparison of Home-based, Residential and Outpatient treatment.** Randomization stratified by level of functioning. Children assessed 2 months prior to intervention, during six month treatment, and at two month follow-up. Home-based treatment was intensive (6–8 hours, 5 days a week). An instructor worked with the child directly to increase skills and decrease maladaptive behaviour. Parents were trained in behaviour modification principles. Residential children remained in hospital 5–7 days a week, parents were not involved. The Outpatient programme was a less intensive version of the home-based programme (5–8 hours a week). Parents were actively involved in learning the principles of behaviour modification and direct instructor-child contact was minimal	**Child outcomes**: Behaviour observations; Hung Functional Behavioural Checklist; Developmental, Checklist – No useable data
Smith *et al.* (2000a)	ABA *n* = 15 PT *n* = 13 Mean age 36 months (between 18 and 42). Diagnostic measures unspecified	**Intensive home based Applied Behaviour Analysis vs. Parent training:** Random matched pair allocation based on diagnosis and IQ. Three-month pre-treatment measures, and a mean follow-up of 4 years and 10 months. ABA was based on Lovaas *et al.* (1981) treatment manual with 30 hours intervention per week for 2–3 years. In the Parent training group (PT) parents were taught the same treatment method. Parents received five hours instruction per week for three to nine months. Children spent 10–15 hours in a special education unit for the duration of parent training	**Child outcomes:** IQ (Bayley or Stanford-Binet) (ABA > PT), Merrill-Palmer (ABA > PT), Reynell (NS), Vineland Adaptive Behaviour Scales (NS), Child Behaviour Checklist (NS), lower parent rating of child workload (ABA > PT) **Parental outcomes:** lower parental stress (ABA > PT), higher parental rating of treatment quality (NS), lower parental workload (ABA > PT)

SCT, social communication training; RC, routine care; JAFPT, joint attention focussed patient training; ABA, applied behaviour analysis; NS, no significant difference; PRT, pitvotal response training; ITB, individual target behaviour training.

Table 2 Controlled studies of parent training for young children with autism spectrum disorder

Study	Participants	Design	Outcome measures and Results
Bristol *et al.* (1993)	Intervention *n* = 14 Control *n* = 14 Mean age 3.8 years. Diagnosis used the Childhood Autism Rating Scale	**TEACCH + Parent Training vs. TEACCH.** Measures were at pre-intervention, and at 6 and 18 month follow-ups. The Intervention group (PT) received TEACCH services with maternal psycho-educational treatment. Mothers in this group played an active role in the TEACCH service that their child was receiving, acting as co-therapists. Participants in the control group (CG) did not play an active role in the TEACCH service that their child was receiving	**Parental outcome:** Maternal Depression (Community Epidemiologic Depression Scale) lower (PT > CG)
Ozonoff & Cathcart (1998)	Intervention *n* = 11 Control *n* = 11 Age 31–69 (mean 53 months). Diagnostic approach unspecified, confirmed by the Childhood Autism Rating Scale	**TEACCH + Parent Training vs. TEACCH.** Measures were recorded at pre-intervention and an average of 16.5 week follow-up. Intervention group (PT) combined TEACCH services with a home parent training programme. Participants received 10 weekly one hour sessions and were asked to work with their child 30 minutes per day. Control participants received standard TEACCH services	**Child outcomes:** Psychoeducational Profile-Revised (imitation, perception, fine motor, gross-motor, eye-hand integration, cognitive performance, cognitive verbal) (NS)
Howlin (1981); Howlin *et al.* (1987); Holmes *et al.* (1982)	Intervention *n* = 16 Control (6 month) *n* = 14 Control (18 month) *n* = 16 Age 3–11 (mean 6.3 years). (Second control group mean age 5.2 years.)	**Parent Training vs. Routine Care.** Measures were made at pre-intervention, 6 months (matched controls group 1) and 18 months (matched controls group 2). Intervention group parents (PT) were trained at home in the use of behavioural techniques. Mothers were instructed to work with their children for 30 minutes per day. Participants in the control group(s) (RC) received intermittent outpatient treatment in addition to usual local routine care	**6 month child outcomes:** Language function (NS), Language level (NS) **6 month parental outcomes:** Changes in Mother's behaviour (NS), Changes in Mother's speech: info giving, praise, correct, direct (PT > RC), Changes in total utterances by mother (PT > RC), Mother's speech to child – interjections or incomprehensible (RC > PT) **6 month interaction outcomes:** direct observation of mother and child unoccupied (RC > PT), direct observation of mother and child playing (PT > RC), disruptive (PT > RC) **18 month follow-up child outcomes:** Parental interview: general behaviour problems at home (PT > RC) obsessions/rituals (PT > RC), levels of play (RC > PT); Child language use (NS), Child language level (NS), IQ scores (Merrill Palmer or WISC for older children) (NS). **18 month follow–up interaction outcomes:** Parental interview: response to parents (RC > PT), response to other adults (RC > PT), response to peers (RC > PT)

TEACCH, Treatment and Education of Austistic and Related Communication handicapped Children; NS, no significant difference; RC, routine care; WISC, Wechsler Intelligence Scale for Children.

the sample was recruited, how many individuals refused to participate and why, or the number of individuals who were excluded from the study and the reasons for this, were reported only in the two most recently published randomized controlled studies (Drew *et al.* 2002; Aldred *et al.* 2004). All the randomized studies had small sample sizes.

Controlled studies

Similar methodological weaknesses are apparent in the controlled studies. One study did not specify that diagnosis was made by an independent clinician (Ozonoff & Cathcart 1998). Furthermore, two studies did not use independent clinicians to carry out study assessments (Howlin *et al.* 1987; Bristol *et al.* 1993). Two studies did not report any follow-up data (Bristol *et al.* 1993; Ozonoff & Cathcart 1998). All studies reported some demographic information, with two reporting socio-economic status (Howlin *et al.* 1987; Bristol *et al.* 1993); however, the data were not compared directly to the local population, and again, very little information was reported concerning the numbers of people the study was offered to, nor how many refused or were excluded.

Child outcomes

The outcomes reported for children included direct testing of social-communication skills and IQ, and parent and teacher report of adaptive skills and problem behaviour.

The key outcomes for early intervention involve the areas of core impairment in autism. In the randomized study of Aldred *et al.* (2004), children in the parent training group had significantly lower ratings on the Autism Diagnostic Observation Schedule (ADOS) algorithm score for social-communication impairments ($P = 0.04$, MD −4.30, CI [−8.37, −0.23]), in comparison with the group receiving routine care. The ADOS social interaction algorithm score alone also showed a significant group difference ($P = 0.02$, MD −3.00, CI [−5.60, −0.40]). The authors comment that the ADOS communication score did not adequately reflect change because of how it is rated. However, effects of parent training have been shown in the communication domain in terms of reported child vocabulary. These effects just miss significance in the individual studies; however, when meta-analysis is applied to the MacArthur Communication Development Inventory (Drew *et al.* 2002; Aldred *et al.* 2004), significant effects are shown in favour of treatment on words understood ($P = 0.02$, WMD 75.84, CI [10.95, 140.72]) and on words said ($P = 0.04$, WMD 69.66, CI [2.39, 136.94]).

In the randomized study carried out by Smith *et al.* (2000a), children in the Intensive Applied Behaviour Analysis (ABA) group achieved a MD of 19.33, CI [3.7, 24.92] on IQ (Stanford-Binet Intelligence Scale, Thorndike *et al.* 1986, or Bayley Scales of Infant Development-Mental Development Index, Bayley 1969) in comparison with the Parent Training group. This result is statistically significant ($P = 0.02$), but more importantly it represents a clinical significant benefit, with almost one standard deviation of difference between the two groups. Children in the Intensive ABA group also scored significantly better on the Merrill-Palmer Scale of Mental Tests ($P = 0.04$); again, this level of difference can be interpreted as clinically significant (MD 15.16, CI [0.14, 30.18]). It was notable, however, that outcome reports of children's behaviour and skills, by both parents and teachers, did not differ between Intensive ABA and Parent Training groups. There were two significant treatment-related child outcomes reported in one of the controlled studies (Howlin *et al.* 1987). Parental reports of their children's general behaviour problems at home decreased significantly ($P = 0.005$, MD 6, CI [1.81, 10.19]), so did parental reports concerning child obsessions and rituals, in the parent training group at 18 months ($P = 0.0002$, MD 6, CI [2.89, 9.11]). However, at 18-month follow-up, levels of child play were reported by parents to be higher in the control group ($P = 0.04$, MD 7.30, CI [0.50, 14.10]).

Four significant generalization effects were found in one controlled study (Howlin *et al.* 1987). Parents in the control group rated their children higher in terms of their response to parents ($P = 0.0007$, MD 3, CI [1.26, 4.74]), to other adults ($P = 0.03$, MD 2.50, CI [0.27, 4.73]) and to peers ($P = 0.0001$, MD 2, CI [0.97, 3.03]).

Parental outcomes

Outcomes for parents included knowledge about autism and teaching strategies, observed communica-

tion behaviours when with their child, and stress levels.

There were four significant treatment-related parental outcomes reported from the RCTs. Mothers and child care workers in the parent training and child care group improved their knowledge concerning autism significantly, as measured by the TRE-ADD Autism Quiz (mothers, $P = 0.03$, MD 1.70, CI [0.19, 3.2]; child care workers, $P = 0.008$, MD 2.70, CI [0.70, 4.70]) (Jocelyn *et al.* 1998). However, this result does not suggest clear clinical significance for the improvement, that is, only an average 1 or 2 point difference on a 25-point questionnaire. Parental ratings of stress and workload during treatment were rated significantly lower in the Intensive ABA group compared with the Parent Training group (stress, $P = 0.008$, MD 1.52, CI [0.40, 2.64] workload, $P = 0.005$, MD 1.09, CI [0.47, 1.71], both rated on seven point scales) (Smith *et al.* 2000a). No differences were reported in the studies which utilized a well-validated instrument to measure stress, the Parenting Stress Index (Drew *et al.* 2002; Aldred *et al.* 2004).

In the controlled studies, it was found that mothers' communication behaviours such as information giving ($P = 0.0001$, MD 64.5, CI [31.75, 97.25]), praise ($P = 0.00002$, MD 24.6, CI [13.27, 35.93]), correct responses ($P = 0.007$, MD 10.3, CI [2.88, 17.72]), direct responses ($P = 0.002$, MD 29.9, CI [11.44, 48.36]), and the total number of maternal utterances ($P = 0.02$, MD 120.8, CI [15.85, 225.75]) were significantly greater in the parent training group at 6 months than in a control group (Howlin *et al.* 1987). Moreover, the number of interjections and incomprehensible utterances made by the mothers in the parent training group were fewer than those made by mothers in the control group ($P = 0.0003$, MD 0.94, CI [0.41, 1.47]) (Howlin *et al.* 1987). One generalization effect of treatment concerning parents was reported in the controlled studies: lower levels of maternal depression for mothers receiving parent training at 18 months ($P = 0.04$, MD 7, CI [0.49, 13.51]) (Bristol *et al.* 1993).

Interaction outcomes

The randomized study of Aldred *et al.* (2004) reported a significant difference in parents' observed interaction strategies with their child, in terms of greater parental synchrony ($P = 0.01$, MD 15.60, CI [3.19, 28.01]) in the intervention group than in the control. Koegel *et al.* (1996) found that the observed interaction outcomes of happiness ($P = 0.0006$, MD 0.94 CI [0.41, 1.47]), interest ($P = 0.007$, MD 1.28 CI [0.35, 2.21]), low stress ($P = 0.02$, MD 0.76, CI [0.14, 1.38]) and communication style ($P = 0.01$, MD 0.89, CI [0.21, 0.89]) were rated better for the Pivotal Response training group, than for the Individual Target Behaviour group. It should be noted that the MDs were very small in this latter study, bringing into question the clinical significance of these findings.

In one controlled study, it was found that the interaction between mothers and children during unoccupied periods was found to favour the control group ($P = 0.03$, MD 46.4, CI [5.48, 87.32]) (Howlin *et al.* 1987). However, results favoured the parent training group at play ($P = 0.007$, MD 72, CI [19.44, 125.56] which was associated with a reduction in disruptive behaviour ($P = 0.05$, MD 8.8, CI [0.09, 17.69]).

Discussion

This review aimed to establish the effectiveness of parent-implemented intervention for young children with ASD. The process has revealed the aim to be too ambitious. First, only recently have a few studies evaluated, in a randomized controlled design, the outcomes of parent training as compared with no training (i.e. local services as usual). Comparison with intensive intervention (which also includes a component of parent training), or between two different training approaches, does not in essence tell us about the effectiveness of the parents' involvement. Second, all of the studies included in this review have a number of important methodological shortcomings. In particular, none has a sufficiently large sample size to be able to attribute effects unambiguously to parent training. However, the evidence provided in this review has been treated in such a way as to reduce the probability of mis-attributing effect to confounding factors.

Intervention research generally, and that concerned with parent training for children with ASD can be perceived in terms of three questions relating to stages of knowledge from research evidence: Can the intervention work? Does the intervention work?

Does the intervention work in practice? (Charman *et al*. 2003)

There is sufficient evidence from the review that parent training *can* work in terms of observed improvements in children's social communication skills. This conclusion is further supported by the findings of a number of multiple baseline studies which suggest that parent training in the techniques of applied behaviour analysis produces some positive language (Harris *et al*. 1982; Laski *et al*. 1988; Smith *et al*. 2000b) and behaviour change (Koegel *et al*. 1978; Neef 1995; Lerman *et al*. 2000). Results of multiple baseline studies also indicate a positive effect for most parents, including increased parental knowledge (Harris *et al*. 1982), skills and performance (Koegel *et al*. 1978; Neef 1995; Lerman *et al*. 2000). Smith *et al*. (2000b) report that stress is reduced amongst parents as a result of parent training. In terms of family outcomes, parents involved in the Laski *et al*.'s (1988) study reported generalizing their new skills to other offspring.

Moreover, the review has suggested, from a small number of studies with and without random treatment allocation, that parent training *does* work to produce a positive effect on children's social communication behaviour, parental performance and parent child interactions. Parent training may also serve to reduce maternal depressive symptoms. However, the scope of current studies has been very limited; studies are either short-term, or report only outcomes for parents, or show mixed results for children. Furthermore, the mechanism of effect is not clear from current studies: for example, are children more likely to improve when their mothers have changed in interaction style or mood the most?

Finally, does parent-implemented intervention work *in practice*? This is the question of effectiveness of intervention, answered through studies that evaluate normal clinical practice. Studies of effectiveness must randomly allocate participants to the best available services vs. the new treatment, be tested on a large number of people in the real world, and be generalizable to the entire population (in question). Therefore, the design should involve a number of different sites. It should also provide pragmatic answers to real life questions such as cost in opportunity and money, and in terms of its acceptability to both professionals and families. None of the studies of early

intervention yet reported in the autism literature is an adequately designed study of effectiveness.

Parent training has been successfully applied within other areas of developmental disability and child mental health (e.g. Woolfenden *et al*. 2002). A systematic review of the effectiveness of group parent education programmes that aimed to improve behaviour problems in children up to the age of 3 years concluded that there is good evidence for positive change both in parental perceptions and in objective measures of children's behaviour (Barlow *et al*. 2002, 2005). Thus there is good reason to think that training of parents in specific skills may bring about some positive changes in for children who have ASD.

Implications for future research

Future studies of early intervention for children with ASD must have sample sizes large enough to generate a reasonable degree of statistical power, include a long-term follow-up assessment schedule of at least a year, and involve a full economic evaluation. Studies should make use of widely recognized standardized tools to assess outcome in terms of children's social and communication skills, and secondary behaviour problems, so as to enable others to assess the level of clinical significance. Research must be designed in such a way that a suitable degree of equipoise exists. Equipoise is the ability to offer a choice between two alternative interventions, where no fixed beliefs are held concerning their relative effectiveness by clinicians nor by participants. Equipoise is a necessary requirement for RCT methodology but is particularly difficult to achieve in research on young children with ASD (McConachie 2002).

The context in which parent-implemented intervention is evaluated in future must be carefully considered. Parent training is simply one component of early intervention for children with ASD (Le Couteur 2003). Autism is a complex and multifaceted disorder with a range of severity. Children place a range of demands on their parents, and families have varying capacity to respond to additional pressures such as active support of their children's educational programme. Parents need not only initial training in new skills but also on-going support as their children develop (Harris 1986a,b,c). Future evaluations must

consider what components of intervention may be combined in the most logical way to provide effective packages of care and supportive services for families.

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