

Living Guideline Supplementary Paper

**Aotearoa New Zealand Autism Guideline:**

Supplementary Paper on the effectiveness of parent-mediated approaches for autistic children

Marita Broadstock





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**Contributorship and independence**

*Marita Broadstock* (INSiGHT Research) conducted the umbrella review of systematic reviews, and prepared this Supplementary Paper. She manages the Autism/Takiwātanga Living Guideline programme; a process for ensuring the Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau (3rd edition, 2022) remains up to date.

An advisory panel known as the *Living Guideline Group* (LGG) considered the body of evidence presented in this paper in revising and grading Recommendations and Good Practice Points to update the Guideline. Their decisions are reported in [**Part 3**](#Part3).

Contributors have no financial or other perceived or real conflicts of interest pertaining to the reviewed material.

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**Statement of intent**

INSIGHT Research produces evidence-based best practice guidelines and systematic reviews to assist affected individuals, families/whānau, clinicians, educators, and policy-makers make decisions about the best supports available. The evidence is developed from systematic reviews of international literature and placed within an Aotearoa New Zealand context. Guidelines, including Supplementary Papers, are not intended to replace a clinical specialist/professional’s judgement in individual circumstances.

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This Supplementary Paper relating to parent-mediated approaches for autistic children and young people updates evidence for the Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau (2022) [1] and should be read in the context of the Guideline and previous Supplementary Papers [[2-14](#_ENREF_2)].

Currency review date: 2028

**Kāhore taku toa i te toa takitahi, he toa takitini** We cannot succeed without the support of those around us

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Executive Summary

This report supplements the Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau (‘the Guideline’) [[1](#Ref_1), [15](#_ENREF_15), [16](#_ENREF_16)] by providing an update on the effectiveness of parent-mediated approaches (‘parent-mediated interventions’) for children and young people on the autism spectrum.

Scope

The review considered the effectiveness of services, supports, accommodations, interventions, or programmes mediated by parents (as well as family members, whānau, or other caregivers) to address the needs of autistic people/tāngata whaitakiwātanga. In parent-mediated approaches, the professional specialist/therapist is involved in training the parent or family member in how to deliver the approach/therapy/programme, as well as having an ongoing role in coaching and mentoring them.

Method

A broad search strategy was undertaken of 10 bibliographic, health technology assessment, and guideline databases to identify peer reviewed studies published between January 1, 2004 and 12 July 2022. Including reports identified through citation searching of the bibliographies of retrieved articles, 1595 unique abstracts were considered.

After applying inclusion and exclusion criteria, a ‘best evidence’ approach was employed to restrict eligible studies. This led to including only peer reviewed secondary research studies (systematic reviews and meta analyses) published since 2013, the year a Cochrane meta-analysis was published on the topic [[17](#_ENREF_17)]. Secondary reviews were excluded if they did not have at least one original eligible primary study not already included by another secondary review.

The ‘umbrella review’ included eight systematic reviews, five of which also included meta-analyses. These were critically appraised for quality using a formal checklist for secondary studies, and synthesised narratively and in tables.

No New-Zealand based research was identified that met inclusion criteria for ‘best evidence’ for assessing the effectiveness of these approaches. However, four qualitative and survey-based research studies (reported in three papers) relating to the clinical relevance and applicability of these approaches to Aotearoa were identified and summarised.

Review findings

* These results suggest that there is emerging evidence for small improvements evident for language as a broad outcome, with moderate improvement in expressive language found for children receiving parent-mediated approaches.
* For young children receiving parent-mediated approaches, there is evidence of no or small improvement in joint attention and initiation, and some small improvement in general measures of social communication/socialisation. A review including young children and adolescents found moderate improvement in social skills, and a large improvement in engagement.
* Two reviews suggest moderate to large improvement in the parent-child relationship for recipients of parent-mediated approaches.
* There is reasonable evidence of small to moderate improvement in autism (core) characteristics, and repetitive behaviour. Meta-analyses suggest consistent evidence of moderately large effects of parent-mediated approaches being associated with reduced maladaptive behaviour, and some evidence of moderate improvement in child distress and self-regulation.
* There may be a small improvement in the adaptive functioning of autistic children and young people, based on parent ratings.
* There was no evidence that parent-mediated approaches improved visual reception, or motor skills outcomes. However, there is emerging evidence of a small improvement of parent-mediated approaches in autistic children’s cognition.
* There is consistent evidence from a good number of studies across different reviews that parent-mediated approaches can lead to modest positive benefits for parents, with small improvements in parents’ levels of stress, distress, and self-efficacy.
* Factors that may predict, mediate or moderate parent-mediated intervention effectiveness were investigated in sub-group analyses and suggest no clear or consistent patterns. It is not currently possible to offer specific recommendations about the most successful and necessary components and intensity of approaches delivered by parents and family members to autistic children and young people.
* Four qualitative studies (reported in three papers) conducted in New Zealand illustrate how parent-mediated approaches have been successfully implemented and positively received in Aotearoa. Alignment of approaches with a parent’s parenting style, culture, and connection with the agency/trainer/therapist appeared to be important factors in acceptability. The research suggests ways programmes can be tailored to the needs of the community, including relationship-building, targeted promotion, addressing practical barriers to participation, and employing culturally-specific adaptations to align with social norms and language.

Conclusions

Parent–mediated approaches are a source of intense and growing research interest over the last decade. Improving the relationship between the parent and child may lead the parent to understand their child’s needs better, supporting them to communicate more effectively and reduce behaviours of concern, and as a consequence, reducing the distress for both parent and child.

Limitations of the research, gaps in understanding, and recommendations for future research are discussed.

Revisions to the Guideline based on the review

The Living Guideline Group (LGG), an expert advisory panel, presented their decisions on the implications of the updated body of evidence for the guideline. Their revised and new recommendations and Good Practice Points supplement the guideline on this topic are presented in the [Summary Table](#SummTable). The Preamble, additional text and rationale are presented in [Part 3](#Part3).

Summary Table: Revised and new Recommendation and Good Practice Point

|  |  |  |
| --- | --- | --- |
| Reference | Revised recommendation | Grade |
| Rec 2.1.5a | Parent-mediated approaches (also known as ‘parent mediated interventions’) should be considered by, and provided for, parents/caregivers of autistic children. There is strong evidence that parent-mediated approaches improve the relationship between children and their parents/caregivers, and provide a range of benefits. | A |
| GPP 2.1.5b | The design of parent-mediated approaches (also known as ‘parent mediated interventions’) should be culturally responsive, whānau-led, holistic, mana-enhancing, respectful, easy to use, and tailored to the autistic individual and whānau needs. | ✓ |
| GPP 2.1.5c | Parents/caregivers should determine in what way and how much they are involved in the delivery of supports. | ✓ |
| GPP 2.1.5d | Practitioners should be aware that parents/caregivers may perceive that their involvement in the delivery of supports detracts from their natural parenting/caregiving. | ✓ |

Note: Grades indicate the strength of the supporting evidence rather than the importance of the evidence. Grade A indicates good evidence, B is fair evidence, C is international expert consensus, and I is insufficient, poor quality, or conflicting evidence. See [Table A1.3](#A1_3) in Appendix 1 for detail.

Preamble

Living Guideline process

The first edition of the New Zealand Autism Spectrum Disorder Guideline (as it was then called) was published in April 2008 [[15](#_ENREF_15)]. As part of their commitment to its implementation, New Zealand’s Ministry of Health and Ministry of Education agreed to establish a ‘Living Guideline process’ in 2009. This process aims to ensure that the Guideline is regularly updated and refined to reflect new research and changing user needs.

Every year, the Living Guideline process produces an update of the Guideline on a topic of high priority. These are published as Supplementary Papers, with thirteen completed to date [[2-14](#_ENREF_2)].

Revisions to Guideline Recommendations are directed by an advisory panel called the Living Guideline Group (LGG). The panel includes members with lived experience, clinicians, researchers, educationalists, and community service providers, as well as ex-officio representation by the funders: Whaikaha – Ministry of Disabled People and the Ministry of Education (the current membership is listed in [Appendix 1.1](#A1_1)). The Living Guideline process is directed by the Autism/Takiwātanga Guideline Manager (Marita Broadstock, INSiGHT Research) who prepares the Supplementary Papers and revises the Guideline.

Each year, the Living Guideline Group are responsible for identifying and selecting a topic for update. A systematic literature review is then undertaken by the Autism/Takiwātanga Living Guideline Manager. A draft of the review (representing [Part 1](#Part1) and [Part 2](#Part2) of the Supplementary Paper) is considered by the LGG who assess the body of evidence with respect to its quality, quantity, consistency, applicability, and relevance. The LGG use this assessment as a basis for revising and developing guideline new Recommendations and Good Practice Points. These decisions are presented, accompanied by the LGG’s rationale and additional notes, in [Part 3](#Part3).

A second edition of the Guideline was published in 2016 [[16](#_ENREF_16)]. The current 3rd edition, now titled ‘Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau’, was published in 2022 [[1](#Ref_1)]. New editions of the Guideline incorporate revised and new Recommendations from completed Supplementary Papers alongside other revisions.

The current Supplementary Paper should be read in the context of the Guideline and previous Supplementary Papers.

Full details of review methods including search strategies, appraisal of study quality and data extraction are presented in [Appendix 1](#App1).

[Appendix 2](#App2) presents relevant abbreviations, acronyms, and [glossaries](#Glossary).

[Appendix 3](#App3) presents evidence tables of appraised studies.

Autistic preferences around terminology

In this paper and in the Guideline, the terms ‘autistic person’ and ‘person on the autism spectrum’ are used to refer to someone understood to meet criteria for the diagnosis of Autism Spectrum Disorder. This reflects that increasingly people in the autistic community prefer to use identify-first language by referring to themselves as autistic rather than ‘having autism’. This recognises autism as a central part of one’s identity – of who one is, rather than as something separate to oneself, that can be put aside [[18](#_ENREF_18)].

Many autistic people are also uncomfortable with the acronym ASD. This is because the word ‘disorder’ conveys a sense of autism as a pathological impairment rather than a reflection of neurodiversity [[19](#_ENREF_19)]. Here, the acronym ASD is therefore only used when referring to a person’s clinical diagnosis or diagnostic tools or services.

The term ‘high functioning’ is sometimes used by researchers to define people with higher cognitive functioning either as established by intelligence tests (generally indicated by full-scale IQ scores of 70 or above), or through the diagnosis of ‘high-functioning autism’ or Asperger syndrome (under DSM-IV criteria) [[20](#_ENREF_20)]. In general, the terms ‘high functioning’ and ‘low functioning’ to describe groups of autistic people are considered unhelpful and divisive by many on the autism spectrum [[21](#Ref_21)]. Some people may be described as having high and complex needs, and others as having lower or less obvious support needs.

The terms ‘problem behaviour’ and ‘challenging behaviour’ are avoided in the Guideline and its supplements. The use of ‘problem behaviour’ can be perceived as implying that the autistic individual is deliberately doing something wrong or ‘being naughty’ or ‘difficult’. However the behaviour is the problem, not the person, and the challenge lies in how to support them. The behaviour may be a concern to the autistic individual themselves (such as self-harm) or it may only be a concern to others who do not understand its purpose (e.g., shutdown from sensory overload). Addressing this concern therefore does not necessarily imply the need to eliminate or replace the behaviour. In specific situations, where possible, the ‘behaviour of concern’ should instead be described with respect to the autistic person’s experience (e.g., sensory overload, stimming, expression of distress).

Other problematic terms refer to autistic people who can reliably use speech to communicate as being ‘verbal’, and those who do not as being ‘non-verbal’. However, verbal language is not just speech and includes spoken and written words, signs or visual codes. To be more inclusive, alternative terms include ‘minimally speaking’ or having ‘complex communication needs (CCN)’. In the Guideline, people described as ‘having speech’ are those who can consistently rely on speech for functional communication. Note that non-verbal means something different when describing non-symbolic communication such as body language, facial expressions, tone of voice, and eye gaze to convey meaning.

It is understood that language preferences may continue to evolve. Fundamentally, autistic individuals have the right to self-refer and be referred to as they choose. It’s always best to ask a person what terms work for them, based on their own lived experiences and identity.

Examples of other language changes aimed at de-pathologising the condition include replacing the term ‘comorbidity’ with ‘co-occurring’; ‘normal’ with ‘non-autistic’; ‘symptoms’ with ‘characteristics’; and the terms ‘impairments’ and ‘deficits’ with ‘challenges’ and ‘difficulties’. Terms relating to ‘treatment’, and ‘management’ are generally avoided, especially where they are used in relation to the nature and expression of autism itself. Instead, they have been replaced by ‘supports’, and ‘approaches’.

Māori perspectives

Takiwātanga is a Māori word for autism. This term was coined by Keri Opai after consultation with tāngata whaitakiwātanga (autistic people). It means ‘in my/their/his/her own time and space’ and, as Opai notes, it reflects “a positive, Māori worldview aspect of autism [[22](#_ENREF_22)].”

‘Tangata whaitakiwātanga’ refers to an autistic person

‘Tāngata whaitakiwātanga’ refers to autistic people

Māori cultural concepts and values not only determine how takiwātanga is perceived but also attitudes towards it and how autistic whānau should be supported. Sir Mason Durie (1984) [[23](#_ENREF_23)] provides a helpful framework to guide services and programmes for tāngata whaitakiwātanga and their whānau. Durie’s Whare Tapa Whā model lists four dimensions of wellbeing for Māori. These are: taha tinana (physical wellbeing); taha hinengaro (mental wellbeing); taha wairua (spiritual wellbeing) and taha whānau (family wellbeing). Consequently, in order to be culturally responsive, provisions for tāngata whaitakiwātanga need to incorporate these four dimensions.

A framework that complements Te Whare Tapa Whā is Ka Hikitia’s Outcome Framework[[1]](#footnote-1), Aotearoa New Zealand’s Māori Education Strategy. This is based on extensive consultation and is focused on achieving the following ‘excellent and equitable outcomes’ for Māori:

* Te Whānau: education provision responds to Māori within the context of their whānau
* Te Tangata: Māori are free from racism, discrimination and stigma
* Te Kanorautanga: Māori are diverse and need to be understood in the context of their diverse aspirations and lived experiences
* Te Tuakiritanga: Identity, language and culture matter for Māori
* Te Rangatiratanga: Māori exercise their authority and agency.

Tiriti o Waitangi/Treaty of Waitangi

The Guideline and its Supplementary Papers acknowledge and uphold the principles of Te Tiriti o Waitangi/Treaty of Waitangi. It considers the Treaty principles of partnership, participation and protection central to improving health and education outcomes for Māori.

Consistent with Whāia Te Ao Mārama 2018–2022: The Māori Disability Action Plan [[24](#_ENREF_24)], the Guideline and this Supplementary Paper seek to advance practices and services for tāngata whaitakiwātangaMāori that upholds the significance of te reo Māori, te ao Māori (the Māori world), and ensures access to Māori approaches to practice.

This vision sees tāngata whaitakiwātangaMāori having leadership, choice and control over the supports which enable them to thrive, flourish and live the life they want.

Target audience

The systematic review presented in [Part 1](#Part1) and [Part 2](#Part2) aim primarily to provide an updated synthesis of research evidence on a specific topic for consideration by the Living Guideline Group. As such it is written in an academic style and is not intended for the general reader, though will also be of interest to researchers and those wishing to consider the original research in greater depth.

The Living Guideline Group’s decisions regarding the systematic review’s implications for revising and developing new Recommendations and Good Practice Points (which update the Guideline) are presented in [Part 3](#Part3). This section is intended for a broader audience, including tāngata whaitakiwātanga, their families/whānau, and the providers of professional clinical, education and support services for New Zealanders on the autism spectrum, as well as policy makers and funders.

Part 1: Introduction

1.1 Background

Advantages and disadvantages of parent-mediated approaches

Access for autistic children to approaches, therapies, programmes and professional therapists can be difficult, particularly for supports requiring intensive one-to-one time with a specialist. Services can come at a high cost, which is a key barrier in low-resource settings and for those without insurance or government cover [[25](#_ENREF_25)]. There may also be a lack of local availability, and long waiting lists. In addition, practical challenges of attending clinics outside the home present themselves, including demands of time, inconvenience, transport, and coordination of care needs, particularly for people living in remote or rural areas [[26](#_ENREF_26)].

Alternative approaches which aim to address these barriers include ‘parent-mediated interventions’ (PMIs). These are where parents (or other family members including primary caregivers or siblings) are trained by a specialist to deliver an approach directly to the child. In addition to the practical and economic advantages, parents are arguably ideally suited to the provision of supports to their children. They already spend significant time with their children and likely are to be the most attuned to and invested in their child’s development. A programme which is led by parents aims to harness such intrinsic insight and motivation [[17](#_ENREF_17)].

Placing the approach/programme within a home setting also has the potential to facilitate an intensive, naturalistic, and tailored approach [[27](#_ENREF_27)]. The home environment may also be more ecologically valid, providing incidental opportunities in everyday life to learn and practice new skills. There is also the opportunity to transfer skills across tasks/settings and imbed them over time, promoting generalisation and maintenance [[28](#_ENREF_28)]. Having a parent or caregiver providing the support also provides consistency of approach over time as the child grows and develops [[29](#_ENREF_29)].

There may also be benefits to the wider family. Providing parents with skills to support their autistic child’s needs may offer them a sense of empowerment [[27](#_ENREF_27)], with the potential to enhance their sense of competence, reduce their stress, and contribute to family cohesion [[30](#_ENREF_30)].

However, the time, training and effort required of the parent are not ‘cost-free’. The ability of the parent or family member to be trained in and adept at delivering an approach well is likely to vary across individuals. As discussed in literature reviewed by Shalev et al., (2020) [[28](#_ENREF_28)], parents of children on the autism spectrum tend to have increased stress, and are at high risk of mental health concerns including anxiety and depression, compared to parents of children without disabilities. Common genetic profiles may also be reflected in traits of neurodiversity in the wider family that may impact on members’ delivery of an approach/programme and assessment of progress in unpredictable ways.

Defining parent-mediated approaches

Parent-mediated approaches have been defined inconsistently in the autism literature. Bearss et al., (2015) [[31](#_ENREF_31)] suggested a taxonomy that differentiates *skill-focused parent-implemented* approachesfrom *knowledge-focused parent support* programmes. The current review is concerned with the former. Skill-focused programmes are where the parent or family member takes on the role of specialist/therapist in delivering the approach/programme to their autistic child. Importantly, the parent is the agent of change, and the child is the direct beneficiary of the approach, rather than indirectly as is the case with psychoeducational programmes and care coordination [[31](#_ENREF_31)].

In parent-mediated approaches, the professional specialist/therapist is involved in training the parent or family member in how to deliver the approach/programme, as well as having an ongoing role in coaching and mentoring them. Key components include:

* modeling of strategies
* parent coaching
* provision of feedback, and
* individualization of strategies [[32](#_ENREF_32)].

Parent-mediated approaches include comprehensive programmes for global improvement (e.g., of ‘adaptive functioning’), or target specific areas/skills (e.g., joint attention, language, play) or areas of concern (e.g., disruptive behaviour, insomnia, feeding, toileting). They can also seek to improve quality of life and well-being for the autistic individual, as well as for parents and the wider family unit [[33](#_ENREF_33)].

Training of parents/family members can be undertaken in a clinic or community centre, home, or through distance learning (e.g., tele-health). Delivery of training can be through interactive digital technologies including video-conferencing, interactive websites, and apps [[29](#_ENREF_29)]. However, evaluations of how best to deliver the training itself are not the focus of the current review.

1.2 Parent-mediated approaches in the Guideline

There are currently no Recommendations or Good Practice Points in the Guideline [[1](#Ref_1), [15](#_ENREF_15), [16](#_ENREF_16)] relating specifically to parent or caregiver-mediated supports. There is brief mention of them in text within ‘Part 2, 2.1.a Parents and full-time carers’, under ‘Parent-professional collaboration’:

Observational research acknowledges the important role that parents have in any intervention process, but specific guidance is lacking on the potential advantages and disadvantages of different parent-mediated approaches to providing early intervention. A Cochrane systematic review [[34](#_ENREF_34)], including two small randomised controlled trials (RCTs), was unable to offer guidance for practice from its findings.

The Cochrane review [[34](#_ENREF_34)] cited in the Guideline has been withdrawn and replaced by Oono et al., (2013) [[17](#_ENREF_17)], included in the current review. It considered parent-mediated 'interventions’ evaluated in randomised controlled trials published between 2002 and 2012 for young children (aged 1 to 7 years).

In addition, Good Practice Point 4.3.10a, developed by the LGG and reported in the relevant Supplementary Paper,[8] lists a range of recommended adaptations to cognitive behaviour therapy. The final adaptation listed is: “Involve a **support person**, such as a family member, partner, carer or key worker (if the autistic person agrees) as a co-therapist to improve generalisation of skills learned within sessions.”

1.3 Aotearoa New Zealand research

Qualitative studies and survey-based research were identified which assessed the first-hand experiences of New Zealand-based recipients of parent-mediated approaches (see [Appendix A1.2](#A1_3_NZ) for the methodology).

Studies were limited to those reporting on the relevance, applicability, acceptability and feasibility of these approaches within a New Zealand culture and service context, including where these reflect user preferences and values. (Studies evaluating effectiveness were considered for the systematic review described in [Part 2](#Part2)).

Of the 131 abstracts identified by the search strategy, three papers met inclusion criteria reporting on four separate studies. The earliest study by Birkin et al., (2008) [[35](#_ENREF_35)] considered the EarlyBird program, an early intervention program for parents of autistic children which taught parents how to employ behavioural techniques aimed to increase their child's communication and manage behaviours of concern. To explore accessibility, 77 caregivers of autistic children (aged <five years) who were eligible for the programme were interviewed, with only 15% reporting having received it. Factors associated with non-participation included: lack of awareness and connection with the agency administering the program; being from a minority ethnic group; length of wait time and geographical barriers.

A second study in the same report [[35](#_ENREF_35)] explored barriers to uptake through semi-structured interviews of 12 Māori, Pasifika and Asian parents of autistic children in New Zealand. Themes identified included: lack of awareness of autism; stigma for the family attached to autism; shyness and anxiety around group discussion and videotaping; and competing demands of work for low-income earning parents. Ways to improve access included the need for relationship-building and trust prior to a programme being offered; and offering the programme in the parent’s first language. Suggestions to address barriers include more targeted promotion through culture-specific networks; running smaller programmes more often in rural areas; and employing culturally-specific adaptations.

A qualitative study by Pretorius, Clendon, and McLaughlin (2020) [[36](#_ENREF_36)] considered the experience of three parents of young autistic preschool-aged children after receiving training and coaching in a parent-implemented intervention. A Speech-Language Therapist taught parents interaction promoting strategies and embedded naturalistic interaction practices in 4 group workshops and 8 in-home coaching sessions. Recorded open-ended interviews with participants were transcribed and themes identified. Parents found the approach acceptable, and valued the personalised nature of developing communication goals for their child. The group workshops were found to be initially overwhelming and complicated. However the addition of individualised coaching was highly valued as it provided opportunities for planning, reflection and problem-solving of ways to embed naturalistic interaction practices into everyday activities and routines. Families reported feeling less stressed and more confident alongside improvements in child-related outcomes.

Finally, a study by Waddington, van der Meer, Sigafoos and Bowden (2020) [[37](#_ENREF_37)] considered parents’ perceptions of a 12-week home-based training programme for delivering the Early Start Denver Model (ESDM). The ESDM is a naturalistic developmental behavioural intervention for young autistic children aged under five years. Mothers of five young autistic boys (two NZ European, one Indian, one Asian, and one Māori) completed the Treatment Acceptability Rating Scale-Revised (TARF-R). All reported positive ratings for programme acceptability. During the semi-structured interviews, parents favoured the simplicity and flexibility of the play-based approach. Some mothers found it hard to find time to remember to use the techniques, and read the manual. There was a desire for other family members and professionals involved in the training. Whilst being home-based was convenient for the training, it could be disruptive, and some said it would be helpful to have it elsewhere. Personal qualities of the trainer (flexible, practical, non-judgemental) and their relationship with the child were highly valued. The authors suggest that the mothers’ overall perceptions of the intervention were influenced by whether their child improved on outcomes, alignment of parenting style with the approach, and their relationship with the therapist.

1.4 The current review update

Scope

Since the Cochrane review in 2013 [[17](#_ENREF_17)], there has been a rapid growth in research examining the effectiveness of ‘parent-mediated interventions’ across a broad range of participants, approaches and outcomes. Scoping of the literature identified several good quality systematic reviews and meta analyses published over this time which led to the Living Guideline Group prioritising this topic for update.

An ‘umbrella review’ was undertaken of peer reviewed secondary research studies (systematic reviews and meta analyses) where they investigated the effectiveness of parent-mediated approaches/'interventions' for autistic children and young people.

Umbrella reviews aim to provide a summary of existing research syntheses relating to a topic, rather than a re-synthesis of primary studies. This approach allows assessment of whether review teams addressing similar questions independently observe similar results and arrive at similar conclusions [[38](#_ENREF_38)]. Umbrella reviews also allow broadly scoped topics to be considered, comparing and contrasting the findings from a range of reviews with varying selection criteria. This permits consideration of the impact of different features of the approach/programme, its delivery, the population, and target outcomes on effectiveness, and to determine areas of consistency and discrepancy.

Aims

The specific aims of this Supplementary Paper were to:

* systematically identify, select, and synthesise secondary research studies (systematic reviews and meta analyses) that evaluate the effectiveness of parent-mediated approaches in leading to desired outcomes for autistic children and young people, and explore the characteristics of effective supports
* consider this evidence as it supplements the Guideline [[1](#Ref_1), [15](#_ENREF_15), [16](#_ENREF_16)] in order to inform the LGG’s revision of any existing relevant Recommendations/Good Practice Points and/or the development of new ones.

Part 2: Systematic review

This chapter describes the findings of an umbrella systematic review (review of systematic reviews) relating to parent-mediated approaches/programmes for autistic people.

2.1 Method

Identification of studies

A broad and inclusive systematic search was undertaken on July 12 2022 using a combination of terms for autism, intervention, and systematic reviews (see [Appendix 1](#App1) for the full search strategy). Titles, abstracts and subject fields of 10 bibliographic, health technology assessment, and guideline databases were searched. Results were initially limited to those published in the English language; however, the search strategy was run again for non-English publications and identified only 8 reports, none of which met selection criteria for the current review.

Where database limits permitted, publications were restricted to those involving human participants and peer reviewed journals. Hand searching of journals and contacting of authors for unpublished research were not undertaken.

To identify additional eligible studies, bibliographies of retrieved publications and recent narrative reviews were also examined. This led to 1595 unique abstracts being identified, after removal of duplicates. Guidelines from the Preferred Reporting Items for Systematic Review and Meta-Analyses statement (PRISMA) [[39](#_ENREF_39)] were employed for the screening and selection process. The PRISMA flowchart is presented in [Figure 1](#fig1).

Study selection

Studies were selected if they met predefined inclusion criteria which were structured around parameters of the PICO framework of Population, Intervention, Comparators, and Outcomes (see [Table 2.1](#Table2_1)). Papers were initially screened based on their title and abstract, and then potentially eligible papers were retrieved as full text and assessed for eligibility. As a paper may be excluded for multiple reasons, reasons for exclusion were identified sequentially from a list ordered as follows: wrong study design, wrong population, wrong comparator, wrong scope/intervention, wrong outcomes, and to a final set, wrong date of publication. For each excluded paper, the first criteria that applied was recorded as the reason for exclusion, with results presented in [Figure 1](#fig1).

Figure 1: Overall flowchart of articles screened

**Identification of studies via databases and other sources**

Records identified from:

Databases (n = 2090):

Medline (n = 649)

CINAHL\* (n = 39)

PsycINFO (n = 478)

PsycARTICLES (n = 12)

SocINDEX (n = 38)

ERIC (n = 182)

Education Research Complete (n = 240)

NZresearch.co.nz (n = 1)

EMBASE\* (n = 432)

Cochrane Database of Systematic Reviews (n = 19)

Citation searching (n=3)

Duplicate records removed *before screening*:

(n = 498)

**Identification**

Records screened

(n = 1595)

Records excluded

(n = 1523)

Reports excluded:

Wrong Study Design (n = 11)

Wrong Population (n = 10)

Wrong Scope/Intervention (n = 27)

Wrong Outcomes (n = 6)

Retrieved reports

(n = 72)

**Screening**

Reports excluded:

Published < 2013 (n = 6)

Superseded, no new studies (n = 4)

First set of studies meeting selection criteria

(n = 18)

Final set of studies published since 2013 included in review

(n = 8)

**Included**

\* searches excluded Medline records

Table 2.1: Inclusion and exclusion criteria for selecting eligible studies

|  |  |
| --- | --- |
| Characteristic | Inclusion criteria |
| Participants | Parents, siblings, guardians, primary caregivers, or other family/whānau members of children/young people aged 2-18 years diagnosed with ASD |
| Intervention | Supports, programmes, services, or accommodations offered to address the needs of autistic people  Mediated by a parent, sibling, guardian, primary caregiver or other family/whānau member |
| Comparator | No intervention, ‘treatment’ as usual, waiting list, alternative child-centred intervention not mediated by parents, or alternative parent-mediated intervention of hypothesised lesser effect than the experimental condition. |
| Outcomes | At least one primary outcome was required for eligibility  Primary outcomes (relating to the autistic individual):   * Global scales of clinical improvement * Adaptive functioning * Communication * Socialisation * Play * Behaviour * Cognition * Skills training * Mental health and well-being   Secondary outcomes:   * Family/whānau quality of life (e.g., self-efficacy, confidence, stress) * Support’s perceived helpfulness, satisfaction, preference |
| Study Design | Peer reviewed systematic reviews and/or meta-analyses of relevant scope including at least one eligible RCT (i.e., level II study) |
| Publication date | Initially, eligible reports were published between 1 January, 2004 and 12 July, 2022 (see ‘superseded evidence’ exclusion) |
| Characteristic | Exclusion criteria |
| Excluded publication type | The following were excluded:   * dissertations, book chapters, poster presentations, abstract-only reports, narrative reviews, unpublished data, correspondence, editorials, commentaries |
| Excluded scope | Studies which were not deemed relevant to the research question or nature of the review were excluded, including if they:   * concerned parent support programmes * concerned parent education or parent training programmes which predominantly provided psychoeducation * concerned evaluated delivery of parent training/coaching * concerned the development of an intervention/approach or outcome measure/s * reported on studies comparing autistic people with non-autistic people * reported on studies solely conducted in the developing world * were animal, prenatal, genetic, brain, biomarker, or pharmacological studies |
| Superseded evidence | Limited to ‘best evidence’, excluded secondary reviews published prior to 1 January, 2013, or which included no ‘original’ primary studies (i.e., studies not already included in other eligible reviews) |

Participants

The study population were parents, siblings, guardians, primary caregivers, or other family/whānau of autistic children aged 2-18 years. Autistic children were those who have received a clinical diagnosis of ASD, or (for no fewer than 20% of the sample) self-identify as autistic in the absence of a formal diagnosis.

Intervention

Studies were included where:

* they evaluated supports, programmes, services, accommodations, or other interventions which aimed to address the specific needs of autistic children or young people aged under 18 years
* the evaluated supports were mediated by a parent or family member who had been trained by a professional to act as the primary specialist/therapist delivering the intervention, and who received ongoing mentoring/coaching/feedback from the trainer.

Studies were within scope where they reported the effectiveness of an eligible approach/programme as a stated aim or as a significant and clearly delineated component of the results.

Comparator

For comparative studies, the comparator could be a control condition including no intervention, usual/standard care, or waiting list control. Or the comparator could be an active control group involving an alternative child-centred intervention not mediated by parents, or an alternative parent-mediated intervention hypothesised to provide lesser effect than the experimental condition (e.g., lower intensity, lower dose, group versus individual parent training).

Outcomes

Included studies needed to report on at least one quantifiable measure of effectiveness listed as a primary outcome (all of which relate to the autistic child or young person). Where a study met this criteria, secondary outcomes were also reported.

Primary outcomes:

Note: Thefollowing outcomes were identified in scoping as commonly included in evaluation studies of parent-mediated approaches. However, their inclusion should not be taken to imply endorsement. The ethical choice of outcomes in research relating to autistic individuals is discussed under [2.4](#Sec2_3): Limitations.

* Autism (core) characteristics (the degree of social communicative impairments and restricted repetitive behaviors that are specific to ASD)
* Adaptive functioning (eg. daily living skills, adaptability, functional communication, independent functioning)
* Communication/language (the ability to produce and comprehend spoken and/or written language where age-appropriate, or display play skills that are known precursors of language; e.g., language, receptive language, expressive language, nonverbal communication, joint attention, engagement, initiation and response to interaction)
* Play (e.g., play skills, mixed for functional or symbolic play
* Socialisation (the ability to engage in social interactions, to demonstrate social awareness of others’ perspectives, and to use social aspects of communication such as pragmatic language at an age-appropriate level; e.g., social engagement, social skills). Pragmatic language measures (e.g. Early Social Communication Scales) were included under socialisation as they assess for communicative behaviours that are required for social interactions, such as turn-taking and responses to social games
* Behaviour (e.g., irritability, noncompliance, disruptiveness, hyperactivity, aggression)
* Cognition (the ability to acquire, retain, attend to, problem-solve, and reason information using concrete objects, abstract ideas, and verbal information at an age-appropriate level)
* Physical (motor skills, vision, sleep onset)
* Daily Living Skills (age-appropriate self-care activities including feeding, dressing, toileting, hygiene, safety behaviour)
* Mental health, emotion, and well-being (e.g., depression, anxiety, stress)

Secondary outcomes:

* Parent/family member’s quality of life (e.g., well-being, self-efficacy, confidence, stress
* Support’s perceived helpfulness, satisfaction, preference (of autistic individual or parent/family member)

Study designs

The approach taken was to consider ‘best evidence’; that is, include studies representing the highest level of evidence first, and only in their absence, consider lower order evidence.

The NHMRC evidence hierarchy [[40](#_ENREF_40)] ranks studies in terms of quality based on their study design (see [Table A1.1](#TableA1_1) in Appendix 1). The most robust level of evidence is Level I which consists of systematic reviews (which may include a meta-analysis) that include at least one Level II study, a randomised controlled trial [[41](#_ENREF_41)]. Further details on levels of evidence and specific criteria for defining a systematic review are presented in [Appendix 1.3.](#A1_3)

Guided by this hierarchy and the principal of best evidence, an ‘umbrella review’ was undertaken of peer reviewed secondary research studies (systematic reviews and meta analyses) where they investigated primary studies assessing effectiveness of parent-mediated approaches for autistic children and young people, and included at least one randomised controlled trial.

Exclusions

Publications were excluded if they were:

* Dissertations, book chapters, conference proceedings
* Poster presentations, abstract only reports, unpublished data
* Narrative reviews, editorials, commentaries
* Grey literature including book reviews, news reports, trade magazine articles, blogs
* Not deemed relevant to the research question or nature of the review, including studies that:
* concerned parent support programmes, parent education or parent training programmes which predominantly provided psychoeducation
* concerned evaluated delivery of parent training/coaching
* concerned the development of an intervention or outcome measure/s
* reported on studies comparing autistic people with non-autistic people
* compared autistic people with non-autistic people
* were predominantly conducted in the developing world
* were animal studies, prenatal studies, genetic studies, brain studies, biomarker studies, pharmacological studies.

In the current umbrella review, systematic reviews of parent education and training programmes were excluded. However it is acknowledged that some reviews [[17](#_ENREF_17)] employed a broader definition of parent-mediated approaches.

Superseded evidence

The Living Guideline process aims to update publications from the date the original searches were completed for the first edition of the Guideline [[15](#_ENREF_15)]: 1 January 2004. The initial search therefore included reports published between 2004 and 12 July 2022, the date searches were run.

Once selection criteria were applied to retrieved reports to provide the first set of reports, a final set was limited with two additional exclusion criteria to identify the ‘best evidence’.

First, publications were limited to those published from 2013. This date was selected because it is the year the Oono et al., (2013) [[17](#_ENREF_17)] Cochrane review of parent-mediated early interventions was published, which updates the review referred to in the Guideline’s first edition [[15](#_ENREF_15)]. Considering secondary research within the past 5–10 years is likely to reflect original/primary research conducted over the previous 30 years synthesised [[38](#_ENREF_38)].

Second, secondary reviews which included no original primary studies were also excluded. That is, without a randomised controlled trial (level II evidence) meeting selection criteria for the current review not included in a more recently published secondary review. This aimed to reduce redundancy of superseded material.

Critical appraisal of included studies

An evidence table template was employed to record pre-defined information (see [Appendix 1.4](#A1_4)) to extract data from each included report ([Appendix 3](#App3)).

The quality of included systematic reviews was formally appraised using the Joanna Briggs Institute (JBI) quality checklist [[38](#_ENREF_38)] (see [Table A1.2](#TableA1_2), and [Appendix 1.5](#A1_5)). Scores categorised study quality as follows: low (score 0-5), medium (score 6-8), or high (score 9–11). (Any) reviews rated as being of low quality were identified for exclusion.

Synthesis of included studies

To identify overlap of studies between included reviews, citations of their included studies were recorded in an excel table, with shared citations highlighted.

Results are presented narratively and in tables. The secondary reviews are summarised with attention to strength and consistency of effects across similar studies, as well as differences, including comparing those of differing scope.

These findings represent the ‘body of evidence’ considered by the Living Guideline Group in developing Recommendations and Good Practice Points using a process described in [Appendix 1.6](#A1_6).

2.2 Body of evidence

Overview

After applying inclusion and exclusion criteria, 8 secondary studies were eligible for inclusion in the review published since 2013. After assessing these studies for study quality using the JBI checklist [[38](#_ENREF_38)], none were excluded as being of low quality (scoring 0-5).

Full extracted data are provided in the evidence tables (see [Appendix 3](#App3)). Summary characteristics of these studies are presented in [Table 2.2](#Table2_2), organised by year of publication (oldest first), and for publications in same year, alphabetically (by first author).

Scope

The eight eligible secondary studies included the 2013 Cochrane review and meta-analysis by Oono et al., [[17](#_ENREF_17)], four other systematic reviews including meta analyses [[25](#_ENREF_25), [26](#_ENREF_26), [42](#_ENREF_42), [43](#_ENREF_43)], and three stand-alone systematic reviews [[27-29](#Ref_27)]. Six secondary studies evaluated the effectiveness of parent/family-mediated interventions [[17](#_ENREF_17), [25](#_ENREF_25), [26](#_ENREF_26), [29](#_ENREF_29), [42](#_ENREF_42), [43](#_ENREF_43)], whereas two focused on examining mediating or moderating factors that may impact on the effectiveness of such interventions [[27](#_ENREF_27), [28](#_ENREF_28)].

Table 2.2: Characteristics of included secondary studies

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| Study | Design  Quality | Scope | Selection criteria | Studies identified |
| Oono et al., (2013) [[17](#_ENREF_17)] | SR/MA  High (11/11) | Effectiveness of parent-mediated approaches  Children diagnosed with ASD aged 1 – 7 years  Primary outcome: communication, social communication, parent-child interaction | Published: 2002 – Aug 2012  Any language  RCTs only | 17 studies: all RCTs  3 RCTs unique to umbrella review  N=919 children  Training duration: 1 week – 2 years |
| Nevill et al., (2018) [[26](#_ENREF_26)] | SR/MA  High (9/11) | Effectiveness of parent-mediated approaches  Children diagnosed with ASD aged 1 – 6 years  Primary outcome: core areas of functioning affected in ASD | Published: 2000 – Dec 2015  English language only  RCTs only  Excluded: children who may be autistic (but not diagnosed) | 19 studies: all RCTs  1 RCT unique to umbrella review  N=608 children  Training duration: 1 week – 1 year |
| Naveed et al., (2019) [[25](#_ENREF_25)] | SR/MA  High  (9/11) | Effectiveness of parent-mediated approaches (and peers and teachers not reported here)  Children diagnosed with ASD aged 2 – 17 years | Published: to Dec 2018  Any language  RCTs only  Excluded: studies with overlapping data sets | 23 PMI studies: all RCTs  5 RCTs unique to umbrella review  N=1409 children  Training duration: 7 weeks – 48 weeks |
| Tarver et al., (2019) [[42](#_ENREF_42)] | SR/MA  High (9/11) | Effectiveness of behavioural parent-mediated approaches  Children diagnosed with ASD aged 2 – 18 years  Primary outcome: disruptive and hyperactive behaviour | Published: to Dec 2017  Any language  RCTs only  Excluded: case studies, n < 10 | 9 studies: all RCTs  3 RCTs unique to umbrella review  N=520 children  Training duration: not reported |

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| Study | Design  Quality | Scope | Selection criteria | Studies identified |
| Trembath et al., (2019) [[27](#_ENREF_27)] | SR  High  (9/11) | Examined mediating or moderating factors affecting effectiveness of parent-mediated approaches  Children diagnosed with ASD aged 1 – 12 years  Primary outcome: adaptive behaviour (social, communication, daily living skills), ‘ASD symptoms’, cognition | Published: 2008 – Sept 2018  English language  Excluded case studies, SCED studies or non-controlled studies; interventions involving diet, exercise, medications, and studies aimed primarily at reducing/preventing social, emotional and behavioural problems | 15 studies: 13 RCTs, 1 non randomised trial, 1 cohort study  5 RCT studies and 2 cohort studies unique to umbrella review  N=1,007 children  Training duration: 8 weeks – 2 years |
| Shalev et al., (2020) [[28](#_ENREF_28)] | SR  Medium  (6/11) | Examined parent characteristics as mediating or moderating factors affecting effectiveness of parent-mediated approaches  Children diagnosed with ASD | Published: 1987 – Sept 2018  English language only | 11 studies: 8 RCTs, 3 quasi-experimental studies  3 studies unique to umbrella review  N=724 children  Training duration: not reported |
| Conrad et al., (2021) [[43](#_ENREF_43)] | SR/MA  High  (10/11) | Effectiveness of parent-mediated approaches of ≥8 sessions  Children diagnosed with ASD aged 2 – 17 years  Primary outcome: adaptive functioning | Published: to March 2020  English and Scandinavian language  RCTs only | 30 studies: all RCTs (26 in MA)  7 RCTs unique to umbrella review  N=1,934 children  Training duration: 8 weeks – 2 years |
| Pacia et al., (2021) [[29](#_ENREF_29)] | SR  Medium (6/11) | Effectiveness of parent- or sibling-mediated focussed behavioural interventions delivered by parents and siblings to young autistic children under 6 years old.  Primary outcome: social communication | Published: 1980 – 2019  English language only  Experimental, quasi-experimental group studies, and SCED studies | 54 studies: 42 SCED studies, 12 group studies (including 9 RCTs)  2 RCTS, 2 group studies, and 42 SCED studies unique to umbrella review  N=444 children  Training duration: 5 days – 1 year |

**Note:** Quality rating assigned using JBI checklist [[38](#_ENREF_38)] (see [**Appendix 1.5**](#A1_5))

**Key**: ASD=Autism Spectrum Disorder; MA=meta-analysis; RCT=randomised controlled trial, SCED=single case experimental design; SR=systematic review. See [**Appendix 3**](#App3) for Evidence Tables

The eight systematic reviews included significant overlap between eligible primary studies. Considering RCTs alone, there were between one and seven unique RCTs in the included reviews. One of the studies also included two cohort studies [[27](#_ENREF_27)], and one review of applied behavioural interventions for young children included 44 single case experimental studies [[29](#_ENREF_29)] (see under ‘Studies identified’ in [Table 2.2](#Table2_2)).

The degree of overlap and number of original studies reflect variations in the selection criteria, and the literature searches employed. These include the following factors: the recency and comprehensiveness of database searching, age range of children, the mediating agent (parent, sibling, etc) for delivering interventions, the targeted primary outcome, and the study designs considered.

Search strategy

With respect to literature searching, three reviews employed no language restriction to their search strategy [[17](#_ENREF_17), [25](#_ENREF_25), [42](#_ENREF_42)]. Four considered English language articles only [[26-29](#_ENREF_26)], and one included literature published in English or Scandinavian [[43](#_ENREF_43)].

Study characteristics

The selection criteria for three of the reviews restricted inclusion to studies of younger autistic children aged up to 6 or 7 years old [[17](#_ENREF_17), [26](#_ENREF_26), [29](#_ENREF_29)], whereas the remainder were open to studies of autistic individuals aged up to 12 years old [[27](#_ENREF_27)], adulthood [[25](#_ENREF_25), [42](#_ENREF_42), [43](#_ENREF_43)], or of unspecified age [[28](#_ENREF_28)].

All reviews considered parent-mediated approaches with the exception of Pacia et al., (2021) [[29](#_ENREF_29)] which considered both parent- and sibling-mediated approaches to the delivery of applied behavioural interventions. However the vast majority were concerned with parent-mediated approaches (50 cf only 4 studies relating to interventions mediated by siblings). In addition to considering PMI, Naveed et al., [[25](#_ENREF_25)] included studies investigating interventions mediated by same aged peers or teachers. These were considered separately and were excluded from the current umbrella review.

Some reviews considered specific outcome domains as their primary outcome, including disruptive behaviour [[42](#_ENREF_42)], adaptive functioning [[43](#_ENREF_43)], communication, and/or social communication [[17](#_ENREF_17), [29](#_ENREF_29)]. The remaining studies were more broadly inclusive of outcomes relating to the core characteristics of autism or did not specify eligible outcomes.

As primary studies rarely reported the amount of time an intervention was advised to be delivered or actually delivered to a child, the measure of ‘treatment dose’ relates to training of the parent in delivering the intervention. This was reported in many ways (number of hours, frequency per week, duration over time) making synthesis across studies difficult. The most consistently reported measure of dose was duration which, where reported, ranged from 5 days to 2 years.

It wasn’t possible to synthesise study characteristics across the eight studies given variable reporting and overlapping studies. However, the 2021 meta-analysis by Conrad et al., [[43](#_ENREF_43)] provides a good approximation of the evidence base given it conducted the most recent search (up to 2020), included the greatest number autistic participants (n=1,934), represented all ages, and considered the most RCTs investigating parent-mediated approaches (n=30), 7 of which were unique. In this review, three-quarters (76%) of included trials included children aged 7 years and younger, and one-quarter (23%) focused on children aged below 4 years. No studies included autistic adolescents aged over 14 years, and only 13% included children aged over 11 years (all of which targeted reducing disruptive behaviour or improving positive behaviour).

In Conrad et al.’s review [[43](#_ENREF_43)], the high majority (n=25, 83%) of studies targeted speciﬁc areas of autism (such as joint attention and social-communicative skills), with the remaining five studies aimed at improving parent-child social interaction and reducing behavioural diﬃculties and ‘demand-avoidant behaviour’. Control groups consisted of waitlist or other passive control conditions in 37% of included studies, usual care in 33%, and an active control with a less extensive educational program, psycho-education, or placebo parent-intervention in 27%. Antipsychotic medicine was a comparator intervention in a single study.

Study quality

Review quality was generally good, with six reviews rated (using the JBI checklist) as being of high quality (scoring 9-11) [[17](#_ENREF_17), [25-27](#_ENREF_25), [42](#_ENREF_42), [43](#_ENREF_43)], and two reviews of medium quality (scoring 6-8) [[28](#_ENREF_28), [29](#_ENREF_29)].

All reviews were impacted by the limitations of the evidence base, including a lack of controlled group studies, small samples, and lack of blinding of family members who were often informants for outcomes. Limitations of primary research in this area, including that appraised in the current review, are discussed further in [Section 2.4](#Sec2_4).

The majority of reviews included study design restrictions in their selection criteria. Of the reviews of intervention effectiveness, five included only randomised controlled trials (RCTs) [[17](#_ENREF_17), [25](#_ENREF_25), [26](#_ENREF_26), [42](#_ENREF_42), [43](#_ENREF_43)], and one considered single case experimental studies, RCTs and quasi-experimental group studies [[29](#_ENREF_29)]. Both the reviews investigating moderating factors included group studies only, with RCTs and uncontrolled or quasi-experimental group studies identified [[27](#_ENREF_27), [28](#_ENREF_28)].

Narrative summary of included studies

Studies are presented below in chronological order by date of publication, from oldest to most recent. Note that included systematic reviews had overlapping scope, search strategies, and selection criteria, and therefore shared some primary studies. Degree of overlap is represented by the number of unique articles (see [Table 2.2](#Table2_2)). It is important that the quantity of studies are not summated across reviews given this overlap.

Studies investigating effectiveness of parent-mediated approaches

Oono et al., (2013) [[17](#_ENREF_17)]

In their update of an earlier Cochrane review [[34](#_ENREF_34)], Oono et al., (2013) [[17](#_ENREF_17)] identified 17 parent-mediated intervention studies involving autistic children aged under 7 years published between 2002 and 2012. The researchers performed a meta-analysis on 10 of these following a gold standard systematic review with respect to identification, selection, appraisal and synthesis. Individual study quality was assessed using the ‘risk of bias’ tool (considering inconsistency, indirectness, imprecision, and publication bias), which informed the rating of the strength (or certainty) of evidence for each outcome, and overall, using the GRADE framework [[44](#_ENREF_44)]. (This process is similar to that employed by the Living Guideline Group in grading the quality of evidence that supports each Recommendation; see [Appendix 1.6](#A1_6)). For the current review, the study was graded as being of ‘high quality’ using the JBI critical appraisal tool.

Parent-mediated approaches appeared to lead, on average, to positive changes in parent–child interaction, and to possible gains in child language comprehension, with reduction in severity of autism (core) characteristics.

Results were inconclusive for improvements in communication, social communication, language comprehension and expression, frequency of child initiations, reduced maladaptive behaviour, improved cognitive development, and reduced parenting stress.

Due to variability in reporting methods and low sample sizes, moderators such as duration, intensity, type of intervention, parental education, child’s age, child’s IQ, and family socio-economic status were not investigated in subgroup analyses. Whilst individual studies reported potential moderating variables, results were variable and inconsistent, and the reviewers suggest that individual level data meta-analyses are needed.

The reviewers caution that the low precision (small effect sizes and wide confidence intervals), as well as general high risk of bias across studies, led to a low level of certainty (or low quality) of findings. This means that conclusions were judged as likely to change as high-quality RCTs become available.

Nevill et al., (2018) [[26](#_ENREF_26)]

Following a similar scope and methodology to the Cochrane review [[17](#_ENREF_17)], Nevill et al., (2018) [[26](#_ENREF_26)] identified 19 RCTs evaluating parent-mediated approaches for children diagnosed with ASD aged 1 – 6 years. The study was rated as being of ‘high quality’ in the current review. It identified 19 studies, 17 of which were included in meta-analyses. The authors noted that their review included six more studies to those included in the meta-analyses of the Cochrane review [[17](#_ENREF_17)], and that the quality of studies had improved in the intervening five years. Classifying outcomes into four domains of ‘core autism functioning’, the researchers reported that parent-mediated approaches led to small improvements in autism (core) characteristics, socialisation, and cognition, and a significant but ‘trivial’ effect for communication/language.

In sub-group analyses, there was no significant difference in outcomes based on dose of ‘treatment’, and conflicting associations based on informant (parent or clinician) depending on the outcome. Small numbers of studies precluded analyses investigating cognition or autism (core) characteristics as mediating factors.

Reviewers rated the strength (certainty) of evidence for the included studies as being of ‘medium’ quality (based on GRADE assessments) for all the outcome domains with the exception of socialisation studies, which were of ‘very low’ quality.

Naveed et al., (2019) [[25](#_ENREF_25)]

A broader scope was employed by the systematic review and meta-analysis by Naveed et al., (2019) [[25](#_ENREF_25)]. This considered approaches mediated by non-specialists which included parents, but also peers (similar aged children, usually fellow students), and teachers. As these last two groups are outside the scope of the current umbrella review, subgroup analyses relating to parent-mediated intervention group participants provided effect sizes. The review, rated as being of ‘high quality’ for the current umbrella review, considered 33 RCTs, 23 of which related to parent-mediated approaches, delivered to children aged 2 to 17 years. Ten of these studies were unique to this review (i.e., not included in other appraised reviews), five of which were RCTs.

Comparing recipients of PMI with control group participants, a large effect was evident for improved joint engagement (n=5 studies). Moderate effects were identified for reduced autism (core) characteristics (n=9 studies), improved self-regulation (n=3 studies), reduced child distress (n=3 studies), improved social skills (n=10 studies), improved expressive language (n=5 studies), and a beneficial impact to the parent-child relationship (n=6 studies). Small effect sizes were found for reduced repetitive behaviours (n=3 studies), reduced parental distress (n=10 studies), and improved parental self-efficacy (n=8 studies).

Whilst relative benefit of parent-mediated versus other non-specialist mediated interventions is not within scope of the current umbrella review, it is of interest to note that in meta-analyses considering all studies, effect sizes did not vary as a function of delivery agent (i.e., parent, teacher, peer) for most outcomes. Exceptions were found for two outcomes, with conflicting results; for communication, peer- and teacher-mediated interventions revealed greater effect sizes than for PMIs, and for joint engagement, the reverse was true. Caution should be observed however given that the number of studies reporting on peer and teacher administered approaches was relatively small, and for several outcomes, absent altogether. It is therefore not possible to draw conclusions about the relative effectiveness of approaches delivered by different non-specialists.

The majority of studies were assessed as being of low risk of bias according to the Cochrane tool. The authors argued that the findings support the benefits of non-specialist approaches for autistic children and their caregivers, although no particular therapy, approach or delivery agent was recommended.

Tarver et al., (2019) [[42](#_ENREF_42)]

In the same year, Tarver et al.’s (2019) [[42](#_ENREF_42)] systematic review (rated ‘high quality’ in the current review) focussed on the effectiveness of behavioural parent-mediated approaches for addressing comorbidities of autism including anxiety and ADHD, and specifically disruptive behaviour and hyperactivity.

Nine RCTs (reported in 11 papers, three of which were unique to this review) were included in meta-analyses. From parent-reported outcomes, there was a moderate improvement (reduction) in child disruptive behaviour (n=9 studies) and a small effect was found for hyperactivity (n=3 studies).

Considering parental outcomes, a small improvement was found for parental stress. There was no significant improvement for parental efficacy in five studies exhibiting high heterogeneity.

Conrad et al., (2021) [[43](#_ENREF_43)]

Recently published was the systematic review of Danish researchers Conrad et al., (2021) [[43](#_ENREF_43)] which considered parent-mediated approaches for autistic individuals aged 18 months to 17 years. The database search strategy was somewhat limited by following a stepwise process, beginning with secondary research, and then - relying on citation searching from these reviews – primary studies published between 2017 and March 2020. The review methodology was otherwise very robust and judged to be of ‘high quality’ in the current review.

Of 30 included RCTs, 26 were included in a meta-analysis (seven of which were unique to this review). For each outcome, significant improvement for the parent-mediated intervention group compared with the control or comparator group were reported alongside its rated study quality (‘certainty of benefit’, using GRADE).

The meta-analyses suggest a small but clinically relevant improvement in children’s adaptive functioning as rated by parents (n=8 studies), but not observed in clinician-rated assessments (n=2 studies). The authors suggest that this is a particularly strong indicator of the value of parent-mediated approaches given that they do not directly target adaptive functioning. PMI’s also appeared to lead to moderately large improvement in a child’s (parent-rated) disruptive behaviour, which were considered secondary outcomes by the reviewers.

Whilst not statistically significant, a small clinically relevant improvement in autism core characteristics was observed for PMI recipients in clinician-assessed measures (though not parent-assessed). A small improvement (though not clinically relevant) was observed for parental well-being. Adverse events were rare and of low significance.

Notably, the certainty in the evidence was generally assessed as ‘low’ (with the exception of assessments of disruptive behaviour, where it was ‘moderate’). Certainty was reduced due to risk of bias, prominently from lack of blinding, and a risk of imprecision due to few participants being included in the meta-analyses. There were no differences in subgroup analyses comparing outcome target area of the intervention (i.e., language, aggression management, training in social skills) and other intervention types.

The reviewers concluded that clinicians may consider introducing parent-mediated approaches to autistic children, but cautioned that high-quality RCTs are needed as the effects are not well-established.

Pacia et al., (2021) [[29](#_ENREF_29)]

Published in the same year is the systematic review from Pacia et al., (2021) from Ireland [[29](#_ENREF_29)] (rated ‘medium quality’ in the current review). It considered the effectiveness of parent- or sibling-mediated ‘focussed behavioural interventions’ (using approaches and strategies based on applied behavioural principles) for addressing social communication outcomes for young autistic children aged 0 – 6 years old. A total of 54 studies were included representing a broad range of experimental study designs, including 42 single case experimental design (SCED) studies, and 12 group studies, 11 of which were RCTs). Nearly all (n=50) evaluated parent-mediated approaches, with 4 studies involving siblings (aged 4-13 years) delivering the intervention.

Treatment effectiveness was determined differently for the SCED studies compared with the group studies, but there was broad agreement in findings. There was slightly greater effectiveness evident for social engagement outcomes, compared to language and communication, and imitation and play, where improvement was more mixed.

In sub-group analyses considering moderating effects, there was no difference in effectiveness based on agent (parent, sibling), modality (telehealth, in situ), setting (clinic, home), or dose (hours of intervention). Of those studies that evaluated generalisation of outcomes (about 75%), over 90% demonstrated partial or complete generalisation.

Studies investigating mediating factors

Trembath et al., (2019) [[27](#_ENREF_27)]

In an Australian systematic review, Trembath et al., (2019) [[27](#_ENREF_27)] (rated as being of ‘high quality’ in the current review), identified 41 studies assessing effectiveness of parent-mediated approaches for improving adaptive behaviour, autism (core) characteristics, or cognition in autistic children aged up to 12 years. Rather than reporting effectiveness gains, the research team described the use of restrictive selection criteria and study characteristics as raising concerns about whether research in this field can be generalised to the broader community.

The review also identified a subset of 15 studies (including 13 RCTs) which investigated possible factors which may predict, mediate or moderate PMI effectiveness. A broad variety of 45 possible factors were considered across domains relating the baseline child and parent characteristics and socio-demographic variables, child social and language skills, and contextual and intervention characteristics (e.g., recruitment, adherence, fidelity, dose). Whilst no association was observed between parent-mediated intervention effectiveness and parent education or occupation in two studies, there were mixed results in another study. Studies were generally over-represented by higher-educated parents anyway which may dilute the ability to detect true effects. One study considering qualitative data found positive parent behaviours such as sensory-motor support, joining, use of affect, smiling, and reciprocity were related to social communication outcomes.

In general however findings were non-significant or mixed with no clear or consistent patterns identifying moderating factors, with synthesis hampered by the small number of studies investigating each factor.

The reviewers provide important recommendations for ways future research can be improved in terms of study quality, reporting and measurement consistency, and examination of factors that may predict, moderate, and mediate intervention effectiveness for children and their parents (see [Section 2.4](#Sec2_4) for a discussion of these issues).

Shalev et al., (2020) [[28](#_ENREF_28)]

Another study of potential moderators in PMI effectiveness considered parent characteristics specifically. Shalev et al.’s (2020) [[28](#_ENREF_28)] study (rated ‘moderate quality’ in the current review) was hampered by a limited search strategy and lack of formal critical appraisal. Eleven controlled group studies were identified, 8 of which were RCTs. There were contrasting effects for parent stress measured at baseline (n=4), with high baseline stress leading to improved outcomes in some studies and poorer outcomes in others, and for some, there was no moderating effect at all. Similarly mixed results from three studies were found for the moderating effect on outcomes of parent’s socio-economic status, and education. The authors suggest that variability in results may be due to core differences in the interventions and measures employed. No significant moderating effect was found for maternal age or parent’s insight into their child’s cognitions and affect, although these findings were from single studies.

Although this emerging literature is characterized by high variability in regard to the specific treatment employed, measures used, and child outcome targeted, initial results from this review suggest that parent factors have the potential to act as moderators. These results underscore the need for systematic research on the role of parent baseline characteristics in this field (see [Section 2.5](#Sec2_5) for further discussion on future research).

2.3 Synthesis of results

Effectiveness of parent-mediated approaches

Key results relating to the effectiveness of parent-mediated approaches (compared with a control/comparator) are presented in [Table 2.3](#Table2_3). These are grouped broadly into outcome domains, and then within each domain by similar sub-categories, within which studies are ordered by lower to higher number of included studies for the reported outcome (where the number of studies is the same, more recently published reviews are presented last).

Note that assigning outcomes within domains was a fraught process and is somewhat subjective. Language and social communication outcomes were the most difficult to disentangle conceptually due to having multiple and overlapping goals, varying focus (broad domains and specific skills), and different terminology. Moreover, whether outcomes are appropriate from a neurodiversity perspective may differ between sub-categories within a broader domain (for example, social skills compared to other social communication outcomes). Care should therefore be taken in reporting results at the domain level.

Findings from group-controlled studies (predominantly RCTs) are presented from the appraised systematic reviews reporting on these outcomes. For each row, whether there was an improvement found is indicated, and if so to what degree, for the group receiving a parent-mediated approach compared with a control/comparison group. These were based on pooled effect sizes for meta analyses, or descriptive indications of improvement for studies synthesised narratively.

Colours have been used to visually highlight the degree to which improvement is indicated, ranging from possible improvement where results were very mixed and uncertain (highlighted in orange), small improvement (yellow), moderate improvement (blue), to large improvement (green). Where no indication was given for the size of an effect, a finding is described simply as “improvement” and is highlighted in aqua. No colour is used where no improvement was observed.

Within each outcome domain, those reviews contributing a larger number of studies, and reporting larger improvements, increase the strength of evidence and confidence that an intervention is beneficial for that outcome. However, results across reviews within an outcome domain should not to be considered unique findings that can be summated, as the studies reported for each review may overlap with others. Instead, they are a reflection of the consistency (or lack thereof) of conclusions from reviews of different but overlapping scope.

Some variation is to be expected given the heterogeneity of the parent-mediated approaches being considered, their intensity, their therapy goals, measures employed, informants used, and the characteristics of the participants, particularly their age. The findings relating to effectiveness are also limited by small sample sizes of included studies per outcome. This is reflected in the low strength of evidence applied (using GRADE) for many outcomes in the appraised reviews. Methodological limitations are discussed further in [Section](#Sec2_3) 2.4.

Communication/language development

Within the area of language development, including language comprehension (receptive language) and language expression (expressive language), there were mixed findings. Three meta-analyses reported on broad communication and language outcomes. One found no improvement in a broad “communication” measure for children and adolescents (n=10 studies) [[25](#_ENREF_25)]. By contrast, a review of young children reported a possible improvement (n=6 studies) [[29](#_ENREF_29)]. The largest meta-analysis of trials for young children was able to be more definitive, suggesting a small improvement in communication/language outcomes (n=13 studies) [[26](#_ENREF_26)].

Other review syntheses of smaller numbers of studies considered receptive and expressive language outcomes separately. Based on two studies of younger children, the Cochrane review reported a possible improvement for receptive language/comprehension, and from three studies, a possible improvement in expressive language [[17](#_ENREF_17)]. More recently, a meta-analysis considering children across a broader age range [[25](#_ENREF_25)] concluded there was no improvement in recipients of PMI for receptive language (n=4 studies), but a moderate improvement in their expressive language (n=5 studies).

Social communication

Social communication and social interaction are one of two core affected areas of development that lead to a diagnosis of ASD [[45](#_ENREF_45)]. Social communication outcomes where diverse and likely to reflect the different age groups targeted. Considering the more specific skill of joint attention, a small improvement was observed based on two trials of younger children in the Cochrane review [[17](#_ENREF_17)]. However a later more inclusive meta-analysis of seven studies across a broader age group of children reported no improvement [[25](#_ENREF_25)]. No improvement was reported for initiation skills in young children (n=4 studies) in the Cochrane review [[17](#_ENREF_17)].

Table 2.3: Summary of the impact of parent-mediated approaches on outcomes reported in effectiveness studies

|  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- |
| **Outcome *(reported by)*** | **Mediated by** | **Child age (years)** | **Systematic review** | **Included studies** | **Recipients of parent-mediated approaches compared with control** |
| **Language Development** (comprehension and expression) | | | | | |
| Communication/language | Parents | < 6 | Pacia et al., (2021) [[29](#_ENREF_29)] | N=6 | Possible improvement |
| Communication | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=10 | No improvement |
| Communication/language | Parents | 1 – 6 | Nevill et al., (2018) [[26](#_ENREF_26)] | N=13 | Small improvement |
| Receptive language/comprehension | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=2 | Possible improvement |
| Receptive language/comprehension | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=4 | No improvement |
| Expressive language | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=3 | Possible improvement |
| Expressive language | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=5 | Moderate improvement |
| Social communication | | | | | |
| Joint attention | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=2 | Small improvement |
| Joint attention | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=7 | No improvement |
| Initiation | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=4 | No improvement |
| Social communication | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=3 | Possible improvement |
| Social engagement | Parents/siblings | < 6 | Pacia et al., (2021) [[29](#_ENREF_29)] | N=4 | Improvement |
| Socialisation (incl. social communication) | Parents | 1 – 6 | Nevill et al., (2018) [[26](#_ENREF_26)] | N=13 | Small improvement |
| Imitation/Play | Parents | < 6 | Pacia et al., (2021) [[29](#_ENREF_29)] | N=1 | Moderate improvement |
| Engagement | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=5 | Large improvement |
| Social skills | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=10 | Moderate improvement |
| Parent-child relationship | | | | | |
| Parent-child interaction/synchrony | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=3 | Large improvement |
| Parent-child relationship | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=6 | Moderate improvement |

Table 2.3: Summary of the impact of parent-mediated approaches on outcomes reported in effectiveness studies (continued)

|  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- |
| **Outcome (reported by)** | **Mediated by** | **Child age (years)** | **Systematic review** | **Included studies** | **Recipients of parent-mediated approaches compared with control** |
| Autism core characteristics | | | | | |
| Repetitive behaviour | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=3 | Small improvement |
| Autism characteristics | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=6 | Small improvement |
| Autism characteristics | Parents | 1 – 6 | Nevill et al., (2018) [[26](#_ENREF_26)] | N=6 | Small improvement |
| Autism characteristics *(parent-rated)* | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=7 | No improvement |
| Autism characteristics | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=9 | Moderate improvement |
| Autism characteristics *(clinician-rated)* | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=9 | Small improvement |
| Behaviours of concern | | | | | |
| Child distress | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=3 | Moderate improvement |
| Self-regulation | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=3 | Moderate improvement |
| Hyperactivity *(parent-rated)* | Parents | 2 – 18 | Tarver et al., (2019) [[42](#_ENREF_42)] | N=3 | Small improvement |
| Maladaptive behaviour | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=4 | No improvement |
| Disruptive behaviour *(parent-rated)* | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=7 | Large improvement |
| Disruptive behaviour *(parent-rated)* | Parents | 2 – 18 | Tarver et al., (2019) [[42](#_ENREF_42)] | N=9 | Moderate improvement |

Table 2.3: Summary of the impact of parent-mediated approaches on outcomes reported in effectiveness studies (continued)

|  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- |
| **Outcome (reported by)** | **Mediated by** | **Child age (years)** | **Systematic review** | **Included studies** | **Recipients of parent-mediated approaches compared with control** |
| Adaptive functioning | | | | | |
| Adaptive functioning | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=2 | No improvement |
| Adaptive functioning *(clinician-rated)* | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=2 | No improvement |
| Adaptive functioning | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=5 | No improvement |
| Adaptive functioning *(parent-rated)* | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=8 | Small improvement |
| Development | | | | | |
| Cognitive development | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=5 | Possible improvement |
| Cognition | Parents | 1 – 6 | Nevill et al., (2018) [[26](#_ENREF_26)] | N=6 | Small improvement |
| Visual reception | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=3 | No improvement |
| Motor skills | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=5 | No improvement |
| Parent outcomes |  |  |  |  |  |
| Parental stress | Parents | 1 – 6 | Oono et al., (2013) [[17](#_ENREF_17)] | N=2 | Possible improvement |
| Parental stress | Parents | 2 – 18 | Tarver et al., (2019) [[42](#_ENREF_42)] | N=7 | Small improvement |
| Parental efficacy | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=5 | No improvement |
| Parental self-efficacy | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=8 | Small improvement |
| Parental distress | Parents | 2 – 17 | Naveed et al., (2019) [[25](#_ENREF_25)] | N=10 | Small improvement |
| Parental well-being | Parents | 2 – 17 | Conrad et al., (2021) [[43](#_ENREF_43)] | N=12 | No improvement |

Social communication outcomes in young children was considered in three reviews. The early Cochrane review [[17](#_ENREF_17)] found a possible improvement was suggested from three trials, whilst a recent systematic review of four trials [[29](#_ENREF_29)] identified improvement for social engagement. A meta-analysis of 13 trials [[26](#_ENREF_26)] assessing socialisation considered improvement to be small. Considering the more specific outcome of imitation and play skills, moderate improvement was found for young children [[29](#_ENREF_29)], although this was based on a single study.

However, a meta-analysis that included children and adolescents reported stronger effect sizes [[25](#_ENREF_25)]. Large improvement was reported for engagement (from 5 studies), and moderate improvement for social skills (based on 10 studies).

Parent-child relationship

Two reviews reported on the impact of parent-mediated approaches on the parent-child relationship specifically, which Oono et al., (2013) described as measuring interaction or synchrony between a parent and their autistic child. Strong effect sizes were evident in both meta-analyses. The earlier Cochrane review [[17](#_ENREF_17)] of young children reported a large improvement in parent-child synchrony (n=3 studies) whereas a later review [[25](#_ENREF_25)] of 6 studies including children and adolescents reported a moderate improvements for the parent-child relationship.

Autism (core) characteristics

Autism characteristics, commonly assessed by ‘autism symptom severity’ scales, relate to observed traits that are central to the diagnosis of autism (sometimes called ‘core characteristics’). A meta-analysis of nine RCTs including children and adolescents [[25](#_ENREF_25)] found moderate improvement to autism core characteristics (‘severity’). The same study also reported a small improvement in repetitive behaviour from 3 studies. This outcome is relevant to the second core area of autistic characteristics considered in the diagnosis of ASD of ‘restricted, repetitive patterns movements, interests and activities’ [[45](#_ENREF_45)]. Two other meta-analyses of young children aged under 6-7 years observed a small improvement in autism core characteristics [[17](#_ENREF_17), [26](#_ENREF_26)].

A fourth review had different conclusions depending on the assessment’s informant [[43](#_ENREF_43)]. The review of studies including autistic children and adolescents reported a small improvement for *clinician-rated* autism core characteristics (n=9 studies), but no improvement for *parent-rated* assessments (n=7 studies). Given that blind assessment to allocation of an observed individual (to the intervention or control group) is more possible for clinician assessments than parents, it is arguable that one could be more confident in the clinician assessments.

Behaviours of concern

Behaviours of concern included a broad range of outcomes including child distress, self-regulation, hyperactivity, and disruptive behaviour. Findings for this group of outcomes were generally moderately strong and largely consistent.

Whilst the Cochrane review (2013) [[17](#_ENREF_17)] found no improvement from four studies on what they described as maladaptive behaviour, three more recently published meta-analyses of RCTs including children and adolescents all found improvements. These included the review by Conrad et al., (2021) [[43](#_ENREF_43)] of seven RCTS which found a large reduction in *parent-rated* disruptive behaviour. Also considering disruptive behaviour, a meta-analysis of behaviourally based PMI found moderate improvement (n=9 studies), as well as a small improvement in *parent-rated* hyperactivity (n=3 studies) [[42](#_ENREF_42)]. And a third meta-analysis found moderate improvement for child distress (n=3 studies) and self-regulation (n=3 studies) outcomes [[25](#_ENREF_25)].

The difference between these more recently published studies finding moderate and strong effects compared to the Cochrane review’s lack of detected improvement for maladaptive behaviour may be explained by the inclusion of additional studies published in the intervening years increasing statistical power, and potentially the inclusion of older children.

Adaptive functioning

The impact of parent-mediated approaches on children’s adaptive functioning were investigated in three reviews. Two reviews reported no improvements, each based on two studies [[17](#_ENREF_17), [25](#_ENREF_25)]. The third review was the recent meta-analysis by Conrad et al., (2021) [[43](#_ENREF_43)] which concluded there was a small improvement in (unblinded) *parent-rated* adaptive functioning (n=8 studies), but none when assessments were *clinician-rated* (n=2 studies).

Developmental gains

Considering outcomes loosely categorised as physical and developmental outcomes, no improvements were found for visual reception, or motor skills [[25](#_ENREF_25)]. However cognitive development was reported as being possibly improved based on five early childhood studies in the Cochrane review (2013) [[17](#_ENREF_17)]. More recently, a firmer finding of a small improvement was observed for cognition based on six studies in a meta-analysis of trials including children and adolescents [[26](#_ENREF_26)].

Parent outcomes

In the early Cochrane review by Oono et al., (2013) [[17](#_ENREF_17)], possible improvement was reported for parental stress based on two studies including young children. Six years and five additional studies later, the meta-analysis by Tarver et al., (2019) [[42](#_ENREF_42)] concluded that there was a small improvement for this outcome based on RCTs including children and adolescents receiving parent-mediated behavioural interventions (n=7 studies). In the same year, another meta-analysis of studies of children and adolescents [[25](#_ENREF_25)] found parent-mediated approaches had no impact on parental efficacy (n=5 studies). However the review reported small improvements for both reduced parental distress (n=10 studies), and increased self-efficacy (n=8). Finally, the recent meta-analysis by Conrad et al., (2021) [[43](#_ENREF_43)] found that there was no impact on measures of the broader measure of parental well-being from their consideration of 12 studies.

Overall there appears consistent evidence from a good number of studies from different reviews that parent-mediated approaches can lead to modest positive benefits for parents, with small improvements in parents’ levels of stress, distress, and self-efficacy.

Adverse events

In addition to effectiveness outcomes, one review, a meta-analysis by Conrad et al., (2021) [[43](#_ENREF_43)], reported on whether there were differences in reported adverse events by treatment group. The reviewers reported that only two studies mentioned adverse events. Both studies reported finding such events to be rare, and no more likely to occur for parent-mediated approaches groups compared with their control groups.

Moderating or mediating factors

Two systematic reviews were dedicated to investigating factors which may predict, mediate or moderate parent-mediated approaches’ effectiveness. Unfortunately, synthesis was hampered by the small number of studies systematically investigating such variables, and the consequently low sample sizes for sub-group analyses. Trembath et al., (2019) [[27](#_ENREF_27)] found relationships of variables were non-significant or mixed, suggesting no clear or consistent patterns. A single study suggested that parent behaviours (including sensory-motor support, joining, use of affect, smiling, and reciprocity) were associated with improved social communication outcomes in their autistic children. Arguably these factors may be characteristics specifically targeted in some interventions.

Also considering parent characteristics, Shalev et al.’s (2020) systematic review [[28](#_ENREF_28)] reported mixed results for parent’s baseline stress, socio-economic status, and education. The authors suggest that whilst parent factors have the potential to act as moderators, variability in results between studies may be due to other variables, such as the intervention used and measures employed.

Five reviews investigating effectiveness of PMI also attempted to investigate mediating and moderating factors where possible. The Cochrane review’s meta-analysis [[17](#_ENREF_17)] found that subgroup analyses were not possible due to low sample sizes and differing reporting methods. Whilst the reviewers noted that included studies did report on moderating factors, findings were variable and inconsistent.

Four other reviews were similarly unable to identify moderating factors consistently associated with effectiveness. Specifically, there were no significant differences in outcomes found for dose of ‘treatment’ [[26](#_ENREF_26), [29](#_ENREF_29)]; the outcome targeted by the intervention [[43](#_ENREF_43)]; modality (telehealth, in situ) [[29](#_ENREF_29)]; or setting (clinic, home) [[29](#_ENREF_29)]. And there was no significant difference in effectiveness based on mediated delivery agent (parent, sibling) for most [[25](#_ENREF_25)] or all [[29](#_ENREF_29)] outcomes investigated. An exception were the conflicting results reported for two outcomes in one meta-analysis [[25](#_ENREF_25)], with improvements in joint attention favouring parent-mediated delivery, and communication outcomes benefitting more from teacher or peer delivery. There were also conflicting associations reported based on informant (parent or clinician), depending on outcomes [[26](#_ENREF_26)].

2.4 Review limitations

The review is limited by the structured approach inherent in its methodology, as well as the quality of the studies appraised.

Limitations of review methodology

The current review was initially restricted to English language studies. However follow-up searching using the same strategy restricted to non-English Journal articles published in 2013 or later did not identify any eligible reviews. This gives reassurance that the initial language restriction did not affect identification of studies.

The review’s search strategy was very broad and inclusive, using a large list of search terms and an absence of methodology limiters. Studies were initially selected for appraisal by examining the articles’ abstracts. Therefore, it is possible that some studies were inappropriately excluded prior to examination of the full text. To minimise this possibility, where detail was lacking or ambiguous, papers were retrieved as full text. Supplemental searching, including considering the reference list of all retrieved studies, and narrative reviews retrieved as background material, extended the search catchment, increasing the likelihood of inclusion of eligible primary studies.

Geographically, most of the primary studies included in the appraised reviews of effectiveness tended to be conducted by researchers in industrialised, developed countries, and featured predominantly white children [[27](#_ENREF_27)]. The generalisability of the evidence base to the New Zealand population (particularly Māori and Pacific Peoples) and health/disability service context may therefore be limited. Such factors must be considered in implementing the research findings (and this Guideline update) locally. This is particularly needed to honour the Crown’s obligations to Te Tiriti o Waitangi with respect to considering what approaches and resources are needed to achieve equitable health outcomes for Māori[[2]](#footnote-2).

In order to consider the relevance and applicability of the international evidence to Aotearoa’s culture and service systems, qualitative and survey-based research was identified which assessed the lived experiences of members of the Autism community in New Zealand, including Māori. As discussed in [Section 1.3](#A1_3_NZ), these studies provide information on appropriate adaptions that reflect cultural preferences and reduce barriers to accessing services.

Limitations of appraised studies

The ability to interpret and generalize findings on the effectiveness of parent-mediated approaches was limited by the methodological quality and significant heterogeneity of the primary studies that the appraised systematic reviews draw from.

Study design and quality

The quality of the appraised secondary studies was generally very good, with six reviews rated as being of high quality, and two of medium quality. Methodological strengths included employing broad search strategies, independent selection and data extraction by multiple researchers, detailed tabulated data of included studies, and consideration of study design and quality.

Key limitations of reviews included a lack of formal critical appraisal checklists, lack of methodological critique, and limited descriptions of included studies’ design and results. Only one of the eight included reviews, the Cochrane review (2013) [[17](#_ENREF_17)], considered grey literature. Restriction to peer-reviewed Journal articles is likely to lead to publication bias since studies that show an absence of effect are less likely to be published.

Attempts to synthesise primary studies in this area were limited by wide variation in the following areas: population age and characteristics, study setting, intervention strategies, dose and duration of intervention, targeted outcomes, assessment tools and informants, and intervention mediators. These factors lead to substantial methodological heterogeneity in meta-analyses.

The reviews themselves are impacted by the methodological limitations of the primary studies they included. All but one of the six reviews of effectiveness were restricted to RCTs which represent level II evidence in the NHMRC hierarchy (**Appendix 1,** [Table A1.1](#TableA1_1)), however quality within these designs is affected by how they are conducted, as discussed below with respect to different areas of study design.

Sampling and recruitment

Studies included in the appraised reviews were hampered by low sample sizes. When samples are small, and based on limited recruitment, poor response and study completion rates, sampling, detection, and attrition biases can lead to unpredictable effects. They reduce the likelihood that the results represent, and are generalisable to, the population the intervention is targeted towards.

Determination of diagnosis, presence of co-occurring conditions, and level of cognitive functioning were not consistently reported in the primary studies or synthesised in appraised reviews. This makes it difficult to determine their applicability to different autistic individuals, or to investigate any moderating effect on intervention effectiveness.

The included Australian investigation by Trembath et al., (2019) [[27](#_ENREF_27)] of mediating factors in 15 studies provided a descriptive summary of study selection and socio-economic characteristics reported across 41 parent-mediated approaches studies representing 2,707 autistic children. This synthesis found that around half the studies targeted children with minimum specified developmental and verbal ability (20 studies). Commonly, children with co-occurring conditions were excluded (n=23 studies). Children were also excluded on the basis of epilepsy, seizures, and use of medications (n=9 studies). In some studies, parents’ characteristics were used to exclude children, including presence of parents’ psychiatric disorders (n=8 studies), inaccessibility to participation (e.g., language proficiency, distance from clinic), non-acceptance of protocols, and prior exposure to intervention (n=16 studies). The use of restrictive selection criteria can reduce external validity. It is of particular concern in the field of autism, given the high prevalence of co-occurring conditions, and that 25- 30% of autistic children lack functional speech or are minimally verbal [[46](#_ENREF_46)].

Studies evaluating parent-implemented interventions also tend to target pre-school or primary school aged children. In the 2021 meta-analysis by Conrad et al., [[43](#_ENREF_43)], no studies included children aged over 14 years.

Interventions and comparators

All appraised reviews considered parent-mediated approaches with one [[29](#_ENREF_29)] also including four studies evaluating sibling–mediated approaches. However, the form and delivery of the programmes offered varied widely, often involving multiple components, making it difficult to determine effective features.

The type of comparators employed in trials were also diverse, including waitlist or passive control conditions, usual care, or for about a quarter of studies, an active comparator offering a less extensive educational program, psycho-education, or placebo parent-intervention. The use of active comparators that are structurally matched to the intervention aim to control for non-specific factors that may be provided by the intervention but are not core to it. They are more effective placebos than a waitlist control.

The intensity and duration of interventions also varied, ranging from 5 days to 2 years, although investigating dose effects was limited by the inconsistent and incomplete ways this information was reported. Programme fidelity and number of intervention hours delivered by a parent/family member were rarely recorded and is likely to be highly variable. This makes it difficult to ascertain the impact of programme adherence and intensity as mediators of intervention effectiveness [[17](#_ENREF_17)].

An attempt was made to classify outcomes within different domains in [Table 2.3](#Table2_3). This process was challenging due to conceptual overlaps, particularly those between language, communication, and socialisation. It also highlighted differences in how broadly or narrowly an outcome was defined. For example, domains could relate to areas of behaviour (e.g., initiation, joint attention), sub-scale areas (e.g., social awareness), and broader and combined domains (e.g., communication/language, and social communication). To check the face validity of an initial attempt at organisation, two members of the Living Guideline Group (autism researcher Martyn Matthews and child psychiatrist and LGG Chair Matt Eggleston) provided peer review for the classifications used in the Table, which led to some changes. The inherent subjectivity of this process is accepted, recognising the significant underlying heterogeneity of the studies and summated outcomes in each review (as discussed in the next section). The purpose of this process was to assist in considering the weight and consistency of findings across the reviews and to look for patterns.

Assessment

Awareness of the importance of choosing outcomes that are important to people on the autism spectrum is increasing. Some measured changes may not be sought or valued by autistic participants, their families, or the autistic community. Reduction of “undesirable or challenging behaviours” is dependent on for whom it is ‘undesirable’ or ‘challenging’. The degree to which some behaviours may be undesirable may vary in terms of context and the perceptions of others, and be socially constructed [[47](#Ref_47)].

In the appraised systematic reviews, narrative synthesis of outcomes was hampered by the lack of overlap between specific measures, given the very wide range of scales, subscales, and instruments employed to measure the same broad outcome, and use of different informants (e.g., clinician, researcher, parent, child). Synthesis becomes particularly fraught in meta analyses. The calculation of standardized effect sizes is intended to account for variability across measures. However, Nevill et al., (2019) [23] warned that whilst there are statistical processes for standardising mean differences when pooling effect sizes across different assessment instruments, these measures will vary in the outcomes they attempt to capture. To then synthesise the findings of meta-analyses and narrative syntheses in an umbrella review can compound such heterogeneity requiring great caution in interpreting results.

Statistically different scores may not translate to clinically significant changes. Based on an instrument’s psychometric properties and clinical relevance [[48](#_ENREF_48)], some commonly used outcome instruments (e.g., the Autism Diagnostic Interview-Revised, CARS, and Social Communication Questionnaire) may be inappropriate as outcome measures of change [[26](#_ENREF_26)].

A major limitation of research in parent-mediated approaches relates to lack of allocation concealment to participants, and the impact of this on assessments where informants are unblinded to condition, being parents/family members delivering the intervention, their autistic children, or programme staff. Unblinded studies cannot control for reporting biases of observers in seeing an improvement, which may correlate with expectations about the value of the programme, and/or a desire to assist the researchers. Such reporting biases from lack of blinding may artificially inflate ratings of the effectiveness of an intervention. They are less likely when the control group uses an active comparator.

Reviewers have observed that few studies evaluating parent-mediated approaches have assessed longer term follow-up, and studies also lack measures of cost-effectiveness and economic feasibility [[25](#_ENREF_25)]. Longer-term follow-up is crucial to determine whether any improvements to outcomes are sustained after the intervention ends. Equally important, it is imperative to investigate the potential emergence of harms in the longer term; for example, on autistic people’s mental health.

Studies also lack measures of cost-effectiveness and economic feasibility [[25](#_ENREF_25)]. Lack of economic assessments make it difficult to determine whether programmes evaluated in a research context can be applied practically and effectively ‘in the real world’.

2.5 Future research

Addressing the limitations of the current evidence base will inform future research into parent-mediated supports for autistic individuals.

Study design and quality

Fully randomised controlled trials, using robust methods of randomisation and reporting, are needed. Larger sampled studies will increase (statistical) power to detect true effects, and to permit meta-regressions and subgroup analyses to explore both moderation and mediation of effects. Replication studies should also be undertaken to confirm earlier research findings [[43](#_ENREF_43)].

Ideally studies should be mixed methods, which attempt to integrate both quantitative and qualitative elements. These provide complementary evidence of how an intervention is helpful on a personal level, within a social and cultural context. The inclusion of qualitative research can also help in understanding why an approach, therapy or ‘intervention' programme is not effective and how it may be improved, better implemented, or better directed to those most likely to benefit.

Lack of allocation concealment is a major weakness in this area, where children and family members are not unaware/blind to condition [[17](#_ENREF_17), [32](#_ENREF_32)]. Assessments from other informants (e.g., teachers, peers, clinicians), blinded to condition, should be included in future trials to reduce reporter biases [[42](#_ENREF_42)].

Reporting of studies should be more detailed, with research in this field commonly omitting clear information about the participants, setting, the delivery of interventions, and the fidelity of family members to adhering to intervention protocols [[49](#_ENREF_49)].

Sampling and recruitment

Studies need to reflect a diversity of participants representative of varying characteristics of the autistic community, including communication abilities, cognitive functioning, cultural/ethnic backgrounds, socio-economic status, gender, and age. Broader characterised samples will permit the systematic investigation of whether approaches are more or less favoured, used, effective, and appropriate for people across a range of backgrounds and characteristics. Efforts to increase diversity need to be made through targeted sample recruitment.

Interventions and comparators

There is a need to better support parents or other family members in their capacity to be trained in and deliver programmes and strategies. As discussed by Trembath et al., (2019) [[27](#_ENREF_27)], practical consideration must be given to the appropriateness and feasibility of parent-mediated approaches, including access based on geographical location, the time required to deliver the intervention as requested, and the costs for families in terms of time, transport, and lost earnings. The researchers observe, “equally, or perhaps more importantly, is the need for closer consideration of the factors that may have an impact on the appropriateness of parent-mediated interventions for individual families” (p. 1306) [[27](#_ENREF_27)]. This could include considering parents’ culture, psychological stress, cognitive ability, and communication preferences.

Whilst parents and caregivers have been the prime focus of programme delivery in research to date, there is a growing literature on sibling involvement deserving more investigation. Future research could also explore approaches that involve the broader family unit as a whole and to integrate benefits across the family [[29](#_ENREF_29)].

Programme fidelity appears to be low for parent-mediated approaches, although it is rarely measured and reported. Objective information is greatly needed into how family members implement a programme and their use of the skills they have been trained in. Further research is also needed to investigate parent factors that may affect treatment fidelity [[28](#_ENREF_28)]. Adherence may be improved by manualising programmes to ensure consistent delivery [[43](#_ENREF_43)]. Training parents in delivering supports may also be more feasible, accessible and financially viable when conducted outside of clinical-research settings and instead through community-based services [[27](#_ENREF_27)].

Much research relating to parent-mediated approaches has focused on early intervention and supporting young children. There is a need to develop and evaluate parent-mediated approaches suited to older children and adolescents, and addressing a broader range of outcomes [[32](#_ENREF_32)]. More research attention could also be directed to programmes that aim to reduce parents’ stress [[17](#_ENREF_17)], which may lead to indirect benefits for the children the parents care for and in turn, improve their capacity to deliver programmes effectively.

With respect to comparators, when usual care is used as a control condition, comparisons are limited by variability in the quality of community-based services received. As alternatives, active comparison groups such as parent education approaches may provide more consistent comparators, and also reduce risk of response bias in parent-rated outcomes [[26](#_ENREF_26)].

Assessment

Understanding the primary preferences and priorities of autistic individuals is central not only to developing more effective interventions, but to assessing whether supports meet their needs.

Future research should identify a battery of validated, standardised and cross-culturally valid instruments to permit more meaningful inter-study comparisons and syntheses. These should include objective direct assessments of child behaviour, and consistent coding of parent-child interactions, by independent, blinded-to-condition assessors [[17](#_ENREF_17), [32](#_ENREF_32)].

A broader range of outcomes should be considered that relate to goals that are important to autistic people[[3]](#footnote-3). There has been a call for a shift away from outcomes relating to a reduction of ‘symptoms’, a term associated with a ‘deficit model of disability’ as opposed to a ‘social disability model’ which sees people as being disabled by barriers in society, not by their differences [[19](#_ENREF_19)]. Care should be taken to avoid the use of outcome measures that seek to eliminate autistic traits that are stigmatised but not harmful [43]. Instead, there should be a focus on autistic-preferred goals relating to quality of life, well-being, participation in education and employment, meaningful relationships, and living skills [[27](#_ENREF_27), [47](#Ref_47)]. Dawson and colleagues (2022) describe such goals as “neurodiversity-affirming”, observing that:

“…an optimal outcome for any autism intervention can be defined as an enhanced quality of life which for most people means living as independently as possible, making choices about one’s own life, developing satisfying social relationships, communicating one’s needs and desires, and applying one’s talents and interests in a meaningful and productive manner.” (p. E1) [[50](#_ENREF_50)]

As caregiver stress and well-being can impact on children, and vice-versa, it is also important that parental outcomes are assessed to ensure family-wide benefit of interventions mediated by parents [[42](#_ENREF_42)]. Assessing a broader range of parent characteristics as potential moderating factors is recommended, including parental expectations of treatment, parental self-efficacy, and the parent/therapist alliance [25]. Costs of interventions, and adverse effects, should also be routinely measured [[17](#_ENREF_17)].

Consistent methods to measure and report dose of an intervention are needed to determine the minimum amount of intervention required to achieve desired outcomes, while also considering caregiver burden associated with intervention delivery [[26](#_ENREF_26)].

Finally, longer follow-up assessment periods are needed to see whether benefits are maintained over time, ensuring skills and strategies are generalised to new situations and contexts. It is also important to evaluate whether negative impacts may emerge in later years.

Moderating effects

A greater body of high quality research including larger sample sizes will permit more meaningful subgroup analyses, including meta-regression analyses to consider moderating effects. This enables the investigation of critical questions including “why, when, and for whom” improvements are observed.

Trembath et al., (2019) [[27](#_ENREF_27)] has called for a change in the focus of research in parent-mediated approaches from effectiveness to one that prioritises the investigation of moderating effects:

“It is likely that increased precision in parent-mediated research will further reveal an emerging picture of individual variability in child and parent outcomes. Thus, it is timely that systematic reviews move beyond examining the effectiveness of interventions, to consider factors that may influence outcomes as well as the generalizability of findings to the broader clinical population” (p. 1305).

Moderator research can systematically investigate what programme features may be more effective, and more necessary, than others, with respect to support type, theoretical approach, setting, components, and overall dose (frequency, intensity, duration). Such work will inform what key elements should be included, and what may be left out.

Greater investigation of theory-driven predictors is warranted to identify characteristics of autistic children (and family members) a specific programme or approach is most likely to be effective for, and potential target outcomes for its success (e.g., foundation skills) [[17](#_ENREF_17), [27](#_ENREF_27)]. There is also a need to investigate the impact of contextual and cultural factors which may act as mediating variables for support effectiveness.

2.6 Conclusions

Overview

This systematic review of research relating to parent-mediated supports for autistic children and young people updates evidence for the Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau (2022) [[1](#Ref_1)].

A comprehensive search of peer-reviewed studies published since 2004 was undertaken relevant to the review scope. Studies were restricted to secondary evidence published since 2013 with at least one original eligible randomised controlled trial. Eight peer reviewed secondary studies were included in the umbrella review: three dedicated systematic reviews and five meta-analyses. Six reviews evaluated the effectiveness of parent/family-mediated approaches, and two examined mediating or moderating factors that may impact on intervention effectiveness. There was significant overlap between studies included in the eight reviews with the number of original RCTs ranging from one to seven trials.

The reviews appraised were of moderately good quality in their methodology and reporting. The quality of evidence of primary studies in this field has improved [[26](#_ENREF_26)] has improved over the decade since the Cochrane review [[17](#_ENREF_17)] was conducted. However the methodological limitations of the primary studies included in these reviews remain significant (see [Section 2.4](#Sec2_4)), particularly with respect to lack of controlled group studies, small samples, and lack of blinding of family members as informants in outcome assessments. These issues are reflected in the relatively low ‘strength of evidence’ quality ratings applied (using GRADE) for many outcomes in the appraised reviews (where reported).

Synthesising the current evidence base across these studies is also challenging given the heterogeneity of studies considered in the eight reviews with respect to family-mediated approaches being considered, agent of delivery, intensity/dose of intervention, therapy goals, measures employed, informants used, and the characteristics of the participants, particularly their age.

For these reasons, conclusions should be treated with caution.

Key findings

For ease of presentation, outcomes reported below are organised within broad domains, as guided by those used in Oono et al’s (2013) [[17](#_ENREF_17)] Cochrane review: child language and social communication, child behaviour, adaptive functioning and development, and parent outcomes. However as discussed previously, allocation of outcomes within these domains is somewhat subjective. It is also problematic when outcomes are reported which are not considered appropriate from a neurodiversity perspective,  alongside those that are. Care should therefore be taken when reporting findings across different outcomes.

Further, reporting of outcomes investigated in the appraised literature should not be taken as an endorsement of their suitability or acceptability by the systematic review’s author or the Living Guideline Group.

Language

Considering communication and language as broad outcomes, findings across three meta-analyses (MA) included no improvement, possible improvement, and for the largest review of 13 trials, a small improvement [[26](#_ENREF_26)].

Smaller numbers of trials (n=2–5) from two meta-analyses considered receptive and expressive language separately. Whilst the Cochrane (2013) review [[17](#_ENREF_17)] of young children found possible improvement for both receptive and expressive language, a review including a greater number of studies found relatively no improvement for recipients of PMI for receptive language, but a moderate improvement in their expressive language.

These results suggest that there is emerging evidence for small improvements evident for language as a broad outcome, with moderate improvement in expressive language found for children receiving parent-mediated approaches.

Social communication and child-parent relationship

The overlapping domain of social communication had variable results. In studies of young children, there was evidence of no or small improvement reported for the specific skills of joint attention, and initiation. By contrast, moderate improvement was reported in one trial relating to imitation and play in young children.

Considering social communication/socialisation more broadly, three reviews of young children found evidence of possible improvement, some improvement, or – in the largest review of 13 trials [26] – small improvement for children receiving parent-mediated approaches. However, stronger effects were reported in a meta-analysis of studies including children of a broader age range (2-17 years) [25]. It found moderate improvement in social skills based on 10 studies, and large improvement in engagement across five studies.

Evidence from two reviews suggest a moderate to large improvement for the parent-child relationship (measuring interaction or synchrony between a parent and their autistic child) for parent-mediated approaches’ group participants.

Child behavioural outcomes

Evidence across four reviews found small to moderate improvement in autism core characteristics, including reduced ‘symptom severity’ scale scores, and repetitive behaviour. One of the most recent reviews [[43](#_ENREF_43)] found improvement for clinician-rated assessments (which tend to be blinded), and not for parent-rated ones.

From recent meta-analyses, there was consistent evidence of moderately large effects of parent-mediated approaches on reducing maladaptive behaviour. A moderate to large reduction was observed for disruptive behaviour indicated by 7-9 studies across two reviews [[42](#_ENREF_42), [43](#_ENREF_43)], one of which related only to behaviourally based interventions [[42](#_ENREF_42)]. Small numbers of studies suggested parent-mediated approaches led to moderate improvement in child distress and self-regulation, and a small reduction in hyperactivity.

Adaptive functioning and development

Based on the most recent and largest meta-analysis [[43](#_ENREF_43)], there may be a small improvement in the adaptive functioning of autistic children and young people based on parent ratings, although these may be affected by reporting biases given they tend to be unblinded to condition. No improvements to adaptive functioning were found for assessments that were clinician-rated in this review, or for any assessments in two earlier reviews.

There was no evidence that parent-mediated approaches improved visual reception, or motor skills outcomes. However there is emerging evidence of a small improvement of parent-mediated approaches in autistic children’s cognition.

Parent outcomes

There is consistent evidence from a good number of studies across different reviews that parent-mediated approaches can lead to modest positive benefits for parents, with small improvements in parents’ levels of stress, distress, and self-efficacy. Whilst improving outcomes for children and young people on the autism spectrum should remain the priority, evidence for benefits for the caregiver and wider family/whānau are also important. Benefits may go in both directions, as reduced stress and increased well-being of parents may contribute to better family functioning and outcomes for autistic family members also [[32](#_ENREF_32)].

Mediating and moderating factors

For most or all outcomes, intervention effectiveness did not significantly vary as a function of intervention dose/intensity, outcome targeted, modality or setting, or delivery agent. Findings tended to be mixed for the impact of parent variables on intervention effectiveness, suggesting that whilst having the potential to act as moderators, differences in the intervention or assessment tools employed may be more influential. The ability of sub-group analyses to investigate mediating or moderating factors were limited by low numbers of studies and sample sizes.

Suggestions for improving the methodology and design of future research and addressing research gaps are presented in [Section 2.5](#Sec2_5).

Relevance and applicability to Aotearoa New Zealand

Four qualitative studies (reported in 3 reports) were identified which were conducted in New Zealand which implemented parent-mediated approaches targeting behavioural and language outcomes. These suggest generally high acceptability of these programmes. Alignment of approaches with a parent’s parenting style, culture, and connection with the agency/trainer/therapist appeared to be important factors in acceptability. The research suggests ways programmes can be tailored to the needs of the community, including relationship-building, targeted promotion, addressing practical barriers to participation, and employing culturally-specific adaptations to align with social norms and language.

Conclusions

This umbrella review indicates that parent–mediated approaches are a source of intense and growing research interest over the last decade. There is evidence of improvement in a range of language and behavioural outcomes. Strongest effects were evidence for the benefit of parent-mediated approaches in improving expressive language, promoting social skills and engagement, and improving the interactive relationship between parent and child. Parent-mediated approaches may be particularly effective reducing a child or young person’s behaviours of concern and distress. There also appear to be modest improvements for parents’ own well-being. It may be suggested that improving the relationship between the parent and child leads the parent to understand their child’s needs better, supports them in communicating more effectively, and therefore assists in reducing the child’s behaviours of concern. These improvements to communication and behaviour may reduce distress for both parent and child. Parent-mediated approaches provide avenues to guide and support parents in playing and communicating with their child, and understanding and responding to their child’s needs appropriately. Professionals and therapists should be encouraged to work alongside parents in promoting these goals.

Factors that may predict, mediate or moderate parent-mediated approaches’ effectiveness were investigated in sub-group analyses and suggested no clear or consistent patterns. It is not possible to offer specific recommendations about the most successful and necessary components and intensity of approaches.

The Guideline [[1](#Ref_1)] advocates for a shift in language around autism that is strength-based rather than pathologizing (see the [Autistic Perspectives](#Preamble_AutisticPreferences) section in the current Paper’s Preamble). Whilst the current umbrella review describes research outcomes that have been targeted and reported in the international literature, this should not be considered an uncritical endorsement of the choice of those goals, or the often offensive and diminishing language sometimes used in describing scales and outcomes (for example, reports of autism ‘symptoms’). Goals of reducing autistic core characteristics as primary outcomes do not value neurodiversity. Furthermore, ‘interventions’ that are designed to achieve such goals may actually encourage masking or camouflaging of autistic traits, behaviours which have been associated with increased anxiety and depression [56]. Researchers should consider the needs and values of autistic people and their whānau in setting programme goals, designing research studies, selecting appropriate outcome measures, and reporting findings.

Part 3: Recommendation development

The Living Guideline Group ([LGG](#LGG)) were tasked with considering the systematically reviewed evidence reported in [Part 2](#Part2). Specifically, whether the updated body of evidence required revisions to the Guideline’s existing Recommendations and Good Practice Points (GPP) as well as the development of new ones.

The text and graded ‘strength of evidence’ (see Appendix 1, [Table A1.3](#TableA1_3)) of any potential new Recommendations were considered at an all-day face-to-face meeting. The LGG’s decisions are presented below and summarised in the [Summary Table](#SummTable). Where considered helpful, these decisions are accompanied by additional explanatory text, and/or with a brief rationale which highlights any particular issues that the LGG took into account in their deliberations.

Decisions of the Living Guideline Group

Preamble

The update topic considered by the LGG was parent-mediated approaches (also known as Parent Mediated Interventions or PMI). These are skill-focused programmes where the parent or caregiver takes on the role of specialist/therapist in delivering the approach to their autistic child. In parent-mediated approaches, the professional specialist/therapist is involved in training the parent or family member in how to deliver the approach/programme, as well as having an ongoing role in coaching and mentoring them. Key components of parent-mediated approaches include: modelling of strategies, parent coaching, provision of feedback, and individualization of strategies.

In this ‘umbrella review’ (review of reviews), eight systematic reviews evaluating effectiveness and/or mediating factors on effectiveness of parent-mediated approaches were identified as eligible for inclusion, five of which also included meta-analyses. Evaluations of how best to deliver the training itself were not the focus of the current review.

There was a wide range of approaches and outcomes represented in the reviewed research. Parent-mediated approaches included comprehensive programmes for improvement or target specific areas/skills (e.g., joint attention, language, play) or specific areas of concern (e.g., behaviours of concern, insomnia, feeding, toileting). They can seek to improve quality of life and well-being for the autistic individual, as well as for parents and the wider family unit.

It is not currently possible to offer specific recommendations about the most successful and necessary components and intensity of approaches/'interventions' delivered by parents/caregivers to autistic children and young people.

No New-Zealand based research assessing effectiveness was included in any of the appraised reviews. However, in order to consider the relevance and applicability of the international evidence to Aotearoa’s culture and service systems, qualitative and survey-based research was identified which assessed the direct experiences of New Zealand-based service recipients. These support the use of parent-mediated supportive approaches as acceptable but advise on adaptions to reflect cultural preferences and reduce barriers to access.

New Recommendation

**New Recommendation 2.1.5a:** Parent-mediated approaches (also known as ‘parent mediated interventions’) should be considered by, and provided for, parents/caregivers of autistic children. There is strong evidence that parent-mediated approaches improve the relationship between children and their parents/caregivers, and provide a range of benefits [Grade A].

**Rationale**: The evidence suggests that parent-mediated approaches lead to moderate to large improvement in the parent-child relationship; moderate improvement in social skills; large improvement in engagement; moderate improvement in expressive language; moderately large reduction in behaviours of concern (‘maladaptive behaviour’), and moderate improvement in child distress and self-regulation. Improving the relationship between the parent/caregiver and child may lead the parent to understand their child’s needs better, supporting them to communicate more effectively, and as a consequence, reducing the child’s behaviours of concern as well as the distress of both parent/caregiver and child.

**Additional text:** Cross reference to GPP 4.3.13 relating to ethical practice (see over page).

New Good Practice Points

**New Good Practice Point 2.1.5b.** The design of parent-mediated approaches (also known as ‘parent mediated interventions’) should be culturally responsive, whānau-led, holistic, mana-enhancing, respectful, easy to use, and tailored to the autistic individual and whānau needs.

**Rationale:** Recommendations should reflect Autistic perspectives, Māori perspectives[[4]](#footnote-4), and be consistent with Enabling Good Lives (https://www.enablinggoodlives.co.nz) principles. These are discussed in the Guideline’s Overview [[1](#Ref_1)].

**New Good Practice Point 2.1.5c.** Parents/caregivers should determine in what way and how much they are involved in the delivery of supports

**New Good Practice Point 2.1.5d.** Practitioners should be aware that parents/caregivers may perceive that their involvement in the delivery of supports detracts from their natural parenting/caregiving role.

**Rationale:** The Australian National Guideline for supporting autistic children (2022) [[57]](#Ref_57) was released outside the search period for the current systematic review. However it was identified during consultation for this supplementary paper as providing two relevant Good Practice Points (47.4 and 47.5). These relate to issues discussed by the LGG in its consideration of evidence and are endorsed by them. They have been adapted for inclusion in the New Zealand guideline.

The [Summary Table](#SummTable) of the [Executive Summary](#ExecSumm) summarises the new Recommendations and Good Practice Points.

|  |  |
| --- | --- |
| **Good Practice Point 4.3.13:** In choosing a supportive approach, strategy, practitioner or therapist, the following principles for ethical practice are recommended:   * **Accept the person as authentically autistic**. Respect neurodivergence as difference that does not need to be cured. * **Be strengths-based**. Identify and work with an autistic person’s strengths, abilities and potential. * **Be person-centred**. Focus on the needs and autonomy of the autistic person. Consider their culture, needs and choices in identifying an approach and its goals. * **Avoid encouraging masking** of a person’s autism/takiwātanga. Do not target reducing behaviours (such as ‘stims’) which are not harmful or a barrier to desired goals (e.g., do not demand eye contact). * **Understand that behaviour is communication** and where harmful or ‘challenging’, focus on understanding its purpose and achieving positive change through alternative ways. * **Identify, encourage and facilitate access** to use of supports, modifications, and adjustments (rather than getting the autistic person to do all the changing) * **Presume competence and potential**. Assume an autistic person has the capacity to think, learn, and understand regardless of how they communicate. * **Provide access** to communication modalities that facilitate an autistic person’s ability to process and express (e.g., augmentative and alternative communication (AAC) devices, signing, quiet space) * **Do not use** seclusion and restraint, or aversive practices (*see also* [*Recommendation 3.2.5.3*](#Rec3_2_5_3)) * **Be collaborative**. Work alongside supportive family, carers, and professional providers, therapists, and educators * **Regularly assess consent**. Look for signs of disinterest, disengagement, or distress and consider reducing intensity, taking a break, or ceasing an approach altogether. * **Monitor progress** regularly (*see also* [*Recommendation 4.3.3*](#Rec4_3_3)). * **Commit to the dignity, civil liberties and human** **rights** of people served. Comply with United Nations Convention on the Rights of Persons with Disabilities (2008)and the United Nations Convention on the Rights of Children (1989) | ✓ |

Appendix 1: Methods

A1.1 Acknowledgements

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Ex-officio LGG members

Ex-officio members include the programme funders and sponsors.

**Donna Caddie**, Manager; Investment and Strategic Design – Learning Support, Te Pae Aronui, Ministry of Education

**Helen Hayes**, Portfolio Manager; Whaikaha – Ministry of Disabled People.

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The Ministry of Education Library provided access to databases and assisted with inter-loan retrieval. **Margaret Paterson** (Information Specialist, University of Canterbury) provided informal peer review of the search strategy.

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Declarations of interest

None

A1.2 Search strategy

Search strategies were limited to publications from January 1 2004 onwards. Database searches were conducted on 12 July 2022. Full search strategies are available upon request.

Bibliographic, health technology assessment and guideline databases were included in the search strategy, listed below.

Databases

* Medline (EBSCO-Host)
* EMBASE (OVID)
* Cinahl (EBSCO-Host)
* PsycInfo (EBSCO-Host)
* APA PsycArticles (EBSCO-Host)
* SocINDEX (EBSCO-Host)
* ERIC (EBSCO-Host)
* Education Research Complete (EBSCO-Host)
* nzresearch.org.nz
* Cochrane Database of Systematic Reviews (Cochrane Library website)

Search terms

A combination of search terms were developed and adapted for different databases.

Searches were initially limited to those: published in the English language, involving human participants, and published in peer-reviewed Journals since January 2004. Dissertations were excluded but letters, corrections, and retracted articles were retained in the search as potentially relevant to included studies. Reference lists of retrieved publications were also examined to identify any additional eligible studies. Hand searching of journals and contacting of authors for unpublished research were not undertaken. Authors were contacted for clarification where needed.

The following illustrative search syntax is offered for searches conducted using OVID:

1. (autism or autism spectrum disorder).sh. OR (autism or autistic or asperger or aspergers or pervasive developmental disorder or ASD or PDD).ti. OR (autism or autistic or asperger or aspergers or pervasive developmental disorder or ASD or PDD).ab.
2. (family or parents or caregivers).sh. OR (parent or parents or family or families or whanau or mother or mothers or father or fathers or maternal or paternal or carer or carers or care-giver or care-givers or sibling or siblings or at home or home-based or parent-implemented or parent-training or parent-coaching or parent-mediated or parent-teaching or sibling-implemented or sibling-training or sibling-coaching or sibling-mediated or sibling-teaching).ti. OR (parent or parents or family or families or whanau or mother or mothers or father or fathers or maternal or paternal or carer or carers or care-giver or care-givers or sibling or siblings or at home or home-based or parent-implemented or parent-training or parent-coaching or parent-mediated or parent-teaching or sibling-implemented or sibling-training or sibling-coaching or sibling-mediated or sibling-teaching).ab.
3. (systematic review or meta-analysis or meta analysis or meta-analytic or meta analytic or review or guideline).sh OR (systematic review or meta-analysis or meta analysis or meta-analytic or meta analytic or review or guideline).ti.
4. #1 AND #2 AND #3
5. Limit 4 to (human AND English language AND yr=’2004-Current” AND Journal) AND (article OR article in press OR editorial OR erratum OR letter OR preprint OR review)

To check whether the English language exclusion was likely to have missed eligible articles, a follow-up search was undertaken on 21 July 2022 which included all languages excepting those published in English. The search strategy included the original search terms and databases, however the publication dates were limited to those published since 2013 to represent “best evidence” (the year the Oono et al., [[17](#_ENREF_17)] Cochrane review of parent-mediated early interventions was published).

The search strategy identified 9 articles not published in English. None of these met select criteria for the current overview of systematic reviews.

New Zealand-based qualitative research

To consider the relevance and applicability of the international systematic review of effectiveness to Aotearoa’s cultural and service context, a second search was conducted. Eligible studies were those reporting on the first-hand experiences of parent-mediated approaches by New Zealand-based autistic children.

The search strategy employed for the umbrella review was adapted to remove the methodology filter (search #3), and to add a search for “New Zealand” in the search fields for Title, Abstract, or Author Affiliation (which identified New Zealand-based Universities or organisations). The search was limited to peer-reviewed Journal articles published in or since 2004. Members of the LGG also supplied potentially relevant material.

Selection criteria considered qualitative studies and surveys assessing the relevance, applicability, acceptability and feasibility of these approaches within a New Zealand culture and service context, including where these reflect user preferences and values.

Excluded were non peer-reviewed articles; case studies; autobiographical accounts; and qualitative measures of perceived intervention effectiveness where a quantitative approach would have been more appropriate.

This strategy identified 131 unique abstracts, ten of which were retrieved as full text as being potentially eligible, and three of which met criteria for inclusion. These are summarised in [Section 1.3](#A1_3_NZ) in the Background. Only eligible outcomes were reported.

A1.3 Study selection

A single (highly experienced) autism researcher performed study selection, data extraction, critical appraisal, and synthesis. The search strategy was peer reviewed by an independent Information Specialist.

Levels of evidence

Research study designs are broadly associated with particular methodological strengths and limitations in terms of how bias is minimised. This allows studies to be assigned a “level of evidence” within an evidence hierarchy [[40](#_ENREF_40)], so as to rank them in terms of quality from most robust (level I) to least (level IV).

The best study design is determined by the review question. The hierarchy of study designs that best answer questions of effectiveness are presented in [Table A1.1](#TableA1_1).

Level I evidence is represented by systematic reviews and meta analyses including at least one level II study, a randomised controlled trial. Systematic reviews of lower order evidence rank at the same level as that order of evidence.

The primary goal of the review was to determine whether an intervention is effective. Consistent with the evidence-based practice model, this question is most robustly answered using study designs that appear higher in the evidence hierarchy ([Table A1.1](#TableA1_1)) and a lower risk of bias, such as well conducted, controlled experimental studies. Only in their absence is lower order evidence included.

Using this principle, recently published systematic reviews considering studies that represent a subset of interventions within the broader scope can be considered “best evidence”. In this ‘umbrella review’, primary studies were not individually appraised.

Note that New Zealand-based studies considered in [Section 1.3](#A1_3_NZ) were identified to reflect user experiences including relevance, applicability, acceptability, and feasibility, including their preferences and values. To answer this research question, the best study designs employ qualitative or survey designs.

Table A1.1: Hierarchy of evidence for effectiveness studies

|  |  |
| --- | --- |
| **Level** | **Intervention** |
| I | A systematic review of level II studies |
| II | A randomised controlled trial |
| III-1 | A pseudo-randomised controlled trial  (i.e., alternate allocation or some other method) |
| III-2 | A comparative study with concurrent controls:  - Non-randomised experimental trial  - Cohort study  - Case-control study  - Interrupted time series with a control group |
| III-3 | A comparative study without concurrent controls:  - Historical control study  - Two or more single arm study  - Interrupted time series without a parallel control group |
| IV | Case series with either post-test or before-and-after (pre-test/post-test) outcomes. |

**Source**: NHMRC [[40](#_ENREF_40)]

Criteria for systematic reviews

In the current review, only systematic reviews published in or since 2013 were considered.

The criteria used to classify systematic reviews was developed by Salvador-Oliván et al., (2021) [[51](#_ENREF_51)] and included the following:

1. The title or abstract describes the article as a systematic review.
2. The methods section indicates that a systematic literature search was performed, specifies the databases searched, and presents a search strategy or search terms.
3. The criteria for the inclusion/exclusion of a study are specified.
4. A PRISMA [[39](#_ENREF_39)] flow diagram illustrating the process for selecting studies for analysis is included (or described).
5. A meta-analysis is conducted for data synthesis or a qualitative synthesis of the evidence from the studies included in the review is available.
6. The article presents the following: search strategy including information sources/databases and search terms; criteria for study selection; data extraction method; synthesis of data or evidence; and conclusions.

An article was classified as systematic review only if one of the first two criteria (a or b) and one of the last four criteria (c, d, e, or f) were met. Reviews were excluded if the study selection criteria or conclusions were based on authors’ personal and subjective experiences.

A1.4 Data extraction

Study characteristics were extracted for each of the appraised studies. Key features recorded for primary studies included:

* citation
* critical appraisal quality score (see Section A1.5 below)
* aim of the review; type of review
* participant details; setting/context
* search strategy (number of databases searched, date range)
* eligible interventions; included studies (number, type, country of origin)
* critical appraisal tool used and ratings; comparisons/analyses for outcomes relevant to umbrella review (including effect sizes where reported)
* method of synthesis/analysis employed
* key conclusions
* comments/notes from (umbrella review) reviewer
* funding body

Evidence tables for secondary studies included the review search strategy and methodology (see [Appendix 3](#App3)).

A1.5 Critical appraisal

In addition to the level of evidence associated with a study design, the quality of how the study is conducted can be assessed using critical appraisal tools.

Studies were assessed for study quality using the Joanna Briggs Institute (JBI) checklist for systematic reviews and research synthesis [[38](#_ENREF_38)]. Each of the 11 questions posed in the checklist can be scored as being ‘met’, ‘not met’, ‘unclear’ (or rarely, ‘not applicable’). Items are presented in [Table A1.2](#TableA1_2).

Applying a strategy used for an autism umbrella review [[52](#_ENREF_52)], each item was scored 1 if “met” with scores summed to categorise study quality as follows: low (score 0-5), medium (score 6-8), or high (score 9–11). Reviews rated as being of low quality were excluded.

Table A1.2: Critical appraisal checklist for systematic reviews and research synthesis

|  |
| --- |
| 1. Is the review question clearly and explicitly stated? |
| 1. Were the inclusion criteria appropriate for the review question? |
| 1. Was the search strategy appropriate? |
| 1. Were the sources and resources used to search for studies adequate? |
| 1. Were the criteria for appraising studies appropriate? |
| 1. Was critical appraisal conducted by two or more reviewers independently? |
| 1. Were there methods to minimize errors in data extraction? |
| 1. Were the methods used to combine studies appropriate? |
| 1. Was the likelihood of publication bias assessed? |
| 1. Were recommendations for policy and/or practice supported by the reported data? |
| 1. Were the specific directives for new research appropriate? |

**Source**: Joanna Briggs Institute [[38](#_ENREF_38)]

A1.6 Preparing Recommendations

A one-day face-to-face meeting was held on 18 November 2022 where the Living Guideline Group considered the findings of the current systematic review. Using their collective professional judgement and experience, as well as the New Zealand-based qualitative research summarised in the Background, the LGG discussed the body of evidence with respect to the research question and the applicability of the evidence within Aotearoa New Zealand. They considered (any) existing affected Recommendations and Good Practice Points from the Guideline [[15](#_ENREF_15)] and the development of new ones.

Developing Recommendations involves consideration of the whole evidence base for the research question. The quality and consistency of the evidence and the practice implications within an Aotearoa New Zealand context is weighed up by all the LGG members. The grades of Recommendations used by the LGG, and also used in the Guideline [[15](#_ENREF_15)], are presented in [Table A1.3](#TableA1_3).

Each Recommendation is assigned a grade to indicate the overall ‘strength of the evidence’ upon which it is based. Strength of the body of evidence is determined across three domains [[40](#_ENREF_40)]:

* quality (the extent to which bias was minimised as determined by study design and the conduct of the study)
* quantity (magnitude of effect, numbers of studies, sample size or power)
* consistency (the extent to which similar findings are reported.

The wording of Recommendations and Good Practice Points, and the evidence grade, is determined by the LGG through discussion and group consensus during the meeting.

Table A1.3: Grades for Recommendations

|  |  |
| --- | --- |
| Recommendations | Grade |
| The Recommendation is supported by good evidence (based on a number of studies that are valid, consistent, applicable and clinically relevant) | A |
| The Recommendation is supported by fair evidence (based on studies that are valid, but there are some concerns about the volume, consistency, applicability and clinical relevance of the evidence that may cause some uncertainty but are not likely to be overturned by other evidence) | B |
| The Recommendation is supported by international expert opinion | C |
| The evidence is insufficient, evidence is lacking, of poor quality or opinions conflicting, the balance of benefits and harms cannot be determined | I |
| Good practice point | Grade |
| Where a Recommendation is based on the clinical and educational experiences of members of the Living Guideline Group, or feedback from consultation within Aotearoa New Zealand, it is a Good Practice Point. | ✓ |

A1.7 Consultation

Seeking comments from stakeholders is vital for peer-review and quality assurance processes in developing the report. In a focused consultation, 11 key stakeholder organisations/individuals were approached for feedback on a late draft of the report. Particular attention was sought regarding the relevance of the report to Aotearoa New Zealand’s services and needs, clarity and ease of use of the report, and implementability of the revised or new Recommendations and Good Practice Points.

Eight of eleven organisations responded to the invitation, five of which completed the consultation survey: Altogether Autism, Autism New Zealand, Explore Specialist Advice, the New Zealand Psychological Society, and Victoria University of Wellington’s Autism Centre. The Autism/Takiwātanga Living Guideline Manager (INSiGHT Research) collated feedback and drafted revisions for the Living Guideline Group to consider. Suggestions identified in the consultation led to several improvements to the final report. INSIGHT Research and the LGG are grateful to those individuals and organisations who participated in the consultation process.

Appendix 2: Abbreviations and glossaries

A2.1 Abbreviations and acronyms

Miscellaneous Terms

ANOVA analysis of variance

ASD Autism Spectrum Disorder

CBT cognitive behaviour therapy

cf compared with (contraction of the Latin *conferatur*)

GPP Good Practice Point

IQ intelligence quotient

INSIGHT Research INdependent Specialist in Guidelines & Health Technology Research

JBI Joanna Briggs Institute

LGG Living Guideline Group

mth month

M mean

MD mean difference

NA not applicable

N (or n) number (usually, sample size)

ns not significant

NHMRC National Health and Medical Research Council (Australia)

NR not reported

NZ New Zealand/Aotearoa

OT occupational therapist

PDD Pervasive Developmental Disorder

PDD-NOS Pervasive Developmental Disorder – Not Otherwise Specified

PICO Participants, Intervention, Comparator, Outcomes

RCT randomised controlled trial

SCED single case experimental design

SMD standardised mean difference

UK United Kingdom

US United States of America

/wk per week

WMD weighted mean difference

Tests, scales and measures

ABC(L) Aberrant Behavior Checklist

ADOS Autism Diagnostic Observation Schedule

AIR-SD American Institutes for Research Self-Determination Scale

BDI Beck Depression Inventory

CCAPS-34 Counseling Center Assessment of Psychological Symptoms-34 Scale

CGI-S/I Clinical Global Impression – Severity scale/Improvement scale

DERS Difficulties in Emotional Regulation Scale

DSM-IV-TR Diagnostic and Statistical Manual of Mental Disorders - IV (text revision)

DSM5 Diagnostic and Statistical Manual of Mental Disorders – 5th edition

GRADE Grading of Recommendations, Assessment, Development and Evaluations

RCI Reliable Change Indices

RSES Rosenberg Self Esteem Inventory Scale

SACQ Student Adaptation to College Questionnaire

SRS Social Responsiveness Scale

STAI State Trait Anxiety Inventory

UCLALS UCLA Loneliness Scale

WAIS-II Wechsler Abbreviated Scale of Intelligence–Second Edition

Databases

CDSR Cochrane Database of Systematic Reviews

CENTRAL Cochrane Central Register of Controlled Trial

CINAHL Cumulative Index to Nursing and Allied Health Literature

DARE Database of Abstracts of Reviews of Effects

EMBASE Excepta Medica Database

ERIC Education Resources Information Centre

Medline Medical Literature Analysis and Retrieval System Online

PsycINFO Psychology Information Database

A2.2 Glossary of terms

**Accommodations**

Adaptations that remove barriers to enable equal participation. These are based on the premise that students with disabilities should be neither disadvantaged nor advantaged relative to other students. Students can be treated differently if it is achieving equity.

**Before-and-after study**

Case series where measures on an outcome are taken before-and-after the intervention is introduced to a series of people and are then compared (also known as a ‘pre-test/post-test study’).

**Bias**

Bias is a systematic deviation of a measurement from the ‘true’ value leading to either an over- or under-estimation of the treatment effect. Bias can originate from many different sources, such as allocation of patients, measurement, interpretation, publication and review of data.

**Blinding**

The concealment of group assignment - to either the treatment or control group - from the knowledge of study participants, assessors, and/or investigators in a clinical trial of whether an intervention/treatment being administered is a placebo treatment (i.e., the control group) or the intervention/treatment being investigated.

**Case series**

Case series are collections of individual case reports, which may occur within a fairly short period of time. Cases consist of either only the exposed people with the outcomes, or people with the outcome regardless of the exposure. In neither of these examples can the risk for the outcome be determined

**Cross-sectional study**

A study that examines the relationship between exposures (e.g., risk factor) and outcomes (e.g., disease), as they exist in a defined population, at a particular time. A group of people are assessed at a particular point (or cross-section) in time and the data collected on outcomes relate to that point in time; i.e., proportion of people with asthma in October 2019. This type of study is useful for hypothesis-generation, to identify whether a risk factor is associated with a certain type of outcome, but more often than not (except when the exposure and outcome are stable; e.g., genetic mutation and certain clinical symptoms) the causal link cannot be proven unless a time dimension is included.

**Detection bias**

Detection bias refers to systematic differences between groups in how outcomes are determined. Awareness by outcome assessors/respondents of whether an intervention was received or not (i.e., they are not blind to allocated condition) may increase the risk of their measurements/ratings/reports being affected by detection bias.

**Effect size**

A quantitative measure of the strength of a phenomenon, a standardised measure of the size of the difference between two groups. The effect sizes can be interpreted in accordance with common guidelines for interventions in the behavioural sciences where effect sizes of up to 0.2 are considered small, those around 0.5 are moderate, and those at 0.8 and above are large [[53](#_ENREF_53)].

**Effectiveness**

A measure of the extent to which a specific intervention, procedure, regimen, or service, when deployed in the field in routine circumstances, does what it is intended to do for a specified population.

**Generalisability**

Applicability of the results to other populations.

Grading of Recommendations, Assessment, Development and Evaluations (GRADE)

GRADE (Grading of Recommendations, Assessment, Development and Evaluations) [[44](#_ENREF_44)] is a transparent framework for developing and presenting summaries of evidence and provides a systematic approach for making clinical practice recommendation. An effect size is estimated (from a systematic review and/or meta-analysis) and then the quality of evidence is estimated for each outcome. An overall GRADE quality rating can be applied to a body of evidence across outcomes, usually by taking the lowest quality of evidence from all of the outcomes that are critical to decision making.]

GRADE has four levels of evidence – also known as certainty in evidence or quality of evidence: very low, low, moderate, and high. Evidence from randomised controlled trials starts at high quality and, because of residual confounding, evidence that includes observational data starts at low quality.

The certainty in the evidence can be decreased based on the following methodological factors: risk of bias, imprecision, inconsistency, indirectness, and publication bias. In contrast, the certainty in the evidence can be increased based on a large magnitude of effect, and dose-response gradient.

**Level of evidence**

Levels within a hierarchy of study evidence that indicates the degree to which bias has been eliminated in the study design. For example, see **Appendix 1,** [Table A1.1](#TableA1_1).

**Mean**

Calculated by adding all the individual values in the group and dividing by the number of values in the group.

**Mean difference (MD)**

In randomized controlled trials (RCTs), change scores (between endpoint and baseline) are values compared between experimental and control groups, yielding a mean difference for each outcome. In a meta-analysis, if outcomes are all in the same unit (such as when they were all obtained using the same rating instrument), the mean difference values for a specified outcome can be pooled to yield a summary estimate that is also known as a mean difference (MD). Because pooling of the mean difference from individual RCTs is done after weighting the values for precision, this pooled MD is also known as the weighted mean difference (WMD).

Note: Where outcomes between studies use different units of measurement, see *Standardised Mean Difference.*

**Meta analysis**

Meta-analysis is the use of statistical techniques to combine and summarize the results of multiple studies; they may or may be contained within a systematic review. By combining data from several studies, meta-analyses can provide more precise estimates of the effects of health care than those derived from the individual studies. See also Mean Difference, and Standardised Mean Difference.

**Neurodiversity**

An approach to learning and disability which suggests that diverse neurological conditions appear as a result of normal variation in the human genome. This term was coined in the late 1990s as a challenge to prevailing views of neurological diversity as inherently pathological, and it asserts that neurological differences should be recognised and respected as a social category on a par with gender, ethnicity, sexual orientation, or disability status.

**Observational studies**

Also known as epidemiological studies. These are usually undertaken by investigators who are not involved in the clinical care of the patients being studied, and who are not using the technology under investigation. Distinct from experimental studies.

**Performance bias**

Performance bias refers to systematic differences between groups in the care that is provided, or in exposure to factors other than the interventions of interest. After enrolment into the study, blinding (or masking) of study participants and personnel may reduce the risk that knowledge of which intervention was received, rather than the intervention itself, affects outcomes. Effective blinding can also ensure that the compared groups receive a similar amount of attention, ancillary treatment and diagnostic investigations. Blinding is not always possible, however.

**PICO framework**

A mnemonic used to frame a research question’s parameters which is useful for developing search strategies and selection criteria in systematic reviews. The letters stand for

**P** – Participant or population

**I** – Intervention

**C** – Comparison, control or comparator

**O** – Outcome(s)

T is sometimes added to represent dates of population or duration of intervention.

**Post-test**

Case series where only outcomes after the intervention (factor under study) are recorded in the series of people, so no comparisons can be made.

**Power**

The probability that a statistical test or study will detect a defined pattern in data and declare the extent of the pattern as showing statistical significance.

**Prevalence**

A measure of the proportion of people in a population who have some attribute or disease at a given point in time or during some time period.

**Pseudo-randomised controlled trial**

As for a randomised controlled trial except that a pseudo-random method (such as alternate allocation, days of the week, date of birth, or odd-even medical record numbers) is described for allocating individuals into treatment or control group conditions. The outcomes from each group are compared. Sometimes known as quasi-randomised controlled trials.

**Quality of evidence**

Degree to which bias has been prevented through the design and conduct of research from which evidence is derived.

**Randomised controlled trial (RCT)**

An experiment in which a unit (e.g., people, or a cluster of people) are allocated using a fully random mechanism (such as a coin toss, random number table, computer-generated random numbers) into either the intervention condition (e.g., preventive or therapeutic procedure, manoeuvre, or treatment) or a control comparison condition (e.g., placebo, usual care, alternative treatment). The outcomes from each group are compared. Conditions are run in parallel.

**Reliable Change Indices (RCI)**

Reliable change indices are used to calculate significance of change at the individual participant level from baseline to midpoint and baseline to endpoint.

**Secondary study**

An analysis or synthesis of research data reported elsewhere, including systematic reviews, meta-analyses and guidelines.

**Selection bias**

Error due to systematic differences in characteristics between those who are selected for inclusion in a study and those who are not (or between those compared within a study and those who are not).

**Single case experimental design (SCED)**

In contrast to an experimental group design in which one group is compared with another, participants in a single-subject experiment research provide their own control data for the purpose of comparison in a within-subject rather than a between-subjects design. SCEDs typically involve a comparison between two experimental time periods, known as phases. This approach typically includes collecting a representative baseline phase to serve as a comparison with subsequent phases.

There are many variants. Multiple baseline/combined series designs consist of a number of repeated, miniature AB experiments or variations thereof. Introduction of the independent variable (intervention) is staggered temporally across multiple participants or across multiple within-subject conditions, which allows the researcher to demonstrate that changes in the dependent variable (outcome) reliably occur only when the independent variable is introduced, thus controlling for the effects of extraneous factors. Multiple baseline designs can be used both within and across units (i.e., persons or groups of persons).

Reversal designs (ABAB designs) involve collecting a baseline measure of the dependent variable (A phase), introducing the independent variable/intervention (B phase), removing the intervention while continuing to assess the dependent variable/outcome (second A phase), and then reintroducing the intervention (second B phase). Reversal designs are useful when the intervention’s effects on the dependent variable/outcome are expected to reverse or discontinue when the independent variable/intervention is not present.

**Strength of evidence**

The strength of evidence for an intervention effect includes the level (type of studies), quality (how well the studies were designed and performed to eliminate bias) and statistical precision (p-value and confidence interval).

**Standardised mean difference (SMD)**

In randomized controlled trials (RCTs), change scores (between endpoint and baseline) are compared between experimental and control groups, yielding a mean difference for each outcome that can be pooled in meta-analysis to yield a summary estimate known as a mean difference (MD). When different studies use different rating instruments to measure the same outcome, the units of measurement varies and the mean differences across RCTs cannot be pooled. However, these mean differences can be divided by their respective standard deviations (SDs) to yield a statistic known as the standardized mean difference (SMD). SMDs can be pooled in meta-analysis because the unit is uniform across studies. Various approaches can be used for determining this and its significance (e.g., Cohen's d, Glass' delta, Hodge’s *g*) and the magnitude of effect can be interpreted as follows: trivial (< 0.2), small (0.02 – 0.49), medium (0.05 – 0.79), and large (≥ 0.8) [[53](#_ENREF_53)]. See also Mean Difference

**Systematic review (SR)**

A systematic review attempts to collate all relevant evidence that fits pre-specified eligibility criteria to answer a specific research question. It uses explicit, systematic methods to minimize bias in the identification, selection, synthesis, and summary of studies. The key characteristics of a systematic review are (a) a clearly stated set of objectives with an explicit, reproducible methodology; (b) a systematic search that attempts to identify all studies that would meet the eligibility criteria; (c) an assessment of the validity of the findings of the included studies (e.g., assessment of risk of bias and confidence in cumulative estimates); and (d) systematic presentation, and synthesis, of the characteristics and findings of the included studies.

A2.3 Glossary of Māori terms[[5]](#footnote-5)

**Aotearoa** New Zealand

**kanorau ā-io** neurodiversity (kanorau =diversity, ā-io=of the nerves), see also *Kanorau ā-roro*

**kanorau ā-roro** neurodiversity (kanorau =diversity, ā-roro=of the brain); see also *Kanorau ā-io*

**mana atua** wellbeing

**rangatahi** young person/people

**takiwātanga** autism (in his/her/their own time and space) [[22](#_ENREF_22)]

**tamaiti** child

**tamariki** children

**tangata whaitakiwātanga** autistic person (see takiwātanga)

**te reo Māori** the Māori language

**tikanga** customs, protocol, rules, principles

**whānau** extended family

**whanaungatanga** kinship, relationship

**whakataukī** Māori proverb

**Whare Tapa Whā** framework of Māori health (four-sided house)

Papers are presented chronologically by year of publication (oldest first), and within each year, alphabetically (by first author‘s surname).

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Oono et al., (2013)** [[17](#_ENREF_17)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| Country: UK  **Study type**: systematic review & meta-analysis  Evidence level: I  **Study Quality**: JBI checklist score: 11/11 (high quality)  **Aims**: effectiveness of parent-mediated approaches for young autistic children aged 1 to < 7 years.  Updates earlier Cochrane Review (2002) [[34](#_ENREF_34)] | **Databases**: CENTRAL, Medline, Embase, PyscINFO, ERIC, CINAHL, and other databases, trials registers, and grey literature sources  **Search**: Searched 2002 – Aug 2012. Transparent selection criteria. Full search terms for keywords described.  Citation searching performed, field experts contacted, key Journals hand-searched.  Observed PRISMA reporting.  Selection criteria:  Parents were trained by professionals in strategies to improve ‘management of child’s ASD-related difficulties’. Received Parent Training in group or individual sessions. Received ongoing supervision and support from professionals.   * ASD diagnosis * aged 1 to < 7 years * RCTs assessing efficacy | **Method**: independent double study screening and selection of retrieved articles, data extraction, critical appraisal using the Cochrane Risk-of-bias tool, and rating ‘Quality of evidence’ using GRADE (very low, low, moderate, and high) are reported here only for meta-analyses involving PMI studies.  Random effects meta-analyses undertaken of 10 studies. Findings from the remaining 7 studies were reported narratively.  Effect sizes presented as standardised mean difference (SMD)  Due to variability in reporting methods and low sample sizes, moderators such as duration, intensity, type of intervention, parental education, child’s age, child’s IQ, and family socio-economic status were not investigated directly in subgroup analyses. | **Included**: 17 articles (n =919 autistic children, aged 17 months – 6 years).  Study designs: All RCTs  Study quality: High risk of bias was evident in the studies in relation to allocation concealment and incomplete outcome data; blinding of participants was not possible. All findings were considered to be ‘low quality’ from GRADE assessments,  Duration: 12 weeks – 2 years  **Key findings** (no. studies) (effect size)   * Severity of autism core characteristic: (n=6) small effect (SMD -0.30) * Child communication and social development * Shared/joint attention (coded interactions) (n=2) small effect (SMD=0.41) * *Language – expression* (clinician): (n=3) NS. But higher scores indicate improvement, small, uncertain effect (SMD 0.14) * *Language, comprehension* (clinician): (n=2) NS. But scores indicate improvement, small, uncertain effect (SMD=0.29) * Language, comprehension (parent-report): (n=3) small effect (MD=36.26) * Communication: (n=3) The mean value was 5.31 points higher for the intervention group. Higher scores indicate improvement. * Social communication (n=3) inconsistent * *Child initiations:* (n=4) non-significant difference * Maladaptive behaviour: (n=4) non-significant difference * Parent outcomes * *Parent-child interaction/parent synchrony* (n=3) large effect (SMD=0.90)- assumed mediator for positive child outcomes * *Parent stress*: (n=2) small, uncertain effect (SMD=-.17) * Child development: (n=5) mixed findings, small gains in some studies but may reflect child cooperation   Adaptive behaviour: (n=2) non-significant difference | Author conclusions:  Parent-mediated early intervention does appear on average to lead to positive changes in parent-child interaction, and to possible gains for children in child language comprehension, with reduction in severity of autism core characteristics. Noted low precision (small effect sizes and wide confidence intervals), as well as high risk of bias leading to low level of certainty/quality of findings, and that conclusions are likely to change with future publication of high-quality RCTs.  Evidence of whether such interventions may reduce parent stress is inconclusive.  **Reviewer’s comments**: Comprehensive search. Gold standard Cochrane review and MA.  Unique studies: 2 of 17 (2 unique RCTs) |

Appendix 3: Evidence Tables of included studies

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Nevill et al., (2018)**  [[26](#_ENREF_26)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| Country: USA  **Study type**: systematic review & meta-analysis  Evidence level: I  **Study Quality**: JBI checklist score: 10/11 (high quality)  **Aims**: effectiveness of parent training for delivering parent-mediated approaches for young autistic children aged 1 –6 years. | **Databases**: PubMed, Medline, PyscINFO, Google Scholar  **Search**: Searched 2000 – Dec 2015. Transparent selection criteria. Full search terms for keywords described.  Citation searching performed. Observed PRISMA reporting.  Selection criteria:  Parents were responsible for implementing the active intervention during the active treatment phase through the assistance of therapist coaching, strategy modelling, feedback, or education.   * ASD diagnosis * aged 1 – 6 years * RCTs assessing efficacy * published in English * primary outcome of core area of functioning affected by autism   Excluded studies:   * Where children were suspected of autism but not diagnosed. | **Method**: independent double study selection of retrieved articles, data extraction, critical appraisal using the Cochrane Risk-of-bias tool, and rating ‘Quality of evidence’ using GRADE (very low, low, moderate, and high) are reported here only for meta-analyses involving PMI studies.  Meta-analyses undertaken. Reported weighted average effect size due to variability in sample sizes between studies for each outcome.  Effect sizes are presented as weighted mean overall effect sizes (Hedges’ *g*) | **Included**: 19 articles (n =608 autistic children, M=42 months old; aged 15 months – 6 years). PMIs were either social-communication based or naturalistic developmental behavioural.  Study designs: All RCTs  Study quality: *moderate quality* of evidence for autism ‘symptom severity’, communication-language, and cognition; *very low* for socialization. Parents’ assessment not blind to condition. Study findings tended to be imprecise and most were underpowered. Publication bias was likely as search focused on peer-reviewed studies.  Duration: 1 week – 12 months  **Key findings** (no. studies) (weighted *g* ES) *for PMI studies only*  Autism ‘symptom severity’ (n=6) Small effect (ES = 0.22) (Moderate quality)  Socialisation (n=13) Small effect (ES = 0.23) (Very low quality)  Communication/Language (n=13) ‘trivial’ effect’ (ES = 0.18) (Moderate quality  Cognition (n=6) Small effect (ES = 0.24) (Moderate quality)  In sub-group analyses, no difference found for ‘dose of treatment’, or for comparator type (when comparator was treatment-as-usual compared to an active comparison group).  There were conflicting results based on informant type (for assessment). Based on parent report only, treatment effects were significant for communication-language (ES=0.18) and non-significant for socialisation. The opposite was found for clinician-rated tools; ie; non-significant for communication-language and significant (ES=0.27) for socialisation. Significant heterogeneity for both outcomes.  The small sample sizes precluded sub-group analyses by cognition and ‘ASD severity’. | Author conclusions:  Most outcome domains of parent-delivered intervention are associated with small effects, the quality of research (based on GRADE assessments) is improving since Oono et al’s (2013) [[17](#_ENREF_17)].  Argue that whether (usually blinded) clinician ratings or (non-blinded) parent ratings were higher differed for different outcomes suggests that the inability to blind parents may have minimal effects on outcomes (also concluded by Oono et al., (2013) [[17](#_ENREF_17)].  **Reviewer’s comments**: The search strategy was broad though omitted EMBASE and CINAHL. High quality SR and MA procedures undertaken. Acknowledge that prone to publication bias.  Unique studies: 1 of 19 (1 unique RCT) |

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Naveed et al., (2019)**  [[25](#_ENREF_25)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| Country: USA  **Study type**: systematic review & meta-analysis  Evidence level: I  **Study Quality**: JBI checklist score: 9/11 (high quality)  **Aims**: effectiveness of interventions mediated by non-specialist (parent, caregiver, peer, teacher) for autistic children and young people. For studies pooled across different delivery agents, effect sizes are only reported for PMI studies. | **Databases**: PubMed, CINAHL, Scopus, Web of Science, POPLINE, New York Academy of Medicine, PyscINFO, PsycARTICLE.  **Search**: Searched to Dec 31 2018. Transparent selection criteria. Full search terms for keywords described.  Citation searching not mentioned. Observed PRISMA reporting.  Selection criteria:   * Interventions mediated by a parent, caregiver, peer, teacher, or other non-specialist with supervision (onsite/online) by specialist. * ASD diagnosis (with or without comorbidities) * RCTs assessing efficacy * No language restrictions applied.   Excluded studies:   * Excluded interventions among adults, and reports on overlapping data sets. | **Method**: independent triple study screening and selection of retrieved articles, data extraction, and double critical appraisal using the Cochrane Risk-of-bias tool.  ‘Certainty in evidence’ ratings using GRADE (very low, low, moderate, and high) are reported here only for meta-analyses involving PMI studies.  Meta-analyses undertaken. Pooled effect sizes were adjusted for publication bias using Duvall & Tweedie’s Trim and Fill method. | **Included**: 33 articles (number of autistic children not reported) with intervention delivered by: parents (n=23), peers (n=6); school staff (n=4).  Study designs: All RCTs (incl. 1 cluster-randomised RCT)  Study quality: Low risk of bias for majority but high for n=11 studies. Common sources related to lack of reporting of randomisation method, lack of concealment of allocation, lack of blinding of outcome assessors, and attrition bias.  Duration: ranged from 7 to 48 weeks  **Key findings** (no. studies) (effect size) *for PMI studies only*  Adaptive behaviours (n=5) No improvement.  Autism ‘symptom severity’ (n=9) Moderate effect (SMD = 0.44)  Repetitive behaviours (n=3) Small effect (SMD = 0.36) (High certainty)  Self-regulation (n=3) Moderate effect (SMD = 0.54) (Low certainty)  Distress (child) (n=3) Moderate effect (SMD = 0.57)   * Social skills (n=10) Moderate effect (SMD = 0.42)   Communication   * Communication (n=10) No improvement. * Expressive language (n=5) Moderate effect (SMD = 0.45) * Receptive language (n=4) No improvement (Moderate certainty) * Joint attention (n=7) No improvement. * Joint engagement (n=5) Large effect (SMD = 1.01)   Motor skills (n=5) No improvement.  Visual reception (n=3) No improvement  Parent outcomes*:*   * Parental distress (n=10) Small effect (SMD = 0.33) (High certainty) * Parental self-efficacy (n=8) Small effect (SMD = 0.38) * Parent-child relationship (n=6) Moderate effect (SMD = 0.67)   Sub-group analyses of the full sample of 33 studies considered whether delivery agent (parent, teacher, peer) affected results. Though not possible for 5 outcomes where all studies were PMI, findings varied significantly for two outcomes:   * *Communication* (p=0.04) Significant improvement only evident for peers (n=1, ES=0.86) and teachers (n=2, ES=0.46). Moderate/high heterogeneity (I2=73%). * *Joint engagement* (p=0.02) Significant improvement only evident for parents (n=5, ES=1.01) but not teachers or peers. Moderate heterogeneity (I2=66%). | Author conclusions:  Findings suggest that non-specialist mediated interventions demonstrate effectiveness across a range of outcomes for autistic children and their caregivers. Most of the studies were mediated by parents and caregivers and presented low risk of bias. However, the evidence for peer and teacher mediated interventions was poor due to a limited number of studies.  **Reviewer’s comments**: The search strategy was broad though omitted EMBASE and citation searching. High quality SR and MA procedures undertaken.  There were many discrepancies in data reported between Figures, Tables and text. The author was contacted and was very helpful in providing clarification.  Unique studies: 10 of 23 (5 unique RCTs) |

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| **Tarver et al., (2019)**  [[42](#_ENREF_42)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| Country: USA  **Study type**: systematic review & meta-analysis  Evidence level: I  **Study Quality**: JBI checklist score: 9/11 (high quality)  **Aims**: effectiveness of behavioural parent interventions (BPI) on ‘disruptive and hyperactive behaviour’ in autistic children (to address co-occurring conditions including ADHD and anxiety and core impairment) | **Databases**: PubMed, EMBASE, PyscINFO.  **Search**: Searched to Dec 2017. Transparent selection criteria. Full search terms for keywords described.  Citation searching and hand searching of key Journals. Showed flowchart of searching.  Selection criteria:  Unimodal behavioural parent interventions (BPI) mediated by a parent.   * Directly targeted behavioural, or emotional, problems in ASD * Included valid outcome measures of disruptive/non-compliant child behaviour, ADHD symptoms, or emotional problems * Targeted children with clinical diagnosis of ASD, aged 2 – 18 years * RCTs * Published in peer-reviewed Journals * No language or date restrictions applied.   Excluded studies:   * Excluded studies were those that evaluated interventions targeting parental well-being as primary outcome, included multimodal psychosocial interventions with a BPI component, case studies, or samples < 10. | **Method**: independent double study screening and selection of retrieved articles, data extraction, and double critical appraisal using the Cochrane Risk-of-bias tool.  Meta-analyses undertaken. Publication bias not assessed as such tests require substantially more studies | **Included**:11 articles (9 studies) (n=520 autistic children)  Study designs: All RCTs  Study quality: Common sources related to lack of reporting of randomisation method, and attrition. lack of blinding of outcome assessors (parents, and often not reported for research personnel).  Duration: ??  **Key findings** (no. studies) (effect size)  Child disruptive behaviour (n=9) Moderate effect (SMD = 0.67)  Hyperactivity *(parent reported):* (n=3) small effect (SMD = 0.31)  Parent stress (n=7) small effect (SMD = 0.37)  Parent efficacy (n=5) no improvement but substantial heterogeneity (I2=81%), when removed one trial, was significant (SMD=0.60) | Author conclusions:  Review adds to the growing evidence of the efficacy of behavioural parent interventions for child behaviour and parental well-being.  **Reviewer’s comments**: The search strategy was broad though omitted CINAHL. High quality SR and MA procedures undertaken.  Unique studies: 3 of 11 (3 unique RCTs)  Supersedes SR of Postorino et al., (2017) [[54](#_ENREF_54)] |

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| **Trembath et al., (2019)** [[27](#_ENREF_27)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| **Country**: Australia  **Study type**: systematic review  Evidence level: I  **Study Quality**: JBI checklist score: 9/11 (high quality)  **Aims**: to examine factors (including baseline child and parent characteristics and behaviour, and contextual factors) that may mediate and moderate the effectiveness of parent-mediated approaches for ‘children with ASD’.  (Note: the review considered 41 articles to describe study selection and socio-economic characteristics to inform future research. However only data relating to a subset of 15 articles reporting on associations between outcomes and moderating factors are reported here). | **Databases**: Cochrane Library, Medline, EMBASE, PyscINFO, Scopus, Google Scholar  **Search**: Searched 2008 – Sept 2018. Transparent selection criteria. Full search terms for keywords described.  Citation searching. Observed PRISMA reporting.  Selection criteria:   * Interventions delivered by a parent/caregiver after being trained by professionals * Included pre-post *primary* outcome measures of adaptive behaviour (social, communication, daily living skills), ‘ASD symptoms’, or cognition * Targeted children with ASD, aged 0 – 12 years * Employed a group design with a control group * Reported statistics relating to changes in outcome variables * Published in English in peer-reviewed Journals   Excluded studies:   * Excluded studies which: evaluated interventions not primarily PMI; did not report child outcome data; were case studies, SCED studies or non-controlled studies; considered interventions involving diet, exercise, or medications; considered interventions aimed primarily at reducing/preventing social, emotional and behavioural problems | **Method**: independent double study screening and selection of retrieved articles. Single reviewer data extraction, and critical appraisal using the Effective Public Health Practice Project (EPHPP) Risk-of-bias tool. | **Included**: 15 articles (n=1,007 autistic children, age 12 months – 6 years)  Study designs: 13 RCTs and 2 non-randomised controlled trials or cohort studies  Study quality: Using the EPHPP tool, 12 studies were rated as strong, 2 as moderate, and 1 as being of weak quality.  Duration: 8 weeks – 2 years  Key findings  15 studies included an analysis of the relationship between a baseline/descriptive factor and the effectiveness of an outcome for PMI recipients, whether a significant, non-significant, or mixed relationship was found was recorded for 45 factors investigated across the 15 studies (13 of which were RCTs). These factors were grouped into categories and analyses reported below focus on RCT evidence.  Child characteristics (eg; age, ASD ‘severity’, language, non-verbal development): mixed or non-significant findings.  Child behaviours (eg; social and language domain skills such as vocal initiation, joint attention, responsibility to adult communication, smiling, object exploration): nonsignificant or mixed findings  Child contextual: single studies found non-significant findings relating to source of recruitment, caregiver adherence, involvement or competence, and non-significant findings in one study relating to intervention hours, with only vocabulary production outcome significantly increased.  Parent demographic: no association with parent education or occupation in 2 studies, mixed results in another study.  Parent intervention non-significant or mixed findings for programme fidelity. One study found positive parent behaviours (such as sensory-motor support, joining, use of affect, reciprocity) were related to social communication outcomes.  Parent contextual (eg; accessibility of program, training, location): qualitative analysis in one study suggested therapist support, time pressures, access to training in variety of formats, may impact on uptake and impact of intervention. One study found parents in rural areas were less likely to say their children made a lot of progress. | Author conclusions:  The findings indicate that a range of personal (e.g., participant characteristics, co-occurring conditions), intervention (e.g., approach and intensity), and contextual (e.g., referral source) characteristics may be relevant. Yet, despite assessment of a broad range of factors in studies, to date relatively few have included analyses of these.  Remains a need for improved study quality and measurement consistency in research, including a detailed examination of factors that may predict, moderate, and mediate intervention effectiveness for children and their parents.  **Reviewer’s comments**: The search strategy was broad though omitted CINAHL and only considered research published from 2008. Excluded interventions aimed primarily at reducing/preventing social, emotional and behavioural problems.  Of 45 factors explored, there were a handful of isolated positive associations. Overall, there were no clear or consistent patterns identifying moderating factors.  Unique studies:  7 of 15 (5 unique RCTs) |

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| **Shalev et al., (2020)** [[28](#_ENREF_28)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| Country: USA  **Study type**: systematic review  Evidence level: I  **Study Quality**: JBI checklist score: 6/11 (medium quality)  **Aims**: to examine baseline parent characteristics that may mediate and moderate the effectiveness of parent-mediated approaches for ‘children with ASD’. | **Databases**: PubMed, Web of Science  **Search**: Searched 1987 to Sept 2018. Transparent selection criteria. Search terms for keywords described.  Citation searching conducted.  Observed PRISMA reporting.  Selection criteria:   * Parent-mediated interventions (PMI) where parents were actively trained to deliver the intervention * Included at least one participant diagnosed with ASD * Included outcome measures relating to children’s behaviour * Controlled group design or SCED studies * Published in English in peer-reviewed Journals * Investigated the relationship between parent characteristics and children’s treatment outcomes. * Noted that as the review was exploratory, studies were not selected based on intervention efficacy; however, intervention effectiveness data was briefly presented for each included study.   Excluded studies:   * Excluded where parent baseline characteristics were not available or relationship with child outcomes not quantified. | **Method**: independent double study screening and selection of retrieved articles, data extraction.  No critical appraisal tool employed. | **Included**:11 articles (n=724 autistic children), 90% (n=10) studies included children aged under 6 years. All involved group-based training for interventions.  Study designs: 8 group-based RCTs, 3 quasi-experimental studies  Study quality: not formally appraised  Duration: not reported  **Key findings** Relevant analyses reported below focus on RCT evidence  (no. studies) (effect size)  Baseline parent stress (n=4): mixed moderating effects   * Better response for PMI for parents with high family stress for IQ but not for problem behaviours * Better response for parent home-based massage programme for parents with lower baseline stress * Better PMI treatment response for parents with higher baseline ‘parenting reward’ and lower ‘parenting burden’ * No impact on joint engagement based on baseline parental stress   Demographic variables (n=3): possible effect   * 1 of 2 studies found low socio-economic status, but not maternal education, related to increased IQ but not problem behaviours * 1 study found maternal age not related to treatment effects for expressive and receptive language * Insight (parent’s awareness of child’s cognitions and affect observed in video clips) (n=1): no moderating effect * 1 study found no relationship between insight and outcomes (expressive language). | Author conclusions:  Although this emerging literature is characterized by high variability in regard to the specific treatment employed, parent measures utilized, and child outcome targeted, initial results suggest that parent factors may act as moderators in the context of some interventions. These results underscore the need for systematic research on the role of parent baseline characteristics in PMI.  **Reviewer’s comments**: The search strategy was narrow, restricted to 2 databases. No formal critical appraisal performed and synthesis was mainly descriptive.  Unique studies: 3 of 11 |

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| **Conrad et al., (2021)**  [[43](#_ENREF_43)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| **Country**: Denmark  **Study type**: systematic review & meta-analysis  Evidence level: I  **Study Quality**: JBI checklist score: 10/11 (high quality)  **Aims**: effectiveness of parent-mediated interventions (PMI) for children ‘with ASD’ aged 18 months–17 years on beneficial and adverse outcomes. | **Databases**: Medline, CINAHL, PsycINFO, Embase, ERIC  **Search**: Searched using SR and MA filters, then searched for primary studies between 2017 to March 2020 also appraised in this umbrella review). Transparent selection criteria. Full search terms for keywords described.  Citation searching conducted. Observed PRISMA reporting.  Selection criteria:   * PMI included 8 or more sessions * Aged 8 months – 17 years * ASD diagnosis (with or without comorbidities) * Adaptive functioning (as primary outcome), and other prespecified secondary outcomes (see Results) * RCTs only * published in English and Scandinavian | **Method**: independent double study selection of retrieved articles, and double critical appraisal using the Cochrane Risk-of-bias tool.  Rated ‘certainty in evidence’ using GRADE as very low, low, moderate, and high based on overall risk of bias, inconsistency, indirectness, imprecision, and publication bias. | **Included**: 30 articles reporting on 30 trials (n=1,934 autistic children, all aged < 14 years). Meta-analysis undertaken on 26 studies were outcome data permitted.  Study designs: All RCTs. Comparators included waitlist/passive control (n=11), management as usual (n=10), active control (n=8), pharmacotherapy (n=1).  Study quality: Risk of bias table presented. The certainty in the evidence was downgraded due to serious risk of bias, lack of blinding, and serious risk of imprecision due to few participants included in meta-analyses.  Duration: 8 weeks to 24 months  **Key findings** (no. studies)  Primary outcome:   * Adaptive functioning (VABS) * Parent rated (n=8) Small (clinically relevant) effect on parent-rated adaptive functioning (SMD=0.28) (low heterogeneity, low certainty of evidence). * Clinician-rated (n=2) no effect (low certainty)   Secondary outcomes:   * Core symptoms of ASD   Parent rated (n=7) no clinically relevant effect   * Clinician-rated (n=9) Small clinically relevant effect (SMD=-0.35) (high heterogeneity, low certainty) * Disruptive behaviour (Parent-rated) (n=9) Moderate (clinically relevant) effect (SMD=-0.55) (low heterogeneity, moderate certainty) * Parental well-being (n=12) small but not clinically relevant effect (low certainty of evidence) * Adverse events (n=2) No effect (low certainty of evidence)   Improvements in outcomes for the PMI groups compared to comparators did not differ as a function of outcome target areas (language, aggression management, training in social skills) and other intervention types in subgroup analyses. No studies reported on child’s quality of life, or child anxiety outcomes. | Author conclusions:  PMI’s may slightly improve parent-rated adaptive functioning but there was no effect when rated by a clinician. PMIs probably improves parent-rated disruptive behaviour considerably. PMI’ s may slightly improve clinician-rated autism core characteristics, and may slightly improve parental well-being. PMI’s had no effect on core characteristics of autism (rated by parent) but may slightly improve clinician-rated core characteristics. PMI’s had no effect on adverse events  Findings suggest that clinicians may consider introducing PMIs to children with ASD, but more high-quality RCTs are needed because the effects are not well-established, and the results are likely to change with future studies.  **Reviewer’s comments**: search strategy somewhat limited by original limit to secondary literature, and then follow-up search from 2017, with several papers only identified through citation searching. Very high-quality review procedures and reporting undertaken.  Limited outcomes considered in scope. Notably did not consider joint attention, and social interaction outcomes as prespecified outcomes.  The review informed the national Clinical Practice Guideline for autism published by the Danish Health Authority in 2021.  Unique studies: 9 of 33 (9 unique RCTs) |

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| **Pacia et al., (2021)** [[29](#_ENREF_29)] | | | | |
| Country, study type, aim | Search strategy | Appraisal methods | Results | Conclusions |
| **Country**: Ireland  **Study type**: systematic review  Evidence level: I  **Study Quality**: JBI checklist score: 6/11 (medium quality)  **Aims**: effectiveness of applied behavioural interventions delivered by parents and siblings to young autistic children aged 0 – 6 years old to support social communication outcomes. | **Databases**: Psychology and Behavioural Sciences Collection, PyscINFO, ERIC, Web of Science  **Search**: Searched 1980 – 2019. Transparent selection criteria. Full search terms for keywords described.  Citation searching conducted. Observed PRISMA reporting.  Selection criteria:   * Parent- or sibling-mediated social communication interventions. * All diagnosed with ASD aged 0 – 6 years in group designs, or at least one in single case experimental designs (SCED) * Parent or sibling implemented 100% of intervention * Intervention included one or more ‘focussed intervention practice’; i.e., based on applied behavioural principles * Included measure of at least one social communication skill * Experimental design (group-controlled trial or SCED) * Published in peer reviewed Journals, in English. | **Method**: independent double study screening and selection of retrieved articles, and data extraction.  Critical appraisal used indicators that permit inclusion of SCED studies [[55](#_ENREF_55)] (see Reviewer’s comments).  Treatment effectiveness was presented separately for SCED and group studies.  For SCED studies, PND (percentage of nonoverlapping data) were determined from graphically presented data. PND scores were graded as follows: PND≥90%=very effective; 70-89%=effective, 51-69%=questionable, <50%=ineffective. Could be determined for n=39 SCED studies.  Cohen’s *d* was used to calculate effect Sizes (ES) for groups studies, and improvement/effect graded as follows: < 0.2 (trivial), 0.02 – 0.49 (small), 0.05 – 0.79 (medium), or ≥ 0.8 (large) [[53](#_ENREF_53)]. Could be determined for n=8 experimental controlled group studies. | **Included**: 54 articles (n=444 autistic children, Age range=19-69 months old, 71% male, 19% female, 10% not reported). Parents mediated intervention in 50 studies, siblings (aged 4-13 years, M=8 years) in 4 studies.  Study designs: 42 SCED studies, and 12 were group studies. Of the group studies, 9 were RCT’s (2 unique to this review), 2 were RCTs where the comparator was also a PMI (different components), and one was a before and after study.  Study quality: Determined but not reported here (see Reviewer’s comments)  Duration: 5 days – 1 year  Key findings  Social engagement   * SCED studies (n=25): 60% effective/very effective * Group studies (n=4): 75% medium/large effect * Language and communication*:* * SCED studies (n=35): 37% effective/very effective * Group studies (n=6): 50% medium/large effect * Imitation and play * SCED studies (n=16): 44% effective/very effective * Group studies (n=1): 100% medium effect * No difference in proportion of very effective/effective treatment between PMI (46%) and sibling-mediated interventions (50%), but only 4 of the latter. No difference for setting (clinic, home, telehealth), or dosage (hours of intervention). * Among studies (75%) that evaluated the generalisation, over 90% demonstrated partial or complete generalisation outcomes. | Author conclusions:  ‘Treatment’ was evaluated as slightly more effective for social engagement skills when compared to language and communication skills, and imitation and play skills, although not by a wide margin.  **Reviewer’s comments**: The search strategy was broad though omitted health and nursing databases.  The quality checklists used were designed by Reichow, Volkmar and Cicchetti (2008) expressly because “we feel it is imperative that single subject research be included in a definition of Evidence Based Practice (EBP) used to review treatments for young children with autism” (Note 1) [[55](#_ENREF_55)].  Checklists developed/used by major Health Technology Assessment and Guideline development organisations were not employed. Of the 2 RCTs receiving a ‘weak’ rating, “one was given this rating only because it did not specify the gender of the participants” (p. 15). The authors and other researchers have expressed concerns about this tool and alternative or additional approaches are recommended for future research. This reviewer lacks confidence in the assessments of study quality and therefore they are not reported.  Unique studies: 46 of 54, (2 unique RCTs) |
| **Key**: ASD=Autism Spectrum Disorder; CENTRAL=Cochrane Central Register of Controlled Trials; cf=compared with; CINAHL=Cumulative Index to Nursing and Allied Health Literature; ERIC=Education Resources Information Centre; JBI=Joanna Briggs Institute; MEDLINE=Medical Literature Analysis and Retrieval System Online; ns=not significant; PMI=parent-mediated intervention; PRISMA=Preferred Reporting Items for Systematic Reviews and Meta-Analyses; RCT=randomised controlled trial; PsycINFO=Psychological Information Database; SCED=single case experimental design; US=United States | | | | |

References

[1] Whaikaha – Ministry of Disabled People and Ministry of Education, *Aotearoa New Zealand Autism Guideline: He Waka Huia Takiwātanga Rau*, 3rd ed. Wellington: Whaikaha – Ministry of Disabled People, 2022.

[2] Broadstock, M., “New Zealand autism spectrum disorder guideline supplementary paper on applied behaviour analysis,” New Zealand Guidelines Group, Wellington, New Zealand, 2010. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[3] Broadstock, M., “New Zealand autism spectrum disorder guideline supplementary paper on three pharmacological interventions,” New Zealand Guidelines Group, Wellington, New Zealand, 2011. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[4] Broadstock, M., *New Zealand autism spectrum disorder guideline supplementary paper on supported employment services*. Wellington, New Zealand: New Zealand Guidelines Group, 2012. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[5] Broadstock, M., *New Zealand autism spectrum disorder guideline supplementary paper on gastrointestinal problems in young people*. Christchurch, New Zealand: INSIGHT Research, 2013. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[6] Broadstock, M., *New Zealand autism spectrum disorder guideline supplementary paper on implications of DSM-5 for the diagnosis of ASD*. Christchurch, New Zealand: INSIGHT Research, 2014. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[7] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline supplementary paper on social skills groups for children and young people with ASD*. Christchurch, New Zealand: INSIGHT Research, 2015. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[8] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline supplementary paper on cognitive behaviour therapy for adults with ASD*. Christchurch, New Zealand: INSIGHT Research, 2016. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[9] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline supplementary paper on the impact of ethnicity on recognition, diagnosis, education, treatment and support for people on the autism spectrum*. Christchurch, New Zealand: INSIGHT Research, 2018a. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[10] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of sexuality education for young people on the autism spectrum*. Christchurch, New Zealand: INSIGHT Research, 2018b. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[11] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of strategies for supporting school transitions for young people on the autism spectrum*. Christchurch, New Zealand: INSIGHT Research, 2019. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[12] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of physical activity interventions for young people on the autism spectrum*. Christchurch, New Zealand: INSIGHT Research, 2021. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[13] Broadstock, M., *New Zealand Autism Spectrum Disorder Guideline’s supplementary paper on the effectiveness of music therapy interventions for children and young people on the autism spectrum*. Christchurch, New Zealand: INSIGHT Research, 2021. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[14] Broadstock, M., *Aotearoa New Zealand Autism Guideline’s supplementary paper on the effectiveness of supports for autistic students in tertiary education*. Christchurch, New Zealand: INSIGHT Research, 2022. Available: <https://www.whaikaha.govt.nz/about-us/policy-strategies-and-action-plans/nz-autism-guideline/>

[15] Ministries of Health and Education, *New Zealand autism spectrum disorder guideline*, First ed. Wellington: Ministry of Health, 2008.

[16] Ministries of Health and Education, *New Zealand autism spectrum disorder guideline*, 2nd ed. Wellington: Ministry of Health, 2016.

[17] Oono, I. P., Honey, E. J., and McConachie, H., (2013). “Parent‐mediated early intervention for young children with autism spectrum disorders (ASD),” *Cochrane Database of Systematic Reviews,* (4). doi: 10.1002/14651858.CD009774.pub2

[18] Autism Self-Advocacy Network, “The A Word: Why use the word Autistic?,” Available: <https://www.asan-au.org/autistic-the-word/>

[19] Silberman, S., *Neurotribes: The Legacy of Autism and the Future of Neurodiversity*. New York, NY: Penguin Random House LLC, 2015.

[20] American Psychiatric Association, *Diagnostic and statistical manual of mental disorders. 4th Edition (Text revision)*. Washington, DC: APA Press, 2000.

[21] O’Neill, S., (2008). “The meaning of autism: beyond disorder,” *Disability & Society,* 7:787-799.

[22] Opai, K., "From autism to takiwātanga: An origin story," presented at the Australasian Society for Autism Research Conference, Wellington, New Zealand, 11 December 2020, 2020.

[23] Durie, M. H., (1984). “"Te taha hinengaro": An integrated approach to mental health,” *Community Mental Health in New Zealand,* 1(1):4-11.

[24] Ministry of Health, *Whāia Te Ao Mārama 2018 to 2022: The Māori Disability Action Plan*. Wellington, New Zealand: Ministry of Health, 2018. Available: <https://www.health.govt.nz/publication/whaia-te-ao-marama-2018-2022-maori-disability-action-plan>

[25] Naveed, S., Waqas, A., Amray, A. N. *et al.*, (2019). “Implementation and effectiveness of non-specialist mediated interventions for children with Autism Spectrum Disorder: A systematic review and meta-analysis,” *PloS one,* 14(11):e0224362. doi: 10.1371/journal.pone.0224362

[26] Nevill, R. E., Lecavalier, L., and Stratis, E. A., (2018). “Meta-Analysis of Parent-Mediated Interventions for Young Children with Autism Spectrum Disorder,” *Autism: The International Journal of Research and Practice,* 22(2):84-98.

[27] Trembath, D., Gurm, M., Scheerer, N. E. *et al.*, (2019). “Systematic review of factors that may influence the outcomes and generalizability of parent-mediated interventions for young children with autism spectrum disorder,” *Autism research : official journal of the International Society for Autism Research,* 12(9):1304-1321. doi: 10.1002/aur.2168

[28] Shalev, R. A., Lavine, C., and Di Martino, A., (2020). “A systematic review of the role of parent characteristics in parent-mediated interventions for children with autism spectrum disorder,” *Journal of Developmental and Physical Disabilities,* 32(1):1-21. doi: 10.1007/s10882-018-9641-x

[29] Pacia, C., Holloway, J., Gunning, C., and Lee, H., (2021). “A Systematic Review of Family-Mediated Social Communication Interventions for Young Children with Autism,” *Rev J Autism Dev Disord*:1-27. doi: 10.1007/s40489-021-00249-8

[30] Koegel, R. L., Symon, J. B., and Kern Koegel, L., (2002). “Parent Education for Families of Children with Autism Living in Geographically Distant Areas,” *Journal of Positive Behavior Interventions,* 4(2):88-103. doi: 10.1177/109830070200400204

[31] Bearss, K., Burrell, S., Stewart, L., and Scahill, L., (2015). “Parent Training in Autism Spectrum Disorder: What's in a Name?,” *Clin Child Fam Psychol Rev,* 18(2):179-182. doi: 10.1007/s10567-015-0179-5

[32] Althoff, C. E., Dammann, C. P., Hope, S. J., and Ausderau, K. K., *Parent-Mediated Interventions for Children With Autism Spectrum Disorder: A Systematic Review, The American Journal of Occupational Therapy*, vol. 73, pp. 7303205010p7303205011-7303205010p7303205013, 2019.

[33] Rutherford, M., Singh-Roy, A., Rush, R., McCartney, D., O'Hare, A., and Forsyth, K., (2019). “Parent focused interventions for older children or adults with ASD and parent wellbeing outcomes: A systematic review with meta-analysis,” *Research in Autism Spectrum Disorders,* 68. doi: 10.1016/j.rasd.2019.101450

[34] Diggle, T., McConachie, H. R., and Randle, V. R. L., (2004). “Parent-mediated early intervention for young children with autism spectrum disorder,” *Cochrane Database of Systematic Reviews,* 2.

[35] Birkin, C., Anderson, A., Seymour, F., and Moore, D. W., (2008). “A parent-focused early intervention program for autism: Who gets access?,” *Journal of Intellectual & Developmental Disability,* 33(2):108-116. doi: 10.1080/13668250802036746

[36] Pretorius, E., Clendon, S., and McLaughlin, T., (2020). “Parent perspectives on receiving support for enhancing parent-child interactions,” *Journal of Clinical Practice in Speech-Language Pathology,* 22(2):74-78.

[37] Waddington, H., van der Meer, L., Sigafoos, J., and Bowden, C. J., (2020). “Mothers’ perceptions of a home-based training program based on the Early Start Denver Model,” *Advances in Neurodevelopmental Disorders,* 4(2):122-133. doi: Available: <https://doi.org/10.1007/s41252-019-00146-6>

[38] Aromataris, E., Fernandez, R., Godfrey, C. M., Holly, C., Khalil, H., and Tungpunkom, P., (2015). “Summarizing systematic reviews: methodological development, conduct and reporting of an umbrella review approach,” *JBI Evidence Implementation,* 13(3).

[39] Haddaway, N. R., Page, M. J., Pritchard, C. C., and McGuinness, L. A., (2022). “PRISMA2020: An R package and Shiny app for producing PRISMA 2020-compliant flow diagrams, with interactivity for optimised digital transparency and Open Synthesis,” *Campbell Systematic Reviews,* 18(2):e1230. doi: <https://doi.org/10.1002/cl2.1230>

[40] National Health and Medical Research Council, “NHMRC additional levels of evidence and grades for recommendations for developers of guidelines: pilot program 2005-2007,” NHMRC. Available: <https://www.nhmrc.gov.au/sites/default/files/images/NHMRC%20Levels%20and%20Grades%20(2009).pdf>

[41] Merlin, T., Weston, A., and Tooher, R., (2009). “Extending an evidence hierarchy to include topics other than treatment: revising the Australian 'levels of evidence',” *BMC Medical Research Methodology,* 9(34).

[42] Tarver, J., Palmer, M., Webb, S. *et al.*, (2019). “Child and Parent Outcomes Following Parent Interventions for Child Emotional and Behavioral Problems in Autism Spectrum Disorders: A Systematic Review and Meta-Analysis,” *Autism: The International Journal of Research and Practice,* 23(7):1630-1644.

[43] Conrad, C. E., Rimestad, M. L., Rohde, J. F. *et al.*, (2021). “Parent-Mediated Interventions for Children and Adolescents With Autism Spectrum Disorders: A Systematic Review and Meta-Analysis,” *Frontiers in Psychiatry,* 12. doi: 10.3389/fpsyt.2021.773604

[44] Guyatt, G. H., Oxman, A. D., Vist, G. E. *et al.*, (2008). “GRADE: an emerging consensus on rating quality of evidence and strength of recommendations,” *BMJ,* 336(7650):924. doi: 10.1136/bmj.39489.470347.AD

[45] American Psychiatric Association, *Diagnostic and statistical manual of mental disorders. 5th Edition*. Washington, DC: APA Press, 2013.

[46] Brignell, A., Chenausky, K. V., Song, H., Zhu, J., Suo, C., and Morgan, A. T., (2018). “Communication interventions for autism spectrum disorder in minimally verbal children,” *Cochrane Database of Systematic Reviews,* (11). doi: 10.1002/14651858.CD012324.pub2

[47] Ne’eman, A., (2021). “When disability is defined by behavior, outcome measures should not promote ‘passing’,” *AMA Journal of Ethics,* 23(7):E569-575. doi: doi: 10.1001/amajethics.2021.569

[48] Anagnostou, E., Jones, N., ., Huerta, M. *et al.*, (2015). “Measuring social communication behaviors as a treatment endpoint in individuals with autism spectrum disorder,” *Autism,* 19(5):622–636.

[49] Ratliff-Black, M. & Therrien, W., (2021). “Parent-Mediated Interventions for School-Age Children With ASD: A Meta-Analysis,” *Focus on Autism and Other Developmental Disabilities,* 36(1):3-13. doi: <https://dx.doi.org/10.1177/1088357620956904>

[50] Dawson, G., Franz, L., and Brandsen, S., (2022). “At a Crossroads-Reconsidering the Goals of Autism Early Behavioral Intervention From a Neurodiversity Perspective,” *JAMA Pediatr*. doi: 10.1001/jamapediatrics.2022.2299

[51] Salvador-Oliván, J. A., Marco-Cuenca, G., and Arquero-Avilés, R., (2021). “Development of an efficient search filter to retrieve systematic reviews from PubMed,” *J Med Libr Assoc,* 109(4):561-574. doi: 10.5195/jmla.2021.1223

[52] Rydzewska, E., Dunn, K., and Cooper, S.-A., (2020). “Umbrella systematic review of systematic reviews and meta-analyses on comorbid physical conditions in people with autism spectrum disorder,” *The British Journal of Psychiatry,* 218(1):10-19. doi: 10.1192/bjp.2020.167

[53] Cohen, J., *Statistical power analysis for the behavioural sciences*, 2nd ed. Hillsdale, NJ, : Erlbaum, 1988.

[54] Postorino, V., Sharp, W. G., McCracken, C. E. *et al.*, (2017). “A Systematic Review and Meta-analysis of Parent Training for Disruptive Behavior in Children with Autism Spectrum Disorder,” *Clinical child and family psychology review,* 20(4):391-402. doi: 10.1007/s10567-017-0237-2

[55] Reichow, B., Volkmar, F. R., and Cicchetti, D. V., (2008). “Development of the Evaluative Method for Evaluating and Determining Evidence-Based Practices in Autism,” *Journal of Autism and Developmental Disorders,* 38(7):1311-1319. doi: 10.1007/s10803-007-0517-7

[56] Cook, J., Hull, L., Crane, L., Mandy, W. (2021). Camouflaging in autism: a systematci review. *Clinical Psychology Review,* 89:102080. doi:1.1016/j.cpr.2021.102080

[57] Trembath, D., Varcin, K., Waddington, H., et al. (2022). National guideline for supporting the learning, participation, and wellbeing of autistic children and their families in Australia. Autism CRC. Brisbane.

1. <https://www.education.govt.nz/our-work/overall-strategies-and-policies/ka-hikitia-ka-hapaitia/> [↑](#footnote-ref-1)
2. See https://www.health.govt.nz/system/files/documents/publications/achieving-equity-in-health-outcomes-summary-of-a-discovery-process-30jul2019.pdf [↑](#footnote-ref-2)
3. An example of work which investigates the research priorities of members of the autistic and autism communities is provided by this Aotearoa New Zealand study led by the University of Canterbury: <http://dx.doi.org/10.26021/13952> [↑](#footnote-ref-3)
4. Readers of this Supplementary Paper are referred to the Paper’s Preamble section (i.e., not Part 3’s Preamble) for a summary of [Autistic perspectives](#Preamble_AutisticPreferences) and [Māori perspectives](#Preamble_MaoriPreferences). [↑](#footnote-ref-4)
5. Autism specific terms were developed in consultation with Prof Jill Bevan-Brown who acted as expert advisor for the 3rd edition of the Guideline, and were also informed by Keri Opai’s *Te Reo Hapai* resource (https://www.tereohapai.nz) [↑](#footnote-ref-5)