Universidade Federal de Minas Gerais
Faculdade de Medicina
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Uma revisão de revisões sobre o que tem sido feito para potencializar a aprendizagem de crianças e adolescentes com deficiência intelectual

Linha de pesquisa – Distúrbios do neurodesenvolvimento

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Dissertação apresentada ao Programa de Pós-Graduação em Ciências da Saúde - Saúde da Criança e do Adolescente da Faculdade de Medicina da Universidade Federal de Minas Gerais, como requisito parcial para obtenção do grau de Mestre em Medicina, sob orientação da Profª. Drª. Débora Marques de Miranda.

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PROGRAMA DE PÓS-GRADUAÇÃO EM CIÊNCIAS DA SAÚDE
SAÚDE DA CRIANÇA E DO ADOLESCENTE

FOLHA DE APROVAÇÃO

UMA REVISÃO DE REVISÕES SOBRE O QUE TEM SIDO FEITO PARA POTENCIALIZAR A APRENDIZAGEM DE CRIANÇAS E ADOLESCENTES COM DEFICIÊNCIA INTELECTUAL

KEVIN AUGUSTO FARIAS DE ALVARENGA

Dissertação defendida em 10 de dezembro de 2021 como requisito parcial para obtenção do grau de Mestre em CIÊNCIAS DA SAÚDE, pelo Programa de Pós-Graduação em Ciências da Saúde-Saúde da Criança e do Adolescente e aprovada pela Comissão Examinadora designada pelo Colegiado do Programa de Pós-Graduação supramencionado da Universidade Federal de Minas Gerais, constituída pelas seguintes Professoras Doutoras: Débora Marques de Miranda - Orientadora (UFMG), Ana Cristina Simões e Silva (UFMG) e Juliana Gurgel Giannetti (UFMG).

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UNIVERSIDADE FEDERAL DE MINAS GERAIS
FACULDADE DE MEDICINA

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Orientadora: Profª. Drª Débora Marques de Miranda

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Faculdade de Medicina da Universidade Federal de Minas Gerais

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NOTA EXPLICATIVA

A apresentação da presente dissertação foi organizada sob a forma de redação de artigo científico, de acordo com a resolução 03/2010 aprovada pelo Programa de Pós-Graduação em Ciências da Saúde – Área de Concentração em Saúde da Criança e do Adolescente, da Faculdade de Medicina da Universidade Federal de Minas Gerais, disponível em http://www.medicina.ufmg.br/cpg/programas/saude_criança/arquivos/Resolucao03-2010.pdf.

A revisão de revisões produzida avaliou sistematicamente a literatura publicada a partir do ano 2000 contendo estratégias para melhoria da aprendizagem de crianças e adolescentes com deficiência intelectual, organizando as intervenções de acordo com suas categorias (cuidado, educação, medicamentos e dieta, psicoterapia, educação física, fisioterapia e terapia ocupacional, e intervenções baseadas em tecnologia) e principais desfechos (pessoal-social, motricidade fina-adaptativa, linguagem e motricidade grossa) analisados, avaliando a qualidade da evidência, e gerando recomendações que podem guiar profissionais de saúde, educadores, responsáveis e outros interessados no cuidado destes indivíduos.

As referências bibliográficas estão dispostas ao final de cada artigo ou seção, conforme as normas de Vancouver (*Uniform Requirements for Manuscripts Submitted to Biomedical Journals: Writing and Editing for Biomedical Publication* - www.ICMJE.org).

RESUMO

Contexto: A deficiência intelectual (DI) afeta de 1 a 3% da população pediátrica, e ensinar a essas crianças habilidades básicas de vida, além de otimizar seu capital mental - resultando em melhor adaptabilidade -, é um desafio. Não há consenso atual quanto às estratégias mais eficazes, e a literatura existente é ampla, mas ainda incerta.

Objetivos: Esta revisão de revisões visa compilar as informações e estratégias disponíveis para intervir na aprendizagem de crianças e adolescentes com deficiência intelectual, auxiliando a tomada de decisões na prática clínica, e o cuidado familiar.

Métodos: O protocolo de pesquisa foi registrado no PROSPERO sob o número CRD42020195913. Pesquisas foram feitas nos bancos de dados Medline, Cochrane, Scopus, Web of Science, Lilacs, SciELO, ERIC e PsycINFO para reunir revisões sistemáticas e metanálises publicadas desde o ano 2000 que investigaram os impactos de vários tipos de intervenções na aprendizagem dessa população. A qualidade metodológica foi avaliada usando a ferramenta AMSTAR-2.

Principais resultados: 59 estudos foram selecionados após leitura completa dos artigos. Esses foram divididos pelo domínio do principal desfecho avaliado (pessoal-social, motor fino-adaptativo, motor grosseiro ou linguagem) e pelo tipo de intervenção proposta (relacionadas ao cuidado, educação, medicamentos e dietas, fisioterapia e estratégias semelhantes, ou tecnologia).

Conclusões dos autores: Recomendações foram feitas para cada domínio de resultado usando as evidências coletadas. Os dados mostram benefícios da estimulação de habilidades específicas pelo cuidador, com explicação cuidadosa das atividades, feedback corretivo, engajamento em comportamento lúdico; educação com instrução sistemática (por exemplo, sequências de comando de alta probabilidade), e instrução embutida, com suporte de estratégias de análise

comportamental; intervenções motoras específicas, encorajando práticas de esportes, atividades físicas e terapia assistida por animais; o uso de ferramentas de comunicação alternativa e aumentativa, e dispositivos tecnológicos para suporte da aprendizagem. A baixa qualidade metodológica geral dos estudos é um fator limitante para a maioria das recomendações, e a implementação bem-sucedida também dependerá do treinamento, participação e status socioeconômico dos cuidadores.

ABSTRACT

Background: Intellectual disability (ID) affects 1 to 3% of the pediatric population, and teaching these children basic living skills, as well as optimizing their mental capital — resulting in better adaptability —, is a challenge. There is no current consensus as to the most effective strategies, and the extant literature is broad but still uncertain.

Objectives: This overview aims to compile the most useful information and strategies for intervening in the learning of intellectually disabled children and adolescents, aiding clinical practice, decision making, and family care.

Methods: A research protocol has been registered on PROSPERO under the number CRD42020195913. We searched the databases Medline, Cochrane, Scopus, Web of Science, Lilacs, SciELO, ERIC, and PsycINFO to amass systematic reviews and meta-analyses published since the year 2000 which investigated the impacts of multiple types of interventions on the learning of the selected population. Methodological quality has been assessed using the AMSTAR-2 tool.

Main results: 59 studies were selected after the full-text examination. Those were divided by outcome domains and subdivided by the type of intervention. Outcome's domains were personal-social, fine motor-adaptive, gross motor, or language-related. Interventions were

related to caregiving, education, medication and diet, physical therapy, and similar strategies, or technology.

Authors' conclusions: Recommendations were made for each outcome domain using the evidence gathered in the studies. The data shows benefits of: caregiver stimulation of specific skills with careful explanation of activities, corrective feedback and engaging in play-behavior; education with systematic instruction (e.g., high probability command sequences), and embedded instruction, with the support of behavioral analysis strategies; specific motor interventions, encouraging sport practicing, physical activities and animal-assisted therapy; the use of augmentative and alternative communication, and technological devices for support learning. The overall low methodological quality of the studies is a limiting factor for most recommendations, and the successful implementation will also depend on caregivers' training, participation, and socio-economic status.

LISTA DE ABREVIATURAS E SIGLAS

AAC - Augmentative and alternative communication.

AAT - Animal-assisted therapy.

ABC - Aberrant behavior checklist.

ADHD - Attention deficit hyperactivity disorder.

ASD - Autism spectrum disorder.

AVD - Active video gaming.

CAI - Computer-assisted instruction.

CBI - Computer-based interventions.

CGI - Clinical global impression.

CP - Cerebral palsy.

DCD - Developmental coordination disorder.

DD - Developmental disability.

DI - Deficiência intelectual.

DS - Down syndrome.

ECA - Estatuto da Criança e do Adolescente.

FMS - Fundamental movement skills.

FXS - Fragile X syndrome.

GMS - Gross motor skills.

ID - Intellectual disability.

IDD - Intellectual disability disorder.

LAC - L-acetylcarnitine.

MeSH- Medical Subject Headings

MID - Mild intellectual disability.

MPH - Methylphenidate.

PDD - Pervasive developmental disorder.

PRISMA-P - Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols

PROSPERO - Prospective Register of Systematic Reviews

PWS - Prader-Willi Syndrome

QI - Quociente de inteligência.

RCT - randomized clinical trials.

RSE - relationship and sexuality education.

SG - Serious games.

SO - Special Olympics.

SRF - skill-related fitness.

SUS - Sistema Único de Saúde.

TAI - Tablet-assisted instructions.

TSIF - Test of sensory integration function.

VMI - Visual-motor integration.

VR - Virtual reality.

WHO - World Health Organization.

WWC - What works clearinghouse.

SUMÁRIO

1. INTRODUÇÃO

A deficiência intelectual (DI) é caracterizada por limitações nas funcionalidades intelectuais e adaptativas, afetando pelo menos um dos três domínios adaptativos (conceitual, social e prático), iniciadas ainda no período de desenvolvimento (antes dos 18 anos de idade) [1]. Afeta 1 a 3% da população pediátrica. A introdução precoce de estratégias de estímulo à aprendizagem é crucial para o desenvolvimento de habilidades sociais, motoras e de linguagem e habilitação da criança para uma vida com qualidade e autonomia [2].

Os processos de aprendizagem em diferentes contextos, desde o ensino formal até o aprendizado de habilidades básicas para a vida independente podem estar prejudicados. Conceituada pelo *Medical Subject Headings (MeSH)* [3], aprendizagem, amplia-se além da aquisição de conhecimento. Inclui modificações de comportamento resultantes de experiência ou prática. Desta maneira, considera-se em seu escopo o ganho de qualquer tipo de habilidade a partir de estímulos, exercícios e treinamento.

A potencialização da aprendizagem, tarefa que é muitas vezes atribuída aos profissionais da educação, é de responsabilidade multiprofissional. Há densa literatura da área médica, psicológica e educacional demonstrando resultados de diferentes estratégias utilizadas para trazer melhoria na aprendizagem. Como o tema é amplo, mesmo as sínteses disponíveis em forma de revisões sistemáticas são fragmentadas em diversos tipos de intervenções e desfechos. Não há, portanto, diretrizes bem estabelecidas para guiar o cuidado multiprofissional destas crianças.

A infância e adolescência são uma janela para estímulos, e uma coordenação de estratégias, terapias e outras intervenções baseadas em evidências é chave para a potencialização da aprendizagem destes indivíduos.

O Estatuto da Criança e do Adolescente (ECA) [4] garante, como direito fundamental, o atendimento educacional especializado. Também garante atenção integral à saúde, por intermédio do Sistema Único de Saúde (SUS), com acesso universal e igualitário às ações e serviços para promoção, proteção e recuperação da saúde.

Nesta mesma ideia de proteção ao desenvolvimento da criança com DI, a atual legislação americana, incluindo os atos *No Child Left Behind* de 2001 [5], e *Individuals with Disabilities Education Improvement* de 2004 [6], recomendam que profissionais utilizem práticas de educação baseadas em evidências, em uma tentativa de incentivar a utilização de estratégias comprovadamente eficazes e eficientes, e a produção de estudos bem delineados.

Práticas baseadas em evidência podem otimizar recursos e aumentar o alcance de intervenções. Na educação especial, pesquisa experimental em caso único pode ser considerada uma metodologia para estabelecer práticas baseadas em evidência, uma vez que ensaios clínicos randomizados com múltiplos indivíduos são raros [7]. Esse tipo de estudo, no entanto, é sujeito a diversas formas de viés: desvio do observador (possibilidade de que os observadores possam alterar suas definições observacionais do construto que está sendo medido ao longo do tempo, não tornando as pontuações comparáveis nas fases do experimento); viés de observação (possibilidade de que os observadores possam ser influenciados por uma variedade de fatores associados aos resultados experimentais desejados); e reatividade (possibilidade de que as pontuações observacionais sejam mais altas como resultado do pesquisador monitorando os observadores ou processo observacional) [8]. A produção de revisões sistemáticas e meta-análises pode suprir essas deficiências, confirmando ou refutando seus resultados, e avaliando a possibilidade de recomendação da aplicação de certa intervenção no âmbito populacional.

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2. REVISÃO DE LITERATURA

A compreensão da etiologia e fisiopatologia da DI, processo diagnóstico e classificação da gravidade é importante para o entendimento das possíveis indicações e limitações das intervenções propostas.

2.1. Etiologia e fisiopatologia da DI

A etiologia da DI pode ser identificada em 40-80% dos casos. As técnicas de sequenciamento de nova geração têm contribuído para a identificação de mutações genéticas que podem estar envolvidos nas situações em que a etiologia não é identificada.

As possíveis causas, sejam pré-natais, perinatais ou pós-natais muitas vezes se sobrepõem. As causas pré-natais podem ser intrínsecas, como síndromes genéticas (Síndrome de Down, Síndrome do X frágil) ou mutações específicas, malformações do sistema nervoso central e distúrbios metabólicos (anemia ferropriva, fenilcetonúria); ou extrínsecas, como intoxicações e exposição a teratógenos (medicações e drogas de abuso). Causas perinatais incluem asfixia neonatal, e a prematuridade, com suas complicações. As causas pós-natais incluem negligência, fatores psicossociais, infecções, trauma e exposição a toxinas [1]. Estas, muitas vezes são importantes fatores de contribuição para a potencialização de um déficit já existente. São também uma possível justificava para a maior prevalência da condição em crianças em situação de pobreza [2].

A fisiopatologia da DI é multifatorial. Uma contribuição da desregulação de vias moleculares, de processos celulares e/ou da organização tecidual, leva a defeitos críticos da estrutura ou função sináptica e conectividade neuronal, comprometendo a capacidade do cérebro de processar informações. O grau do déficit cognitivo depende da extensão desta

desregulação. Outros transtornos podem coexistir com a condição, como o transtorno do espectro autista, o transtorno de déficit de atenção e hiperatividade, e diversas outras doenças neuropsiquiátricas [3].

2.2. Diagnóstico

O diagnóstico da DI é feito pela identificação de limitações significativas nas funcionalidades intelectuais e adaptativas, afetando pelo menos um dos três domínios adaptativos (conceitual, social e prático). Essas limitações devem ser iniciadas ainda no período de desenvolvimento (antes dos 18 anos de idade).

O diagnóstico pode ser formalizado a partir dos 5 anos de idade, quando é possível avaliar as funcionalidades intelectuais de maneira objetiva e confiável, por meio de testes que medem o quociente de inteligência (QI) [4].

O QI é medido utilizando-se baterias de testes que avaliam funcionalidades intelectuais esperadas para cada idade. Linguagem, raciocínio, pensamento abstrato, capacidade de julgamento e de resolução de problemas, dentre outras habilidades são mensuradas. Os testes mais comumente utilizados são as escalas Wechsler de inteligência [2]. A versão para crianças está em sua 5ª edição (2019) [5], sendo que apenas a 4ª edição (publicação original em 2003; adaptada para o português em 2013) está atualmente disponível para a população brasileira [6]. A escala pontua, além do QI total, quatro índices: índice de compreensão verbal, índice de organização perceptual, índice de memória operacional e índice de velocidade de processamento.

As funcionalidades adaptativas também precisam estar afetadas para o diagnóstico da condição. Sua avaliação é feita pela detecção clínica em múltiplos ambientes (escola, casa, comunidade etc.) de prejuízos em, pelo menos, um dos três seguintes domínios:

- Domínio conceitual (habilidades de linguagem: leitura e escrita; compreensão de conceitos: números, dinheiro, tempo; memória; raciocínio; juízo).
- Domínio social (habilidades de comunicação social interpessoal, relacionamentos, empatia, resolução de problemas; autoestima; habilidade de seguir regras; possibilidade de culpabilização).
- Domínio prático (habilidades de cuidado pessoal e de vivência diária: alimentação, higiene; manter rotinas; vestir as próprias roupas) [2].

A escala de comportamento adaptativo, *Víneland*, pode ser utilizada para medir essas funcionalidades de maneira objetiva. Está disponível no Brasil em sua versão atual (3ª edição)[7].

2.3. Classificação da DI e seu impacto na aprendizagem

A DI pode ser graduada de leve à profunda e sua gravidade tem grande impacto na expectativa de aprendizagem. Esta graduação não é feita por intervalos de QI (este não reflete com boa acurácia a funcionalidade, não tendo bom valor prognóstico), mas pelo nível de apoio demandado [1].

DI leve

A DI leve corresponde a cerca de 85% dos casos. O déficit é observado geralmente no final do período pré-escolar ou no início do escolar, quando o nível de exigências acadêmicas aumenta. O pensamento e a comunicação são geralmente mais concretos, havendo prejuízos funcionais em atividades práticas complexas, no entendimento e organização do tempo, e manejo de dinheiro. Há a impressão de que são mais imaturas do que os colegas de classe.

Quando adultos, estes indivíduos necessitarão de auxílio intermitente. A deficiência, no entanto, nem sempre compromete a possibilidade de autonomia e de uma vida independente. O QI destas crianças encontra-se geralmente entre 55 e 70 [1].

DI moderada

A DI moderada corresponde a cerca de 10% dos casos. Nestas crianças o déficit já é percebido de maneira mais precoce. Há dificuldades marcantes na linguagem e no aprendizado, que já são observados no período pré-escolar.

O nível de suporte é maior e mais constante, pois há prejuízo no convívio social, na comunicação, cuidado pessoal (vestir-se, utilizar o banheiro, alimentação etc.). Esse nível de suporte se estende por toda a vida. Quando adultos, são capazes de trabalhos que exigem pouca habilidade comunicativa ou cognitiva. O QI está geralmente entre 40 e 55 [1].

DI grave

A DI grave (3-4% dos casos) é caracterizada por limitação importante na comunicação. Há dificuldades para entender a linguagem verbal e não verbal. Demandam supervisão e auxílio extensivos de cuidadores em todas as atividades diárias, por toda a vida. O QI está geralmente entre 25 e 40 [1].

DI profunda

Na DI profunda (1-2% dos casos), há grave prejuízo na capacidade de abstração. Habilidades conceituais restringem-se ao concreto. Poucas habilidades, inclusive motoras, desenvolvem-se, e quando presentes, geralmente são restritas apenas à manipulação de objetos. A compreensão da linguagem simbólica é prejudicada. Em alguns casos há compreensão de instruções básicas, mas a presença e suporte do cuidador é essencial em todos os aspectos da

rotina diária. O nível de apoio demandado é universal. O QI estimado destes indivíduos é geralmente inferior a 25 [1].

2.4. O que pode ser feito para potencialização da aprendizagem na DI?

A identificação e avaliação de estratégias específicas de potencialização da aprendizagem são o objetivo desta revisão de revisões. Algumas medidas gerais, no entanto, são essenciais para o desenvolvimento adequado das crianças com DI.

Diagnóstico e estímulo precoces

O diagnóstico e estímulo precoces são essenciais na potencialização da aprendizagem dessas crianças. A investigação diagnóstica, em busca da etiologia deve priorizar causas tratáveis (hipotireoidismo, anemia ferropriva, distúrbios metabólicos). A identificação e tratamento precoce podem modificar todo o curso do desenvolvimento, evitando a deficiência, ou reduzindo seu grau. Por outro lado, após a exclusão de causas tratáveis, uma busca obstinada por diagnóstico etiológico pode trazer malefícios para o paciente e família, como a submissão a exames de alto custo. Nestes casos, deve haver compartilhamento com a família da decisão de investigar a etiologia [8].

Uma vez constatado atraso no desenvolvimento ou a deficiência intelectual, a estimulação das habilidades deficientes deve ser imediatamente iniciada, e a criança necessita de acompanhamento multidisciplinar, envolvendo pediatras, educadores, psicólogos, fonoaudiólogos, fisioterapeutas e outras especialidades.

Cuidado integral em saúde

Nessa população, o cuidado integral em saúde é um desafio. Há barreiras para inserção da criança em serviços de saúde, uma vez que os profissionais de saúde nem sempre são treinados para lidar com os comportamentos não colaborativos destas crianças. Sentem-se por vezes incapazes de prestar atendimento. O acesso ao serviço de saúde é complexo. A carga sobre os cuidadores é grande [9].

Apesar destas dificuldades, a inserção na rede básica de saúde é extremamente importante. Os profissionais envolvidos devem incentivar a prática de atividades físicas, que além de combaterem a obesidade (frequente nessa população) [10], estimulam o aprendizado de habilidades motoras.

A educação em saúde também faz parte do processo de aprendizagem da criança com DI visando autonomia: hábitos saudáveis de alimentação, higiene, orientações sobre prevenção de acidentes [10].

Educação inclusiva

O ensino tradicional, em geral, não é adequado para crianças com DI. A inclusão destas crianças na escola, no entanto, é extremamente importante para o desenvolvimento pessoal-social, linguagem e motricidade, além de aprendizagens específicas relacionadas ao conteúdo ministrado. O suporte individual, por meio de tutoria, a instrução com paciência, no ritmo da criança, evitando provocar ansiedade e sentimento de incompetência, levando a desinteresse são medidas gerais para o sucesso da educação.

A medida deste sucesso deve ser diferente da dos demais alunos. Um bom alvo é prover uma educação que equipará a criança para viver da maneira mais independente quanto possível

para sua condição individual, em uma comunidade que pode nem sempre estar totalmente ciente de suas necessidades [11].

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3. OBJETIVOS

3.1. Objetivo geral

Realizar ampla busca na literatura científica por revisões sistemáticas que demonstrem quaisquer estratégias utilizadas para potencialização da aprendizagem, e sintetizar seus resultados, propondo recomendações baseadas em evidências.

3.2. Objetivos específicos

- Sintetizar de maneira narrativa as principais conclusões dos estudos incluídos, organizando as intervenções por tipo e desfecho.
- Realizar avaliação crítica da metodologia de cada estudo incluído, apontando a confiança nos seus resultados.
- Propor recomendações baseadas em evidência que possam ser utilizadas como substrato científico para a elaboração de uma diretriz de cuidado focada na potencialização da aprendizagem durante o desenvolvimento de crianças e adolescentes com deficiência intelectual.

4. METODOLOGIA

4.1. Delineamento

A metodologia utilizada é denominada revisão de revisões sistemáticas, também conhecida como *overview*, revisão guarda-chuva ou meta-revisão. Nela utiliza-se um mecanismo sistematizado de busca na literatura, com critérios de inclusão e exclusão bem definidos, assim como análise de qualidade dos estudos incluídos. O objetivo principal é sumarizar evidências de maneira prática para profissionais ou outros interessados terem acesso a uma ampla quantidade de pesquisa, sem a necessidade de buscar ou avaliar os estudos individualmente. As revisões de revisões estão se tornando cada vez mais populares uma vez que a produção de pesquisa original e de revisões sistemáticas aumenta em ritmo exponencial, sendo impossível para o "tomador de decisão" - aquele que precisa aplicar condutas práticas, baseado em evidências disponíveis - acompanhar toda a produção científica em sua área [1].

As revisões de revisões são mais amplas em seu escopo se comparadas às revisões sistemáticas comuns, sendo capazes de examinar diversas opções de tratamento ou intervenções para determinada área, podendo fornecer diferentes opções terapêuticas visualizadas em um texto amigável ao usuário, facilitando o encontro de informações. Os diferentes tipos de intervenções e seus resultados podem ser comparados, e escolhidos para determinado fim pelo tomador de decisão, de maneira a alinhar-se com sua expertise, ou mesmo valores e preferências[2].

4.2. Elaboração e registro do protocolo

O protocolo foi elaborado baseado nos *Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols* (PRISMA-P) [3], e registrado no *Prospective Register of Systematic Reviews* (PROSPERO) [4]. O número de registro do protocolo é CRD42020195913 e está publicamente disponível no endereço eletrônico: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020195913.

4.3. Critérios para considerar revisões para inclusão

Tipos de estudo

Revisões sistemáticas e meta-análises publicadas a partir do ano 2000, que investigaram o impacto de estratégias de melhoria da aprendizagem nos seguintes desfechos em crianças com deficiência intelectual: cognição, desenvolvimento pessoal-social, habilidades motoras grosseiras ou finas-adaptativas, linguagem, autonomia, saúde e qualidade de vida. Não houve restrição por linguagem.

Os critérios de exclusão foram:

- Revisões não sistemáticas.

- Revisões que focaram exclusivamente em melhoria comportamental sem desfechos diretamente relacionados a melhoria da aprendizagem.

- Revisões relacionadas a estudos observacionais, pesquisa laboratorial; resumos; opiniões pessoais; cartas e pôsteres.

- Revisões publicadas antes do ano 2000.

Foi decidido pela restrição de data de maneira a concentrar os resultados das pesquisas em intervenções modernas, adequadas ao contexto global atual.

Tipos de participantes

Foram incluídas todas as revisões sistemáticas ou meta-análises que incluíram crianças e adolescentes com diagnóstico de DI. A idade mínima para o diagnóstico é 5 anos, desta maneira, a idade de interesse deste estudo foi de 5-18 anos.

Tipos de intervenção

Qualquer tipo de intervenção com foco em praticar ou ensinar habilidades e conceitos foram considerados para inclusão nesta revisão de revisões.

Comparadores

Como comparadores foram incluídos: nenhuma intervenção, tratamento usual, ou comparação entre intervenções.

Tipos de medida de desfecho

Qualquer medida qualitativa ou quantitativa da aprendizagem e de suas consequências com relação a autonomia, saúde e qualidade de vida.

4.4. Métodos de busca na literatura

Fontes de informação

As estratégias de pesquisa sistematizada (Apêndice 1) foram elaboradas e adaptadas para cada uma das seguintes bases eletrônicas utilizadas: Medline, Cochrane, Scopus, Web of Science, Lilacs, SciELO, ERIC, and PsycINFO.

A pesquisa nas bases de dados foi concluída em 31 de maio de 2021.

O aplicativo Mendeley Desktop (v1.19.4) ® foi utilizado para organização e manejo das referências.

4.5. Coleta e análise de dados

Seleção das revisões

Dois revisores, de maneira independente, avaliaram títulos e resumos para identificar estudos elegíveis, utilizando o aplicativo online Rayyan ® (*Qatar Computing Research Institute*). Após este passo, foi realizada leitura do texto completo pelos mesmos dois revisores. Todas as discrepâncias foram resolvidas por discussão até consenso ser estabelecido.

Os estudos excluídos após leitura completa e as justificativas para exclusão estão detalhadas no Apêndice 3.

Extração e manejo de dados

Dois revisores, de maneira independente, extraíram os dados pertinentes que foram posteriormente checados entre si para acurácia. Informações coletadas foram: autor, ano de publicação, revista científica, título, objetivos ou perguntas da pesquisa, número e tipo dos artigos incluídos, população, cenário, intervenção, comparação, desfechos, resultados,

conclusão, ferramentas de avaliação de qualidade e viés, financiamento e conflitos de interesse. Todos esses dados estão disponíveis no Apêndice 2.

4.6.Avaliação da qualidade metodológica das revisões incluídas

A ferramenta AMSTAR-2[5] foi utilizada para avaliar a qualidade das revisões incluídas. Esta ferramenta foi desenhada para avaliação crítica de revisões sistemáticas. Inclui 16 questões que abrangem a completude da pergunta clínica, o registro de um protocolo, a justificativa dos critérios de inclusão, o método de pesquisa, a qualidade da pesquisa na literatura e da extração dos dados, a descrição dos estudos excluídos e das justificativas para exclusão, avaliação do risco de viés, a qualidade da meta-análise (quando aplicável), e a presença de financiamento e de conflitos de interesse. Destas 16 questões, 7 são consideradas domínios críticos (vide Tabela 1 do artigo na seção resultados).

Essa ferramenta permite classificar a confiança global nas conclusões da revisão em alta (nenhuma ou uma falha não crítica), moderada (mais de uma falha não crítica), baixa (uma falha crítica, com ou sem falhas não críticas), criticamente baixa (mais de uma falha crítica com ou sem falhas não críticas).

Dois revisores preencheram de maneira independente o checklist do AMSTAR-2. Discrepâncias foram discutidas até haver consenso.

4.7. Síntese dos dados

Considerando a amplitude dos tipos de intervenções, as revisões incluídas foram divididas em categorias do domínio do desenvolvimento relacionado ao seu desfecho principal

(pessoal-social, motor fino-adaptativo, linguagem, motor grosso). Após, foram divididas pelo tipo de intervenção utilizada.

As categorias de intervenções foram:

- Intervenções do cuidador: intervenções promovidas por pais, familiares ou cuidadores;

- Intervenções educacionais: promovidas por professores em qualquer contexto/cenário.

- Intervenções farmacológicas/dietéticas: intervenções farmacológicas ou nutricionais.

- Fisioterapia, educação física ou terapia ocupacional: intervenções profissionais para aprendizagem motora.

- Tecnologia: intervenções que utilizaram quaisquer equipamentos tecnológicos (tablets, smartphones, realidade virtual etc.).

Os resultados foram sintetizados de maneira narrativa, e sumarizados em recomendações. Uma descrição completa de todos os estudos incluídos pode ser encontrada no Apêndice 2.

4.8. Aspectos éticos

Este estudo foi uma revisão de revisões da literatura. Não foram utilizados dados pessoais de pacientes, prontuários, amostras biológicas, ou modelos animais. Desta maneira, não foi necessária aprovação por comitês de ética ou aplicação de termos de consentimento.

Os autores envolvidos não possuem conflitos de interesse com relação ao tema do estudo.

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5. RESULTADOS E DISCUSSÃO – ARTIGO DE REVISÃO DE REVISÕES

An overview about what has been done to improve learning in intellectual disability

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ABSTRACT

Background: Intellectual disability (ID) affects 1 to 3% of the pediatric population, and teaching these children basic living skills, as well as optimizing their mental capital — resulting in better adaptability —, is a challenge. There is no current consensus as to the most effective strategies, and the extant literature is broad but still uncertain.

Objectives: This overview aims to compile the most useful information and strategies for intervening in the learning of intellectually disabled children and adolescents, aiding clinical practice, decision making, and family care.

Methods: A research protocol has been registered on PROSPERO under the number CRD42020195913. We searched the databases Medline, Cochrane, Scopus, Web of Science, Lilacs, SciELO, ERIC, and PsycINFO to amass systematic reviews and meta-analyses published since the year 2000 which investigated the impacts of multiple types of interventions on the learning of the selected population. Methodological quality has been assessed using the AMSTAR-2 tool.

Main results: 59 studies were selected after the full-text examination. Those were divided by outcome domains and subdivided by the type of intervention. Outcome's domains were personal-social, fine motor-adaptive, gross motor, or language-related. Interventions were related to caregiving, education, medication and diet, physical therapy, and similar strategies, or technology.

Authors' conclusions: Recommendations were made for each outcome domain using the evidence gathered in the studies. The data shows benefits of: caregiver stimulation of specific skills with careful explanation of activities, corrective feedback and engaging in play-behavior; education with systematic instruction (e.g., high probability command sequences), and embedded instruction, with the support of behavioral analysis strategies; specific motor

interventions, encouraging sport practicing, physical activities and animal-assisted therapy; the use of augmentative and alternative communication, and technological devices for support learning. The overall low methodological quality of the studies is a limiting factor for most recommendations, and the successful implementation will also depend on caregivers' training, participation, and socio-economic status.

Keywords: Intellectual disability, learning, cognition, social skills, language, motor skills, personal autonomy, health, quality of life.

BACKGROUND

Intellectual disability (ID) is defined by limitations in both intellectual functioning (measured by an Intelligence Quotient lower than 70) and adaptive behavior (American Psychiatric Association, 2013) — which, like the term's life skills and independent living, includes being able to conform to and to interact with different environments and minimizing social conflicts. This condition affects 1 to 3 percent of the pediatric population (Purugganan, 2018).

Intellectually disabled children and adolescents often struggle in the process of learning in different contexts, from academic concepts to basic living skills, which translates to low functionality and a high level of dependence. The absence of specific health programs that include general and academic skill acquisition and social inclusion, marginalizes those individuals, which in turn creates poor employability and low quality of life in adulthood (Garrels & Sigstad, 2021).

Maximizing functioning through rehabilitative approaches and treating associated health problems are the main goal of therapeutics, which aims to potentiate the quality of life, autonomy, independence for self-care, academic performance, and social inclusion (Kishore et

al., 2019). The provision of effective care for this population is a challenge because of the wide array of known and unknown etiologies (van Karnebeek et al., 2014), the frequent presence of comorbidities (Coppus, 2013), and the uncommon use of efficient teaching and learning strategies (Cornelius & Balakrishnan, 2012). Moreover, little conclusive research is available, which broadens the scope of possible courses of action, while simultaneously weakening the overall efficacy of the treatments provided.

In childhood, evidence-based multidisciplinary follow-up with health providers and caregiver support is necessary to assess and treat the underlying cause, when possible (e.g., iron deficiency anemia), and the comorbidities such as malnutrition, obesity, motor and language impairments, and inappropriate behavior; and, to stimulate the developmental process through the instruction of new skills. In adolescence, effective training skills may impact adulthood employability and social inclusion. Furthermore, sexual education and education about risky behavior may prevent diseases and addictions.

The Medical Subject Headings (Learning - MeSH - NCBI) definition for learning is "*Relatively permanent change in behavior that is the result of past experience or practice. The concept includes the acquisition of knowledge".* A possible implication is that learning skills by experience and practice and acquiring knowledge in its different forms may amplify the ID children's adaptive behavior and functional repertoire. Therefore, this overview aims to assemble the extant body of academic literature regarding different approaches to the improvement of learning in intellectually disabled populations.

OBJECTIVES

We reviewed systematic reviews of the literature to compile the currently available strategies to improve learning and to propose a rational, pragmatic, and evidence-based

recommendation of which strategies are best for the improvement of each developmental domain, providing a guide for multidisciplinary teams involved in the care of ID children.

METHODS

Protocol registration

The protocol was elaborated based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Protocols (PRISMA-P) (Page et al., 2020), and registered at the Prospective Register of Systematic Reviews (PROSPERO). It holds the registration number CRD42020195913 and is publicly available at:

https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020195913.

Criteria for considering reviews for inclusion

Types of studies

Meta-analyses and systematic reviews published from the year 2000 onwards that investigated the impact of learning improvement strategies on the following domains: cognition, personal-social, fine motor-adaptive, and gross motor abilities, language, personal autonomy, health, and quality of life of children and adolescents with intellectual disability. No language restriction was applied.

The exclusion criteria were: (a) studies that did not meet the minimum criteria for systematic review; b) reviews focusing exclusively on behavior improvement without outcomes directly related to learning improvement; (c) reviews of observational studies, and laboratory research; abstracts, personal opinions; letters; and posters, and (d) published before the year 2000. We have decided to set this time restraint to limit results to modern interventions adequate to our current global context.

Types of participants

We included any systematic review or meta-analysis that included children or adolescents diagnosed with ID. The minimum age for the diagnosis of ID is 5 years, thus the age range of our interest population was 5-18 years.

Types of interventions

Any type of intervention focused on practicing or teaching skills and concepts was considered for this overview.

Comparators

No intervention, treatment as usual, or comparisons between interventions.

Types of outcome measures

Any qualitative or quantitative measures of learning and its consequences on personal autonomy, health, and quality of life.

Search methods for identification of reviews

Information sources

Systematic search strategies (Appendix 1) were elaborated and adapted for each of the following electronic databases: Medline, Cochrane, Scopus, Web of Science, Lilacs, SciELO, ERIC, and PsycINFO.

Database searching was concluded on 31 May 2021. Mendeley Desktop (v1.19.4) was used for reference management.

Data collection and analysis

Selection of reviews

Two independent reviewers screened titles and abstracts to identify eligible studies using the online software (Rayyan®, Qatar Computing Research Institute). Full-text reading of eligible studies was performed by the same two reviewers. Any discrepancies were resolved by discussion until a consensus was reached between the reviewers. Excluded studies after full-text reading and the justification of exclusion are detailed in Appendix 3.

Data extraction and management

Two independent reviewers extracted pertinent data that were further cross-checked for accuracy. Information collected was author, year of publication, journal, title, objectives or research questions, number, and type of included primary studies, population, setting, intervention, comparison, outcomes, results, conclusion, bias and quality assessment tools, and funding and conflict of interests. See Appendix 2 for extracted data.

Assessment of the methodological quality of included reviews

AMSTAR-2 (Shea et al., 2017) was used to assess the quality of the included reviews. It is designed to be a practical critical appraisal tool of systematic reviews. It includes 16 questions, approaching the completeness of the clinical question, the registration of a protocol, the rationale for inclusion criteria, the search method, the quality of search and extraction, the studies exclusion justification, the risk of bias assessment, meta-analysis correctness when applicable, and funding or conflict of interest assessment. Of those questions, 7 are considered critical domains (see Table 1).

This tool allows rating the overall confidence of the review conclusions in high (no or one non-critical weakness), moderate (more than one non-critical weakness), low (one critical flaw with or without non-critical weaknesses), critically low (more than one critical flaw with or without non-critical weaknesses).

Two authors independently completed the AMSTAR-2 checklist. Discrepancies were discussed until consensus.

Data synthesis

Considering the broad range of types of interventions, we divided the reviews into categories of developmental domain related to the main outcome (personal-social, fine motor-adaptive, language, or gross motor skills), which were further divided by the type of intervention applied. A complete description of all included studies can be found in Appendix 2.

The categories of interventions were:

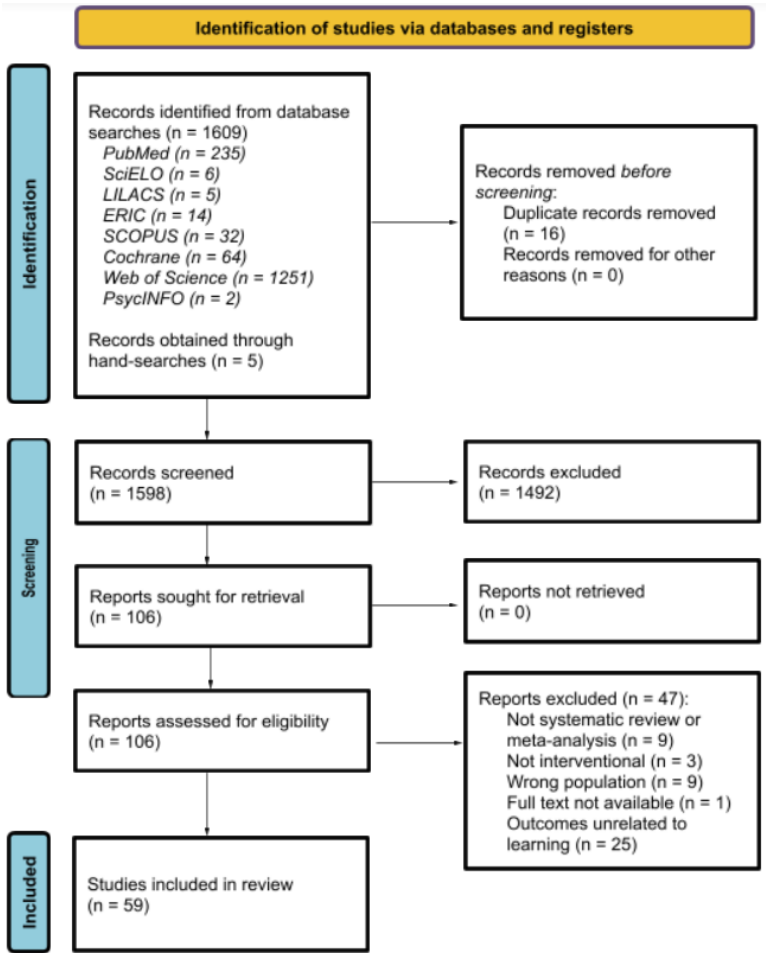
- The caregiving category includes interventions promoted by parents or caregivers; or actions that aimed to enable caregivers for interventions.
- The educational category includes teacher-led interventions in any setting.

- The pharmaco-dietary category included pharmacological and nutritional interventions.
- Physical therapy, physical education, or occupational therapy category included professional intervention on motor skills.
- The technology category included any use of technological devices (e.g., tablets, smartphones, Virtual Reality).

We narratively synthesized the results and discussed findings summarizing data into recommendations.

RESULTS

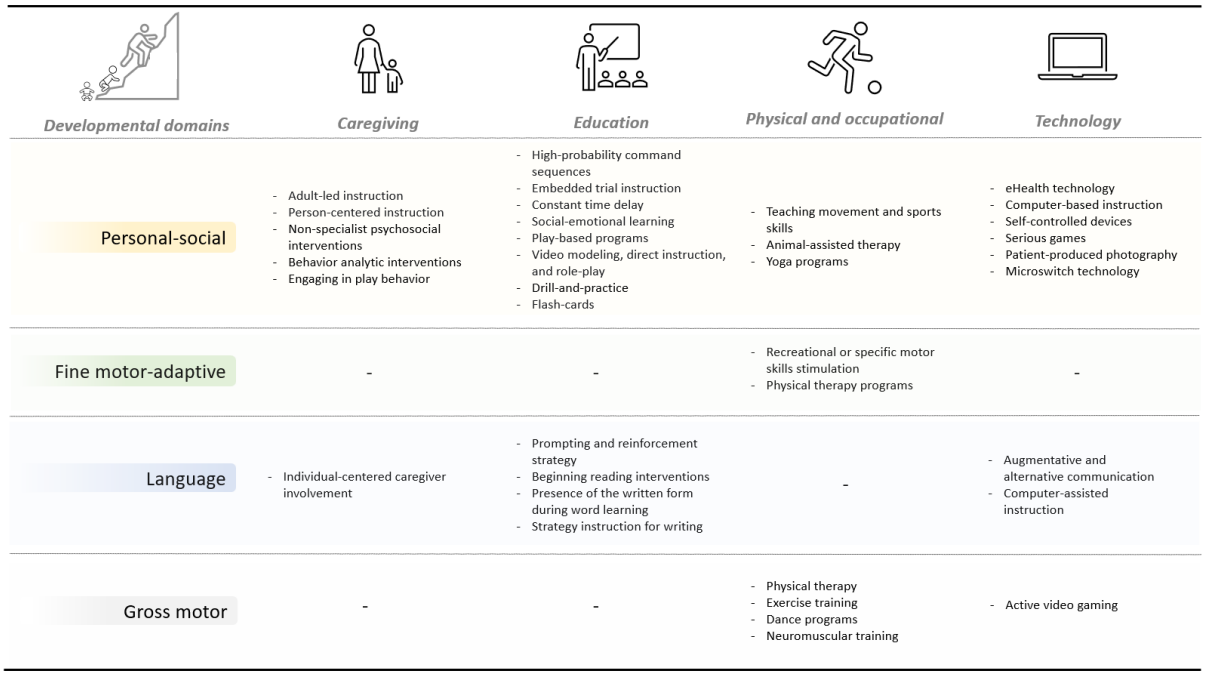
Figure 1. Study flow diagram.



Description of included reviews

Our search yielded 1609 records. 106 systematic reviews and meta-analyses were selected based on titles and abstracts. 59 articles were included in this overview after the full-text examination.

The potentially beneficial interventions found were described in the Figure 2.



Developmental domains	Caregiving	Education	Physical and occupational	Technology
Personal-social	- Adult-led instruction - Person-centered instruction - Non-specialist psychosocial interventions - Behavior analytic interventions - Engaging in play behavior	- High-probability command sequences - Embedded trial instruction - Constant time delay - Social-emotional learning - Play-based programs - Video modeling, direct instruction, and role-play - Drill-and-practice - Flash-cards	- Teaching movement and sports skills - Animal-assisted therapy - Yoga programs	- eHealth technology - Computer-based instruction - Self-controlled devices - Serious games - Patient-produced photography - Microswitch technology
Fine motor-adaptive	-	-	- Recreational or specific motor skills stimulation - Physical therapy programs	-
Language	- Individual-centered caregiver involvement	- Prompting and reinforcement strategy - Beginning reading interventions - Presence of the written form during word learning - Strategy instruction for writing	-	- Augmentative and alternative communication - Computer-assisted instruction
Gross motor	-	-	- Physical therapy - Exercise training - Dance programs - Neuromuscular training	- Active video gaming

Figure 2 – Potentially beneficial strategies separated by developmental domains of the main outcome evaluated and type of interventions.

Personal-social outcomes

Personal-social outcomes were the most frequent aim of the included reviews. Thirty-four articles were designated to this domain, including four meta-analyses and twenty-nine systematic reviews. The interventions applied to attain this outcome consisted of caregiving, educational, pharmaco-dietary, physical, and technological programs.

Caregiving interventions

One meta-analysis and three systematic reviews analyzed interventions related to caregiving.

Hong et al., 2016, conducted a meta-analysis to ascertain whether family-implemented interventions improved the social and other communicative behaviors of individuals with developmental disabilities, and they found moderate effects in the two types of instructional methods: adult-led instruction, which consists of one-on-one instruction carried out in a structured setting; and person-centered instruction, that consists in instruction developed based on interests, carried out in a naturalistic setting. No statistically significant difference between the two strategies was observed (critically low confidence).

Montgomery et al., 2008, investigated the role of personal assistance in developmental outcomes, daily living skills, and other outcomes of interest such as health and quality of life, concluding that it may have some benefits for some recipients and may benefit caregivers, but is a costly intervention (critically low confidence).

Non-specialist psychosocial interventions could be an alternative when professional care is not easily available. A meta-analysis of Reichow et al., 2013, demonstrated that cognitive rehabilitation, training, and support (e.g. reading programs, language instruction) delivered by non-specialists (e.g., mental health care providers who are not psychiatrists, psychologists, or psychiatric nurse practitioners) were demonstrated effective for improving developmental outcomes to ID children, and maximum effectiveness was reached when the training was conducted in clinical settings. Behavior analytic interventions (treatments based on the science of applied behavior analysis) seem useful for daily skills improvement, and parent training for delivering those interventions was also beneficial (low confidence). Target skills (e.g., engaging in play behavior, correcting summing problems, correcting written letters)

can be improved by interventions implemented by siblings, such as demonstration or explanation of the skill, and feedback (critically low confidence) (T. Kim & Horn, 2010).

Educational interventions

One meta-analysis and eight systematic reviews investigated personal-social outcomes of educational interventions. Defiant behavior in classrooms, often reported by educators, could be a barrier to learning. A meta-analysis by (Losinski et al., 2017) evaluated interventions to improve the compliance of students with disabilities in educational settings. Effective instructional delivery (adapting instruction to improve compliance), high-probability command sequences (easy tasks to reinforce compliance), and behavior-specific praise are the most studied interventions. High-probability command sequences were rated as a potentially evidence-based strategy, and further studies should address this intervention (critically low confidence).

Hudson et al., 2013, reviewed the experimental research on academic learning by students with moderate and severe ID in general education. Embedded trial instruction (using the natural environment for teaching) was found to be an evidence-based practice. Practitioners are encouraged to use embedded instructional trials with constant time delay (delay to prompt after instruction) (critically low confidence).

Social-emotional learning strategies were studied by (Hagarty & Morgan, 2020). Play-based programs, used with children aged 5 to 8 and 6 to 13, appeared to be effective in improving social skills. Social skills programs based on behavioral psychology and social learning theory, including techniques such as video modeling, direct instruction, and role-play, appeared to be effective for children aged between 8 and 16 (critically low confidence).

Butler et al., 2001, assessed different mathematics instruction strategies for students with mild to moderate ID. Encouraging but preliminary results have shown that cognitive strategies are better learned when frequent feedback, explicit instruction, and ample drill-and-practice are employed, with improvement in student independence and mathematics performance (critically low confidence). Hord et al., 2012, explored the impact of the changes in mathematics education on students with mild intellectual disabilities. Conceptual and procedural instruction was used, with most of the very limited extant literature being focused on procedural instruction, namely flashcards. Multiple studies in the review suggest teachers can rely on flashcards as an educational tool for teaching mathematical facts to students with mild ID. By teaching students with mild ID methods for storing/registering information on paper, while working through challenging, and multi-step problems, teachers give these students an important tool for overcoming deficits in working memory (critically low confidence).

Kuntz & Carter, 2019, focused on interventions for secondary school students and found a broad portfolio of beneficial interventions, highlighting systematic instruction (e.g., task direction, corrective feedback), peer support arrangements, self-management strategies, peer-mediated communication interventions, and educational placement changes, with positive changes to academic, social, and behavioral skills (critically low confidence).

Walton & Ingersoll, 2013, conducted a review investigating how to improve social skills in adolescents with autism and severe to profound intellectual disability and found that video modeling, developmental/relationship-based interventions, peer-mediated interventions, behavioral interventions, and structured teaching, all positively affected social skills. However, the evidence is weak (critically low confidence).

Relationship mediation and sexuality education programs for adolescents with ID are important in holistic education. Brown et al., 2020, searched for the design, content, and

delivery strategies of those programs. They found that two professional groups are key in delivering the programs' content: teachers and nurses. ID adolescents should be educated to build their knowledge, skills, and confidence. These programs should be designed by collaborative work that includes people with disabilities, their families, and professionals (critically low confidence).

Gilson et al., 2017, sought to review the instructional methods used to teach intellectually disabled secondary students' employment-related skills. They found that between self-management, video-based, audio-based, picture and tactile-based, direct, augmentative and alternative communication (AAC)-assisted, simulation, and peer-delivered instructions, most shows important positive effects (critically low confidence). However, the strategies are varied and not comparable.

Pharmaco-dietary interventions

Most of the research on pharmacological interventions is directed to treat fragile X syndrome (one meta-analysis and three systematic reviews). Three other systematic reviews addressed pharmacological or dietary interventions on other conditions.

In a meta-analysis, Rueda et al., 2015, investigated the effect of L-acetylcarnitine (LAC) compared to placebo for treating fragile X syndrome (FXS), measuring psychological and learning capabilities, behavior, or social performance, safety, and caregiver burden. Their findings did not support LAC as an effective treatment for children with FXS (high confidence). Those results about LAC are like those found back in 2009, in a systematic review of the same author about pharmacological treatments in FSX. In this study, dextroamphetamine and methylphenidate in patients with an additional diagnosis of attention deficit hyperactivity disorder (ADHD), and ampakine compound CS516 for autism and FXS patients. Results were

not robust enough to support recommendations on pharmacological treatments for this population, considering intelligence, behavioral, emotional, and/or learning capabilities (critically low confidence).

In 2011, Rueda et al. published a systematic review of 5 randomized clinical trials (RCTs) on folic acid administration versus placebo on borderline to severe ID patients with FXS, including children. None of the studies found evidence of clinical benefit of folic acid medication in FXS either in psychological and learning capabilities or behavior and social performance (low confidence). Thomson et al., 2009, examined the effectiveness of methylphenidate and dextroamphetamine on ADHD symptoms of children with ID due to FXS. The results show no improvement with either amphetamine, with increased adverse effects (critically low confidence).

In a Down syndrome population, dietary supplements and drugs on cognitive function were reviewed in Salman, 2002. Vitamin B5, 5-hydroxytryptophan, vitamins and minerals, vasopressin, piracetam, niacin, pituitary extract, thiamine. None of the eleven trials reviewed reported cognitive-enhancing effects in subjects with Down syndrome. The quality of the trials was poor with few subjects and generally inadequate allocation concealment of the treatments given (critically low confidence).

Tarrant et al., 2018, reviewed the effectiveness of methylphenidate in the management of attention deficit hyperactivity disorder (ADHD) in people with ID, including children and teenagers, concluding that there is weak evidence to support a beneficial effect of MPH in this population initially, but the evidence is lacking to either support or refutes its use in the long-term (critically low confidence).

A large systematic review of van der Vaart et al., 2015, included 169 trials concerning the treatment of cognitive deficits in genetic disorders. Various dietary and pharmacological interventions were tested for different conditions, such as Down syndrome, phenylketonuria,

Fragile X syndrome, Turner's syndrome, Prader-Wille syndrome, and Rett's syndrome, and Niemann-Pick disease type C. Despite many reports presenting positive conclusions in the abstracts, only 2 treatments have established clinical impact on cognitive outcomes: dietary restriction for phenylketonuria and miglustat for Niemann-Pick disease type C (critically low confidence).

Physical therapy, physical education, or occupational therapy interventions

One meta-analysis and five systematic reviews were found for physical interventions. A meta-analysis of Kapsal et al., 2019, evaluated the effects of physical activity on psychological, emotional, behavioral, and social outcomes, beyond physical health. Teaching movement and sports skills benefit physical and psychosocial health, especially when the activity is social, performed in groups (e.g., basketball, soccer) (critically low confidence). A systematic review of the physical and psychosocial correlates of Special Olympics participation among individuals with intellectual disability from Tint et al., 2017, included 46 studies and 3931 participants with mild to severe ID. Positive changes in participants' self-perception and general self-worth were described. Physical activities seem to increase adaptability, but the results were mixed. Other positive outcomes described were positive emotions, motor skills, increasing social skills and friendships, and social inclusion (critically low confidence).

Animal-assisted therapy's (AAT) effects on social and motor skills were assessed by Charry-Sánchez et al., 2018, sixty one Down syndrome participants, in two included reviews, were submitted to unorthodox animal-assisted interventions: interactions with elephants and dolphins. The study using elephant-assisted therapy demonstrated significantly higher visual-motor integration scores, while the dolphin study demonstrated significant time-group interactions and long-term verbalization benefits (critically low confidence). Those exotic

interventions are not easily available for most ID children. Maber-Aleksandrowicz et al., 2016, reviewed more accessible animal-assisted therapy interventions, including mainly dogs and horses. They concluded that AAT may be a potentially useful supportive intervention for improving the quality of life, and to increase social interaction, improving communication, cognitive functions such as concentration or attention, and emotional well-being, but good quality research is lacking (critically low confidence).

Yoga programs in schools were evaluated by Serwacki & Cook-Cottone, 2012, as it relates to ID, autism spectrum disorder (ASD), and other children. Positive effects on academic, cognitive, and psychosocial outcomes were described, but the quality of evidence is low, and more methodologically rigorous trials are needed to reach definitive conclusions or recommendations (critically low confidence).

Barton et al., 2015, evaluated sensory-based treatments for children with disabilities. Interventions were mainly in classroom or clinical settings and focused mainly on ASD children, and Down Syndrome children. Learning outcomes such as academic performance, language, motor skills, and adaptive behavior were assessed. Sensory-based treatments are more likely to be ineffective than effective for children with disabilities (critically low confidence).

Technology interventions

We have found eight studies regarding technology interventions for personal-social outcomes. Kim et al., 2017, evaluated the effectiveness of tablet-assisted instructions (TAI) for improving academic learning in disabled children. The results show that TAI may have the potential to enhance learning for this population, but it cannot yet be considered an evidence-based practice (critically low confidence).

Boulton et al., 2018, set out to examine the effects of patient-produced photography on emotional, educational, and behavioral levels, mostly in educational settings. The results suggest an improvement in patients' sense of value, empowerment, self-esteem, learning, communication, and attention (critically low confidence).

Oudshoorn et al., 2020, weighed the evidence of the use of eHealth technology, including smartphones, earpieces, and instructional videos, to support the learning of practical daily living skills by mostly ID individuals. The evidence has shown promise, but more research is needed (critically low confidence).

Ramdoss et al., 2012, set out to review the effectiveness of computer-based interventions (CBI) for teaching intellectually disabled individuals' life skills, such as purchasing items at grocery stores, placing orders in restaurants, and using public transportation. 93% of the participants showed positive results, and follow-up reported skill maintenance. The study concludes that CBI is a promising intervention strategy for teaching daily living skills to individuals with moderate ID. (critically low confidence).

Roche et al., 2014, reviewed studies reporting on the use of microswitch technology (i.e., microphone, vibration, lever, and optic switches) to enable self-determined responding in children with profound and multiple disabilities. The results supported this type of intervention in educational programs for this population (critically low confidence).

Den Brok & Sterkenburg, 2015, investigated the use of self-controlled devices, such as mobile devices, virtual reality, and robots, concerning learning and independence for people with ID and some with an additional diagnosis of ASD. This review has found promising advances in using technology to support attaining new daily living skills and cognitive concepts (e.g., time perception, imagination) in persons with moderate to mild ID and/or ASD (critically low confidence).

Kagohara et al., 2013, has reviewed 15 studies using iPads and iPods for teaching developmentally disabled individuals with ASD and/or ID academic, communication, leisure, employment, and transition skills. The study then concluded that these electronic devices are viable aids in the instruction of this population (critically low confidence).

Tsikinas & Xinogalos, 2019, researched the use of serious games (SG) for improving conceptual, practical, and cognitive skills of youth with ID. Positive effects were improvements in social interaction, motor skills, attention and understanding, working memory, punctuation and comprehension, and cognitive skills, and thus SGs can be used for enhancing learning (critically low confidence).

Fine motor-adaptive outcomes

Three systematic reviews were designated to this domain, making it the least frequent objective of the included studies. Physical therapy, physical education or occupational therapy, and technological approaches were used.

Physical therapy, physical education, or occupational therapy interventions

A single systematic review met the inclusion criteria. Houwen et al., 2014, reviewed the evidence related to motor interventions designed to improve motor, cognitive, and/or social outcomes in people with severe and profound intellectual disabilities. Results reported improvement in basic motor skills and recreational or more specific motor skills (critically low confidence).

Technology interventions

Two systematic reviews were included in this category. Hickman et al., 2017, examined the evidence on the use of active video gaming (e.g., Nintendo Wii Sports) for improving motor function in children with movement disorders, including Down Syndrome (DS). There was clear support for their use to improve general motor function (e.g., fine motor precision and integration, manual dexterity, strength, visual perception, motor coordination, and postural movement control) in children with neuromotor conditions. However, the literature did not support the use of active video gaming as a standalone intervention capable of creating permanent neuromotor improvements (critically low confidence).

Kokol et al., 2020, assessed the application of serious games (such as Wii Fit games) in children with developmental disabilities, including DS. Wii-Fit games in addition to the traditional physical therapy program yielded a significant improvement of balance, postural stability, limits of stability, upper-limb coordination, manual dexterity, and running speed and agility standard scores (critically low confidence).

Language outcomes

Sixteen studies spanning caregiving, educational, and technology interventions have been included. The second most frequent outcome in the selected articles gathered seven meta-analyses and eleven systematic reviews.

Caregiving interventions

One meta-analysis and one systematic review have met the inclusion criteria, both regarding parent-related interventions. Liao et al., 2021's meta-analysis was performed to determine the effects of caregiver involvement for promoting communication skills for

individuals with ASD and ID. This intervention is defined as "an individual-centered connection between caregivers at home and professionals in school settings (e.g., teachers, therapists, and service providers) who share responsibility for the development of individuals with ASD and ID". Results indicated that this promotes reduction in caregivers' stress and anxiety, and improvement in caregiver-child interaction, parental quality of life, the understanding of the condition, and in the child's communication skills (critically low confidence).

O'Toole et al., 2018, have conducted a systematic review to assess the effects of parent-mediated interventions for improving communication and language development in young children with Down syndrome. A small number of studies reported gains in language and socialization, and with very low certainty of evidence (high confidence).

Educational interventions

Two meta-analyses and three systematic reviews have been included. Neil & Jones, 2018, meta-analysis aimed to identify effective intervention strategies for communication in individuals with Down syndrome. Most studies used a prompting and reinforcement strategy. Morphosyntactic grammar intervention, manualized reading, and language intervention, and speech recasting, among other interventions, were also present. The results suggest that behavior analytic strategies are a promising approach, and future research should focus on replicating the effects of these interventions with greater methodological rigor (critically low confidence).

Reichow et al., 2019, performed a meta-analysis of seven randomized clinical trials (RCT) to assess the effectiveness of interventions for teaching beginning reading skills to children and adolescents with intellectual disabilities. Beginning reading interventions that

include elements of phonological awareness, letter-sound instruction, and decoding are associated with small-to-moderate improvements in phonological awareness, word reading, decoding, expressive and receptive language, and oral reading fluency (high confidence).

Colenbrander et al., 2019, had the purpose to determine when, why, and how the presence of a word's written form during instruction aids vocabulary learning, a process known as orthographic facilitation. The findings from a small number of studies suggest that the presence of the written form during word learning is likely to be beneficial, with improvements in vocabulary acquisition, for beginning readers and English language learners, as well as children with Down Syndrome (critically low confidence).

Joseph & Konrad, 2009, demonstrated effective methods for teaching writing to students with intellectual disabilities. Among the types of instruction reviewed, strategy instruction yielded the strongest writing performance outcomes for students with intellectual or developmental disabilities, and self-regulated strategy development was the most frequently used (critically low confidence).

Machalicek et al., 2010, reviewed evidence-based literacy instruction for students with physical and developmental disabilities who use aided augmentative and alternative communication (aided AAC). Improvements were reported for 37% of participants, and positive participant outcomes appear to be related to the use of evidence-based systematic instructional procedures such as direct instruction, least to most prompting, and scaffolding of child communication. However, the certainty of the evidence for an intervention effect was rated as inconclusive for 67% of the reviewed studies, and therefore, positive results must be interpreted with caution (critically low confidence).

Technology interventions

Three meta-analyses and six systematic reviews have been selected. All but one study sought to shed light on augmentative and alternative communication (AAC). Ganz et al., 2017, conducted a meta-analysis report on the social-communication outcome results of research on the use of high-tech AAC, including mobile devices, voice-output communication aids, and speech-generating devices, by individuals with intellectual and developmental disabilities. Implementation of high-tech AAC was effective and likely to be effective under most conditions and for most participants with intellectual and developmental disabilities to improve communication skills. Unfortunately, most of the authors evaluated the expression of wants and needs and few evaluated the impact of more social and more complex communicative behaviors, thus, these results must be interpreted with caution (critically low confidence).

Kent-Walsh et al., 2015, focused their meta-analysis on AAC partner instruction interventions to determine the effects on the AAC user. The partners were instructed on the communication of individuals using AAC, with strategy instruction or individual skill training, and the results have shown that communication partner instruction has positive effects on the communication performance of individuals using AAC (critically low confidence).

Millar et al., 2006, assessed the effect of both aided and unaided AAC on the speech production of individuals with developmental disabilities in their meta-analysis. In the six best-evidence studies, 94% of the participants showed improvements in speech production in different instructional approaches. However, none of the evidence was conclusive (critically low confidence).

Barbosa et al., 2018, analyzed research findings regarding the different instruments of AAC used in children with Down's syndrome. Out of twelve instruments, the methods most used by the studies were speech-generating devices (SGDs) and the Picture Exchange Communication System (PECS). Beyond aid in communication, which was reported in every

modality of AAC, these instruments appear to increase the interaction between individuals, contributing to their quality of life and self-esteem (critically low confidence).

Chung et al., 2012, reviewed intervention aimed at improving peer interaction outcomes for school-aged children with complex communication challenges who regularly used or might benefit from AAC. The interventions included the introduction, modification, and/or instruction to target participants, their peers, or their parents regarding AAC. Although the configurations of these intervention packages varied, most were found to be effective at increasing measures of peer interaction (critically low confidence).

Mandak et al., 2018, searched for the effects of instruction on single-word reading of individuals who use aided AAC in their systematic review. Instruction had positive effects on reading at the single-word level for individuals across ages and diagnostic categories, with phonological approaches, sight-word approaches, and a combination of these two approaches yielding very large effects. However, the findings must be viewed with caution due to limitations in the number of studies and participants and the reporting of these studies (critically low confidence).

Morin et al., 2018, investigated the use of high-tech AAC to teach social-communication skills to individuals with an autism spectrum disorder or intellectual disabilities who have complex communication needs and to determine if this intervention approach meets the criteria for evidence-based practices. Results suggest that high-tech AAC can be considered an evidence-based practice, although it was not indicated that high-tech AAC is significantly better than low-tech AAC (critically low confidence).

Roche et al., 2014, explored the use of tangible symbols (objects or parts of them to represent situations or ideas) as an AAC intervention for students with developmental disabilities, most of them with visual impairment. 54% of the participants learned how to use

at least one symbol for requesting, choice-making, naming, transitioning, or directing the actions of others (critically low confidence).

Snyder & Huber, 2019, examined the effectiveness of Computer-assisted Instruction (CAI), in most cases associated with systematic instruction, for academic content (literacy, math, written expression, social studies, or science), for individuals with ID. Across all studies, CAI interventions were effective for teaching the targeted academic skills. No difference was shown to teacher-led instruction and generalization results were generally positive (critically low confidence).

Gross motor outcomes

Only one type of intervention for this outcome was found, amassing three meta-analyses and three systematic reviews. The studies in this category are heterogeneous, exploring different populations, interventions, and specific outcomes.

Physical therapy, physical education, or occupational therapy interventions

Jeng et al., 2017, aimed to determine whether exercise training improves skill-related fitness (SRF), which includes agility, power, coordination, reaction time, activity speed, and balance, in adolescents with ID in this meta-analysis. The interventions included but were not limited to circuit training, muscle strengthening, dance, aerobic exercises, games, and martial arts. The results support positive exercise training effects on all fields of SRF except for balance. However, only a limited number of studies exhibited high-quality evidence (critically low confidence).

May et al., 2021, synthesized in a meta-analysis the findings of research exploring the psychological, cognitive, social, and physical effects of dance programs for children with disabilities, including ID. A wide array of dance programs has been used, such as creative, aerobic, and traditional Greek dances and generally positive results have been demonstrated for dance skills, physical fitness, balance/coordination, flexibility/agility, perceptual-motor skills, jumping, and balance. Cognitive, psychological, and social outcomes were usually positive as well. This study concludes that dance may have physical, cognitive, and psychosocial benefits for children with disabilities, despite the high heterogeneity and poor quality of the reviewed literature (critically low confidence).

Sugimoto et al., 2016, conducted a meta-analysis to examine the effects of neuromuscular training on general strength, maximal strength, and functional mobility tasks in children and young adults with Down syndrome. The intervention involved strength/resistance, balance/bicycle riding, vibration, and functional mobility training. There were limited but mostly high-quality studies, the results showed that neuromuscular training could be used as an effective intervention in children and young adults with Down syndrome (critically low confidence).

Fonzo et al., 2020, aimed to analyze the role of physical therapy in the management of individuals with Rett syndrome. Interventions involved, among other types, aided and unaided traditional physical therapy, hydrotherapy, and treadmill exercises. All studies included in this review demonstrated that physical therapy improved the quality of life and autonomy in patients diagnosed with Rett. According to the authors, physical therapy programs should be provided early, individualized, and must include appropriate involvement of the patient's family or caregivers (critically low confidence).

Hocking et al., 2016, studied the effectiveness of physical therapy interventions for improving gross motor skills (GMS) in people with an intellectual disability aged 6 years and

older. Interventions included progressive resistance training, strength exercises, treadmill training, modified judo training, among others. The available evidence provides initial support for the consideration of physical therapy interventions for increasing independence, particularly lower limb exercises for cadence, partial body-weight-supported gait training for cadence and nondimensionalized gait velocity, and adapted judo training (critically low confidence).

Maïano et al., 2019, investigated the effects of motor skill interventions designed to improve fundamental movement skills (FMS) in children and adolescents with ID. Balance and/or strength exercises, adapted play training, and handball techniques were some of the specific interventions covered. In all but one study the intervention groups have significantly higher post-test balance skills, and overall fundamental motor skills than the control groups, whereas findings on locomotor skills were mixed. Given the relatively small number of studies and their limitations, these results are not conclusive (critically low confidence).

Registered protocols of ongoing systematic reviews

We did not find any ongoing systematic reviews registered on PROSPERO.

The methodological quality of included reviews

Since our study aims to impact health policies, the recommendations made must be based on quality reviews. Thus, we used a systematic method to evaluate the quality of included systematic reviews.

AMSTAR 2 is an instrument designed as a practical critical appraisal tool to enable the assessment of the methodological quality of systematic reviews. This tool allows the evaluation of systematic reviews of randomized and non-randomized trials. It is a questionnaire that runs

through the completeness of the review question, registration of the review, methodological quality, and risk of bias.

AMSTAR 2

		AMSTAR 2 DOMAIN																	
O	I	Author	Date	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	CONFIDENCE
Reviews with meta-analyses																				
PS	C	Hong	2016																	CRIT
	E	Losinski	2017																	CRIT
	PD	Rueda	2015																	HIGH
	PT	Kapsal	2019																	CRIT
L	C	Liao	2020																	CRIT
	E	Neil	2016																	CRIT
		Reichow	2019																	HIGH
	T	Ganz	2017																	CRIT
		Kent-Walsh	2015																	CRIT
		Millar	2006																	CRIT
G	PT	Jeng	2016																	CRIT
		May	2019																	CRIT
		Sugimoto	2016																	CRIT
Reviews without meta-analysis																				CRIT
PS	C	Kim	2010																	CRIT
		Montgomery	2008																	CRIT
		Reichow	2013																	LOW
	E	Brown	2020																	CRIT
		Butler	2001																	CRIT
		Gilson	2017																	CRIT
		Hagarty	2020																	CRIT
		Hudson	2013																	CRIT

		Kuntz	2019																CRIT
		Walton	2013																CRIT
	PD	Rueda	2009																CRIT
		Rueda	2011																LOW
		Salman	2002																CRIT
		Tarrant	2018																CRIT
		Thomson	2009																CRIT
		Van Der Vaart	2015																CRIT
	PT	Barton	2015																CRIT
		Charry-Sánchez	2018																CRIT
		Maber-Aleksandrowicz	2016																CRIT
		Serwacki	2012																CRIT
		Tint	2017																CRIT
	T	Boulton	2018																CRIT
		den Brok	2015																CRIT
		Kagohara	2013																CRIT
		Kim	017																CRIT
		Oudshoorn	2020																CRIT
		Ramdoss	2011																CRIT
		Roche	2015																CRIT
		Tsikinas	2019																CRIT
A	PT	Houwen	2014																CRIT
	T	Hickman	2017																CRIT
		Kokol	2020																CRIT
L	C	O'Toole	2018																HIGH
	E	Colenbrander	2019																CRIT
		Hord	2012																CRIT
		Joseph	2009																CRIT
		Machalicek	2010																CRIT
		Roche	2014																CRIT
	T	Barbosa	2018																CRIT
		Chung	2012																CRIT
		Mandak	2018																CRIT

		Morin	2018																CRIT
		Snyder	2019																CRIT
G	PT	Fonzo	2020																CRIT
		Hocking	2016																CRIT
		Maïano	2019																CRIT

1. PICO	Did the research question and inclusion criteria include the components of the PICO? (yes/no)
2. Protocol	Was a complete protocol written? (yes/partial yes/no)
3. Study design	Did the authors explain their selection of study designs for inclusion in the review? (yes/no)
4. Search strategy	Comprehensive search strategy? (yes/partial yes/no)
5. Study selection	In duplicate? (yes/no)
6. Data extraction	In duplicate? (yes/no)
7. Excluded studies	List of excluded studies and justification? (yes/partial yes/no)
8. Included studies	Included studies described in detail? (yes/partial yes/no)
9. Risk of bias	Risk of bias assessed? (yes/partial yes/no)
10. Funding sources	Funding sources reported? (yes/no)
11. Meta-analysis	Appropriate methods used? (yes/no/no meta-analysis)
12. Impact risk of bias	Did the authors assess the impact of RoB on results? (yes/no/no meta-analysis)
13. Discussing the risk of bias	Did the authors discuss the potential effects of bias? (yes/no)
14. Heterogeneity	Discussion of any heterogeneity? (yes/no)
15. Publication bias	Investigation of publication bias? (yes/no/no meta-analysis)
16. Conflicts of interest	Did the review author report any conflicts of interest in conducting the review? (yes/no)

Color coding

- Methodological requirements met
- Methodological requirements partially met
- Methodological requirements not met
- Not applicable (no meta-analyses)
- Amstar 2 critical domains

Type of outcomes coding

PS *- Personal-social;* ***A*** *- Fine-motor/adaptive;* ***L*** *- Language;* ***G*** *- Gross motor*

Type of intervention coding

C *- Caregiving;* ***E*** *- Educational;* ***PD*** *- Pharmaco-dietary;* ***P*** *- Psychotherapy;* ***PT*** *- Physical therapy, physical education or occupational therapy;* ***T*** *- Technological.*

Types of confidence coding

HIGH *- High overall confidence in the results of the review;* ***MOD*** *- Moderate overall confidence in the results of the review;* ***LOW*** *- Low overall confidence in the results of the review;* ***CRIT*** *- Critically low overall confidence in the results of the review*

DISCUSSION

In this overview, we have searched for systematic reviews and meta-analyses that aimed to evaluate the evidence of interventions to improve learning in intellectual disabilities. We further categorized those studies by the developmental domain of their main outcome (personal-social, fine motor-adaptive, language, and gross motor) and created sub-categories for each field of intervention (caregiving, educational, pharmaco-dietary, physical therapy, physical education or occupational therapy and technological interventions). Our objective was to identify and provide an evidence-based guide to health providers, teachers, or any professional or caregiver involved in ID rehabilitation.

Quality of the evidence

AMSTAR 2

The confidence in the results was classified as high in 3 studies, low in 2 studies, and critically low in 54. The most important flaw identified in the systematic reviews was concerning the search strategy. The methodological criteria in this critical domain were not achieved in 51 studies, mainly due to language restriction to only studies in English without justification.

Most reviews did not register a protocol (46), did not use a risk of bias tool (43), and did not provide a list of excluded studies with justifications (47). All these criteria are considered critical domains.

Despite the confidence of a study being classified as critically low, the critical appraisal should consider all the other items and strengths, once registration of a protocol, use of a risk of bias tool, and not disclosing excluded studies will not always have a major impact in the estimated effect.

We considered, however, the exclusion of studies in other languages as the most important limitation, once education and learning strategies are intricately linked to cultures and retrieving studies from non-English speaking countries would have provided valuable insight into other systems of teaching and learning. The inclusion of literature from different countries and languages is advised for future research to amplify the range of available strategies.

Summary of main results

Despite the overall low quality of the systematic reviews, we were able to identify potentially beneficial strategies (Figure 2). Thus, we propose general recommendations for each type of intervention. That makes it possible to combine strategies according to the socio-economic possibilities, values, and preferences of caregivers and adaptation of the children.

Caregiving

Interventions delivered by caregivers should be at the center of the rehabilitation process concerning learning in ID. Daily skills learning is context-dependent, and families can be taught how to train and support those children to learn specific skills related to the family routine. **Careful explanation of the skill, feedback, engaging in play behavior are ways in which caregivers can improve specific skills learning.**

Despite the common belief that involving the caregiver in the rehabilitation process increases the care burden, evidence shows that **this involvement promotes reduction in**

caregivers' stress and anxiety, and overall quality of life, and improves the social performance of children.

Thus, family-delivered interventions should be encouraged.

Educational

Formal education is often a challenge in this population since defiant behavior and non-compliance in the classroom or other educational settings are a common problem beyond the limitation in the abilities to learn concepts. **High probability command sequences were rated as the most effective strategy for learning**, and **behavior-specific praise can add to minimize the negative effects of misbehavior**. These actions should be taken with planning and adapting content and learning expectations according to the level of the child.

For this, **embedded trial instruction seems to be an evidence-based practice to be implemented**. It refers to using and arranging the natural environment and pre-existing routines towards learning, gaining the students' interest, for instance, social-emotional skills can be taught in play-based programs (Hagarty & Morgan, 2020).

Education interventions should have the purpose of teaching adaptive abilities and should not be a role exclusively of the teacher. **Video modeling, developmental or relationship-based interventions, and peer-mediated interventions can be used for teaching**. In specific contexts, such as sexual education, other trained professionals, such as health professionals, may be necessary, and family collaboration and acceptance of content are necessary. Family education in this theme also can be required.

Systematic instruction (task direction, corrective feedback) seems to be useful for academic learning, beyond social and behavioral benefits. Methods of systematic instruction were found for language and mathematics.

Behavior analytic strategies (prompting and reinforcement strategy) could facilitate language learning. Literacy instruction is the main goal of structured interventions. Reading is an important adaptive ability for social performance and personal autonomy and can be reached using elements of phonological awareness, letter-sound instruction, and decoding instruction (Reichow et al., 2019), and orthographic facilitation (Colenbrander et al., 2019). Concerning writing, the self-regulated strategy (e.g. STOP strategy: Suspend judgment, Take a side, Organize ideas, Plan) was the most promising intervention (Joseph & Konrad, 2009).

Dealing with numbers is frequently necessary for daily activities. Math concepts are often too complex for teaching to ID children. To facilitate this, constant feedback, explicit instruction, and ample drill-and-practice (Butler et al., 2001), as well as flashcards and diagramming information in word problems, teaching students to store information on paper, and step by step instructions (Hord et al., 2012) could be useful.

Technological tools also can assist formal education (see Technological interventions).

Pharmaco-dietary

Various pharmaco-dietary interventions were included in this overview. Most of the research was directed to treating fragile X syndrome. **No consistent benefit of tested drugs (L-acetylcarnitine, dextroamphetamine, methylphenidate, ampakine compound CS516, folic acid) was demonstrated, and they should not be used routinely in clinical practice**.

There is, however, **evidence of the effectiveness of methylphenidate in the management of ADHD in children and teenagers with other etiologies of ID** (Tarrant et al., 2018).

Diverse pharmaco-dietary interventions were tested in the Down syndrome population, aiming for cognitive improvement (vitamin B5, 5-hydroxytryptophan, vitamins and minerals,

vasopressin, piracetam, niacin, pituitary extract, thiamine) but results do not show beneficial effects (Salman, 2002), and **they should not be recommended for this objective**.

Physical therapy, physical education, or occupational therapy

Physical practices and sports should be encouraged, as they were demonstrated to benefit not only physical health, fitness, and motor skills, but psychosocial health, especially if the activity is of social nature.

Specialized motor interventions also should be implemented, with benefits in learning basic motor skills, recreational motor skills seem from profound to mild ID. This recommendation also applies to Rett syndrome patients (Fonzo et al., 2020), with benefits also demonstrated in autonomy and quality of life.

Concerning occupational therapy strategies, while **sensory-based treatments are likely to be ineffective and should not be routinely recommended**, **animal-assisted therapy was found to improve social abilities and cognitive functions, and it was shown to be a useful support intervention**.

Technological

Technological strategies are the trending type of intervention in the reviewed literature, with a progressively increasing number of publications in the last 20 years. Among those, low to high-tech AAC was the most reported (11 studies). Findings showed improvement in communication skills for most individuals (Ganz et al., 2017; Morin et al., 2018), word reading (Mandak et al., 2018), speech production (Millar et al., 2006), interaction, quality of life, and self-esteem (Barbosa et al., 2018). Thus, **those techniques should be implemented**, and **partners and peers should be trained to improve benefits for the user communication** (Chung et al., 2012; Kent-Walsh et al., 2015).

Other technologies were demonstrated to be beneficial in supporting learning in an academic context. Computer-assisted (Snyder & Huber, 2019), tablet-assisted (Kim et al., 2017), and smartphones were studied (Oudshoorn et al., 2020); for children with profound ID, microswitch technology could enable self-determined responding.

Despite most evidence being weak, the great interest of children in technology and the inevitable use of those devices daily by children and adolescents **make a case for the exploration of these tools to support learning, both in a formal and structured way and informally and recreationally**. Games are an interesting possibility to this goal (Hickman et al., 2017).

Games structured to learn lead children to achieve improvements in conceptual and practical skills, social interaction, motor skills, literacy, and the learning of new daily living skills. Thus, since games probably will be present in the daily lives of intellectually disabled children, we suggest that **serious games should be recommended as a supporting tool for learning in this group.**

Application of recommendations for severe to profound ID

Most of the general recommendations stated are better applied for children and adolescents with mild to moderate ID, since are mainly based on systematic reviews from which most of the sample are classified in this level of severity.

We consider, however, that some of the recommendations should be highlighted for the severe to profound ID population. **The use of microswitch technology to enable self-determined responding; developmental/relationship-based, peer-mediated interventions, and inclusion of these children in the motor intervention and physical activities** seems to

be beneficial and should be tried observing individual results, and child and caregiver adaptation and satisfaction.

Potential biases in the overview process

We aim to perform a comprehensive review. However, gray literature has not been searched. Since this is an umbrella review, we have considered that there is a low risk of relevant systematic reviews being published elsewhere than formal literature. Nevertheless, this is a possible source of bias.

We have also limited the searches to more recent literature (starting from the year 2000). We have considered that previous literature would most likely provide obsolete strategies, not adequate to our current context, and those that still are applicable would be reviewed in more recent systematic reviews.

Authors' conclusions

This overview provided a comprehensive evidence-based summary for strategies that could be implemented in clinical, familial, and scholarly practice to improve learning in ID children.

Recommendations were made for each developmental domain. The low quality of the systematic reviews and meta-analyses reviewed is a limiting factor to most of the recommendations. Future studies should use more rigorous methods to generate high-quality evidence.

Applicability will depend on socio-economic contexts and familiar conditions. Providing familial and staff training, improving school structure, and facilitating access to multidisciplinary follow-up are policies that could improve the learning, and consequently the

personal autonomy, independence, social inclusion, health, and the quality of life of children with ID.

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6. APÊNDICES

Appendix 1 - Search strategies

Search terms used in all databases:
1- ((intellectual* OR mental*) AND (disab* OR retard*)) AND (therapy OR intervention) AND ((learning OR motor OR language OR social) AND (development* OR skill*)) AND child*
2- Intellectual disability AND (learning OR cognition OR language OR motor skills OR personal autonomy OR health OR quality of life)
3- education of intellectually disabled AND child AND (learning OR personal autonomy OR cognition OR language OR motor skills OR health OR quality of life)

Search strategies and filters used per database:

MEDLINE
SEARCH PERIOD: 05/2020-03/2021
TYPES: Meta analyses and systematic reviews
PUBLICATION RANGE: 2000 onwards
SPECIES: Human
AGES: Adolescent; Child (6-12); Child (birth-18); Infant (1mo-23mo); Infant (birth-23mo)
SORTED BY: Best match

SciELO
SEARCH PERIOD: 08/2020-03/2021
TYPES: Revision articles

LILACS
SEARCH PERIOD: 08/2020-03/2021
TYPE OF STUDY: Systematic reviews
PUBLICATION RANGE: 2000-2021

ERIC
SEARCH PERIOD: 08/2020-03/2021
USAGE OF * NOT RECOGNIZED, REMOVED FOR THIS DATABASE
FILTERS USED: Intervention; Children; Disabilities; Information Analyses (FOR PUBLICATION TYPE)
PUBLICATION RANGE MANUALLY CHECKED

SCOPUS
SEARCH PERIOD: 09/2020-03/2021
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STUDY TYPE: Intervention

Appendix 2 – Detailed description of included reviews

1. Personal-social

1.1. Caregiving

Meta-analysis

Hong (2016), Review Journal of Autism and Developmental Disorders.

Title: A Meta-Analytic Review of Family-Implemented Social and Communication Interventions for Individuals with Developmental Disabilities.

Objectives: To present a meta-analysis of single-case experimental research on family-implemented interventions for improving the social and other communicative behaviors of individuals with developmental disabilities.

Included studies: 40 single case studies.

Population: Preschool and elementary school children, adolescents, and adults with developmental disabilities. Sample: 156 participants (14 ID or DS). Range of average age for DD participants: 2-32. Range of average age for ID/DS children: 3-6.

Setting: Typically, home, community, or structured settings.

Intervention: Adult-led instruction or person-centered instruction, both using behavioral techniques, such as modeling, fading, prompting, reinforcement, and time delay.

Comparison: Pre-treatment baselines.

Outcomes: Vocalization, social behaviors, nonverbal communication, AAC use.

Results: This meta-analysis provided empirical support for family-implemented interventions, with no statistically significant difference between adult-led instruction and person-centered instruction. Those may be effective in improving the social and other communicative behaviors for early aged individuals with DD, and may be broadly applied for individuals with DD who are elementary school-aged and younger and can be utilized for individuals with DD regardless of their level of communication/language skills.

Conclusion: Overall, the results indicate that family-implemented interventions have a moderate effect on the social and other communicative behaviors of individuals with DD.

Bias and quality assessment tools: What Works Clearinghouse Design Standard.

Funding and conflict of interests: The authors declare no competing interests.

AMSTAR 2: critically low confidence.

Systematic reviews

Kim (2010), Topics in Early Childhood Special Education.

Title: Sibling-Implemented Intervention for Skill Development with Children With Disabilities.

Objectives: To review the currently available evidence on the effectiveness of sibling-implemented interventions in which siblings serve as the intervention agent for teaching age-appropriate skills to their siblings with disabilities.

Included studies: 8 single-subject designs: 7 multiple baselines, 1 withdrawal.

Population: Children with disabilities. Sample: 23 children (3 ID, 2 DS) and 24 siblings. Age range for disabled children: 3-10. Age range for ID children: 5-10. Age range for siblings: 5-12.

Setting: Therapeutic.

Intervention: Demonstration, explanation or discussion of the targeted skills or strategies, role-playing with trainers and/or practicing with siblings, and providing feedback. Some studies used videotapes for presenting examples or modeling or stories for illustrating the use of the targeted skills.

Comparison: Multiple baseline or withdrawal.

Outcomes: Behavior modification (appropriate response to sibling's elicitation to engage in play behavior, engagement in stringing bead, correcting addition problem, correcting written letter, decreasing out-of-seat behavior, correct responding to the siblings' behavioral modification); Social behavior (social initiation, social response, joint attention, attending to the sibling/on-looking, stereotypical behavior); Language or communication behavior (increased utterances, increased use of specific language target, increased spontaneous use of language target, all verbal behaviors and a topic-related verbal turn); Domestic living skills (bed making, table setting). The studies with ID participants fall into the two last categories.

Results: Across the 8 studies, most targeted children (23/24) demonstrated positive gains in targeted skills or behaviors across the intervention phases, and similarly, positive gains in targeted skills or behaviors were noted for the majority of siblings (22/24) as a function of the intervention. Across the 7 studies reporting maintenance data, positive maintenance effects were reported for 15/18 of target children and 22/23 of siblings. Across the 6 studies that reported generalization data, positive generalization effects were reported for 12/16 of target children and 16/19 of siblings. Across the 5 studies reporting social validity, all of them reported positive results: that the means of intervention or after-intervention were significantly higher than the means of baseline; or that the sibling pairs showed more positive feelings toward each other according to questionnaires filled by parents; or that the participants enjoyed taking part in the study according to a survey.

Conclusion: According to the study, sibling-implemented intervention can be considered an evidence-based practice.

Bias and quality assessment tools: Horner et al. (2005)'s standards of evidence for single-subject research.

Funding and conflict of interests: The authors declare no funding or conflicting interests.

AMSTAR 2: critically low confidence.

Montgomery (2008), Cochrane.

Title: Personal assistance for children and adolescents (0-18) with intellectual impairments (Review).

Objectives: To assess the effectiveness of personal assistance for children and adolescents with intellectual impairments, and the impacts of personal assistance on others, compared to other interventions.

Included studies: 1 RCT.

Population: Children and adolescents with intellectual disabilities. Sample: 1002 participants. Age range: 3-17 y.

Setting: Home.

Intervention: Personal assistance (defined as paid support of at least 20 hours per week).

Comparison: 20h of paid care compared to 15h of paid care per week.

Outcomes: Quality of life, user satisfaction, unmet needs, physical health, impact on others, and costs.

Results: This review indicates that personal assistance may have some benefits for some recipients and may benefit caregivers. Paid assistance might substitute for informal care and cost the government more than alternative arrangements; however, the relative total costs to recipients and society are unknown.

Conclusion: Further studies are required to determine which models of personal assistance are most effective and efficient for particular people.

Bias and quality assessment tools: Cochrane Handbook.

Funding and conflict of interests: Grant from the Swedish government, the Unit for Disabilities Issues and the Institute for Evidence-Based Social Work Practice, the Swedish National Board of Health and Welfare (Socialstyrelsen). Reviewers declare no known conflicts of interest.

AMSTAR 2: critically low confidence.

Reichow (2013), PLoS Medicine.

Title: Non-Specialist Psychosocial Interventions for Children and Adolescents with Intellectual Disability or Lower-Functioning Autism Spectrum Disorders: A Systematic Review.

Objectives: To provide an appraisal of which interventions for children and adolescents with intellectual disabilities or lower-functioning autism spectrum disorders delivered by non-specialist care providers in community settings produce benefits in development, daily skills, school performance, behavior, or family outcomes when compared to either a no-treatment control group or treatment-as-usual comparator.

Included studies: 29 studies (15 RCT, 14 prospective controlled studies).

Setting: School, home, childcare, or clinic.

Population: Children and adolescents with ID or both ID and ASD. Sample: 1305 participants (409 ID only, 541 also ASD, 355 mixed groups). Age range for ID-only groups: 0-16 y. Age range for ID and ASD groups: 0-8 y. Age range for mixed groups: 0-16 y.

Intervention: Non-specialist behavior analytic interventions, cognitive rehabilitation, training, and support, or parent training interventions.

Comparison: No-treatment control group or treatment-as-usual.

Outcomes: Development, daily skills, school performance, behavior, and family outcomes.

Results: For the behavior analytic interventions, the best outcomes were shown for development and daily skills, especially for children with more severe levels of cognitive impairment at treatment onset. Cognitive rehabilitation, training, and support were found to be most effective for improving developmental outcomes in children with intellectual disabilities between 6 and 11 y of age, with mixed effects shown for daily skills and

school performance. Parent training interventions are most effective for improving developmental, behavioral, and family outcomes when training is conducted in clinical settings.

Conclusion: The findings of this review support the delivery of psychosocial interventions by non-specialist providers to children who have intellectual disabilities or lower-functioning autism spectrum disorders.

Bias and quality assessment tools: Adaptation of Cochrane Collaboration's Risk of Bias Tool.

Funding and conflict of interests: The review was supported by a WHO-owned fund by the name of Autism Speaks. The authors have declared no conflict of interest.

AMSTAR 2: low confidence.

1.2. Educational

Meta-analyses

Losinski (2017), Education and Treatment of Children.

Title: A Meta-Analysis of Interventions to Improve the Compliance of Students with Disabilities.

Objectives: To systematically review and analyze the extant literature on interventions to improve the compliance of students with disabilities in the schools.

Included studies: 28 single-case design studies.

Population: Children and adolescents with disabilities. Sample: 89 participants. Age range: 4–14 y. Participants had the following disabilities: ID, emotional disturbance, ASD, learning disability, developmental delay, or at risk of disability.

Setting: Education setting, resource room, self-contained classroom, day-care, special education school, and hospital.

Intervention: Effective instructional delivery (8), high-probability command sequences (6), behavior specific praise (4), desensitizing (2), dog training (2), functional behavioral assessment/behavioral intervention plans (1), sound-field amplification (1), errorless compliance training (1), self-monitoring (1), token reinforcement (1), and video self-modeling (1).

Comparison: Baseline.

Outcomes: Compliance.

Results: Only high-probability command sequences can be rated as potentially evidence-based, while all of the other interventions would be classified as insufficient evidence.

Conclusion: Authors suggest that used strategies should be subjects of high-quality replications to determine the extent of their effectiveness for improving non-compliance in students with disabilities.

Bias and quality assessment tools: Council for Exceptional Children Standards for Evidence-based Practices (2014).

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Systematic reviews

Brown (2020), International Journal of Environmental Research and Public Health.

Title: The design, content and delivery of relationship and sexuality education programs for people with intellectual disabilities: A systematic review of the international evidence.

Objectives: To identify the design, content, delivery, and evaluation approaches used in RSE programs for people with intellectual disabilities across the lifespan.

Included studies: 12 included studies: qualitative designs (9), and quantitative methods (3).

Population: Individuals with ID and educators or other supporters. Sample: 168 individuals with ID and 919 educators or other supporters. Educators and supporters included: peer educators, family caregivers, professionals, and front-line staff, special school educators, teachers, school staff, program developers, parents, and support staff.

Setting: Mainly schools. South Africa (3), Sweden (3), Australia (2), Canada (1), Netherlands (1), United Kingdom (1), and the United States (1).

Intervention: Relationship and Sexuality Education (RSE) Programs.

Comparison: Between types of interventions.

Outcomes: Design, content, delivery, or evaluation of RSE programs specific to people with ID

Results: There is evolving research regarding the design and delivery of RSE programs to address ID participants specific issues. It is recommended that to be acceptable, people with learning disabilities, their families and professionals must all work collaboratively on the program content design. Two key professional groups that emerged as those most involved in the delivery of RSE programs are schoolteachers and registered nurses. It is necessary to provide access to education and practice development initiatives that build their knowledge, skills, and confidence.

Conclusion: It is yet to be established what the impact of RSE programs are in effecting long-term behavior change. There are no longitudinal studies regarding the development of positive and healthy relationships, intimacy, and the expression of sexuality by people with ID. These are aims of RSE programs and are therefore areas requiring further research attention.

Bias and quality assessment tools: Mixed-methods appraisal tool.

Funding and conflict of interests: The researchers declare no funding or conflict of interests.

AMSTAR 2: critically low confidence.

Butler (2001), Mental Retardation.

Title: Teaching Mathematics to Students with Mild-to-Moderate Mental Retardation: A Review of the Literature.

Objectives: To identify and analyze mathematics interventions for students with mild-to-moderate mental retardation.

Included studies: 16 studies. 4 group studies and 12 single-subject studies (1 pre-post, 1 AB, 1 ABABC, 1 alternating treatment, 1 ABCD with multiple probes, 4 multiple baselines, 2 multiple probes, 1 multiple problem).
Population: ID students. Sample: 271 individuals. Age ranges from one study: 8-16 y.
Setting: Elementary school (13), junior/high school (2), special education schools (1).
Intervention: For basic skills, discriminant learning theory (1), cross-age tutoring (1), and equality training (1). For computation, concrete-representational-abstract sequence (1), constant time delay (2), multisensory (1), time trials (1), peer tutoring (2), and technology (3). For problem solving, self-regulation (1) and technology with strategy (2).
Comparison: Direct instruction, control groups, other methods, multiple baselines.
Outcomes: Basic skills (e.g., counting, recognition of numerals, addition) (3), mathematics computation (10), and problem solving (3).
Results: Students with mental retardation learned to employ cognitive strategies effectively when the strategy instruction included frequent feedback, explicit instruction, and ample drill-and-practice, especially in the problem-solving studies and in the concrete–representational–abstract study. Strategy instruction promoted student independence in addition to increasing mathematics performance. Computer use did not significantly improve mathematics performance when compared to traditional teacher-directed instruction. However, computers were used successfully when students practiced cognitive strategies with specially designed software.
Conclusion: Preliminary results from these studies are encouraging, but additional research is needed.
Bias and quality assessment tools: Not reported.
Funding and conflict of interests: Not reported.
AMSTAR 2: critically low confidence.

Gilson (2017), Research and Practice for Persons with Severe Disabilities.
Title: Systematic Review of Instructional Methods to Teach Employment Skills to Secondary Students with Intellectual and Developmental Disabilities.
Objectives: To systematically review research on instructional methods used to teach employment skills to secondary students with IDD.
Included studies: 56 studies. RCT (3), single case studies (53), including multiple baseline/probe (45), alternating treatment (5), withdrawal (2), and reversal (1).
Population: Secondary students with IDD. Sample: 766 students (78,8% ID, 9,5% autism, 11,7% both). Age range: 14-22.
Setting: Workplace (10), employment setting (17), school (21), or other (8).
Intervention: Self-management instruction (12), video-based instruction (12), audio-based instruction (3), picture and tactile-based instruction (7), direct instruction (11), augmentative and alternative communication (AAC)–assisted instruction (6), simulation instruction (3), and peer-delivered instruction (2). Regarding instructional methods, studies commonly reported using performance feedback (36), device-assisted instruction (34), response prompting (33), and community-based instruction (32).

Comparison: Control groups or baseline.

Outcomes: Measures used to evaluate the efficacy of the intervention (e.g., task analysis, rate of target behavior), types of employment-related tasks students were performing during data collection (e.g., clerical, assembly), and whether any aspect of the measurement addressed social-related skills (i.e., any interactions with another person during task completion).

Results: All eight intervention approaches had at least 75% of studies with strong positive or positive effects. All group-design studies showed a strong positive differential effect between treatment and control groups. 60.4% of the interventions evaluated within single-case design studies had strong positive effects, 16.7% had positive effects, and 8.3% had mixed effects. Comparative studies showed null or negligible differences. The efficacy of these interventions is demonstrated widely across a variety of participants, settings, and outcomes.

Conclusion: Literature offers considerable evidence of the efficacy of intervention approaches used to teach employment skills that can be drawn upon in schools. Strong employment skills instruction and vocational training are needed in schools during the transition years to help prepare students with IDD for integrated employment.

Bias and quality assessment tools: Council for Exceptional Children Standards for Evidence-Based Practices.

Funding and conflict of interests: The authors report no conflict of interest and declare a grant from the US Department of Education.

AMSTAR 2: critically low confidence.

Hagarty (2020), Educational Psychology in Practice.

Title: Social-emotional learning for children with learning disabilities: a systematic review.

Objectives: To examine the current state of evidence regarding both behavioral social skills training and broader programs aiming to teach social-emotional skills.

Included studies: 12 experimental or quasi-experimental designs: RCTs (2), Single-Case Experimental Designs (5), one-group pre-post (3), and non-randomized experimental design (2).

Population: Intellectually disabled children. Sample: 225 participants. Age range: 5-19.

Setting: Schools. USA (3), Iran (1), Israel (1), Australia (2), Turkey (1), Nigeria (1), Cyprus (1), Hong Kong (1), Switzerland (1).

Intervention: Social-emotional curricula (4) (including the “Explore” social skills curriculum, emotional intelligence training, socio-cognitive training based on Cognitive Enhancement Therapy, and a residential program focusing on daily living skills); Behavioral approaches to teach specific skills (5) (including video modelling, educational software, group discussion, direct instruction, rewards, or the “Superheroes” program, e.g. in order to teach participants to greet people or engage with peers at lunchtime); Play-based approaches (2) (including the “Learn to Play” and “Theraplay” programs); Computerized program (1).

Comparison: regular school program, no treatment, waitlist, unknown.

Outcomes: Social skills: Matson Evaluation of Social Skills for individuals with severe retardation, social skill rating system; Communication: Vineland Adaptive Behavior Scale; Socialization; Face Recognition: Benton Face

Recognition Test; Emotion recognition: Performance on computer program developed as part of study; Social interaction: Penn Interactive Peer Play Scale; Social disruption; Social disconnection; Social cognitive composite score; Social awareness: Social Responsiveness Scale; Social motivation; Social Communication.

Results: Small effect of computerized training on facial recognition; large effects of emotional intelligence training on communication, socialization, and adaptive behavior; Large effects in reduced social disruption and social disconnection and a medium effect on increased social interaction; Medium effect of Theraplay intervention on social awareness and communication as well as a small effect on social cognition and motivation. The SCED studies provided convincing evidence of intervention effectiveness. The residential program study did not find statistically significant change in children and young people's social skills.

Conclusion: Play-based programs, used with children aged 5 to 8 y and 6 to 13 y of age, appeared to be effective in improving social skills. Social skills programs based on behavioral psychology and social learning theory, including techniques such as video-modelling, direct instruction and role-play, appeared to be effective for children aged between 8 and 16.

Bias and quality assessment tools: Gough's Weight of Evidence framework

Funding and conflict of interests: Funding not stated. Authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Hord (2012), Education and Training in Autism and Developmental Disabilities Journal.

Title: Review of Academic Mathematics Instruction for Students with Mild Intellectual Disability.

Objectives: To explore the impact of the changes on mathematics education on students with mild intellectual disability.

Included studies: 7 experimental studies. Multiple baselines (6) and comparison between groups (1).

Population: Individuals with mild ID. Sample: 66 participants. Age range: elementary school age-23 y.

Setting: School.

Intervention: Procedural instruction: flashcards, and computational instruction; and Conceptual instruction: teaching precurrent skills.

Comparison: Multiple-baseline and not stated in the comparison between groups design study.

Outcomes: Mathematics learning.

Results: The results suggest two main findings: a) Little attention in research to academically based mathematical interventions for students with mild ID with no current studies focusing on mathematics standards at or above middle school levels of general education curriculum, and b) of the limited existing research, most studies are primarily focused on procedural instruction. Multiple studies in the review suggest teachers can rely on flashcards as an educational tool for teaching mathematical facts to students with MID. There are some benefits of diagramming information in word problems. By teaching students with MID methods for storing information on paper while working through challenging, multi-step problems, teachers give these students an important tool for overcoming deficits in working memory.

Conclusion: The results suggest two main findings: a) Little attention in research to academically based mathematical interventions for students with MID with no current studies focusing on mathematics standards at or above middle school levels of general education curriculum, and b) of the limited existing research, most studies are primarily focused on procedural instruction.

Bias and quality assessment tools: None.

Funding and conflict of interests: Authors did not report funding or provided a conflict-of-interest statement.

AMSTAR 2: critically low confidence.

Hudson (2013), Research & Practice for Persons with Severe Disabilities.

Title: Review of Experimental Research on Academic Learning by Students with Moderate and Severe Intellectual Disability in General Education.

Objectives: To evaluate experimental research on academic learning in general education classrooms for students with moderate and severe intellectual disability.

Included studies: 17 single-case experimental designs (1 ABAB/BABA, 7 multiple-probe, 6 alternating treatment, 2 multiple-baseline, 1 parallel treatments).

Population: Students with disabilities. Sample: 44 participants. Age range: 5-21 y. 35 participants were diagnosed with ID, while others had different diagnoses, such as ASD, PDD, and other disabilities.

Setting: Primary and secondary general education settings.

Intervention: Instructional methods: embedded trial instruction (mainly embedded constant time delay, 9) (10), system of least prompts (2), a combination of peer delivered cues, feedback, and error correction (2), peer support (e.g., partner learning, peer tutoring, classwide peer tutoring) (1), teacher-delivered statements embedded during instruction but not delivered within instructional trials (1), non-embedded constant time delay (1).

Comparison: Non-embedded massed trials in separate classes, different intervention durations, alternating treatments, multiple probes, and multiple baselines.

Outcomes: Academic skills in science (n = 7), composition/spelling (n = 3), mathematics (n = 4), social studies/geography/history (n = 5), ELA/literacy/reading (n = 8), art/health/PE (n = 4), and German (n = 1).

Results: Embedded trial instruction, especially when using constant time delay (embedded CTD), was found to be an evidence-based practice. Promise was found for the use of the system of least prompts and task analytic instruction.

Conclusion: Practitioners are encouraged to use embedded instructional trials with constant time delay for academic learning given the strong research foundation for the effectiveness of this practice.

Bias and quality assessment tools: Horner et al. (2005)'s quality indicators.

Funding and conflict of interests: Not reported.

AMSTAR 2: critically low confidence.

Kuntz (2019), Research and Practice for Persons with Severe Disabilities.

Title: Review of Interventions Supporting Secondary Students with Intellectual Disability in General Education Classes.

Objectives: To provide a comprehensive map of the literature addressing interventions delivered within general education classes to middle and high school students with intellectual disability.

Included studies: 40 (39 single-case designs (32 single-intervention, 5 comparative, 1 ABAB, 1 ABACABAC) and 1 group design).

Population: Students with Intellectual Disability. Sample: 177 students with ID (mild to profound). Age range: ~11-18 y (middle- and high-schoolers). Most students were male (62.1%). Middle and high school. Some studies included participants with ASD or learning disabilities.

Setting: urban schools (32,5%), suburban schools (17.5%), rural schools (7.5%), not specified (27,5%), other descriptors (15%) - i.e., "small town," "metropolitan school district," multiple states, or districts.

Intervention: Systematic instruction (e.g., task direction, corrective feedback, constant time delay, simultaneous prompting), peer support arrangements, self-management strategies, peer-mediated communication interventions, and educational placement changes.

Comparison: Not specified, since only one study had a group design.

Outcomes: Academic (14), behavioral (14), social (17), self-management (2), or other (5) skills.

Results: For the 53 dependent variables measured in the single-case studies, 23 (41.8%) showed strong effects (5 academic, 6 behavioral, 7 social, 2 self-management, 3 other), 8 (16.4%) showed positive effects (6 social, 2 academic), 8 (16.4%) showed mixed effects (5 social, 1 academic, 1 behavioral, 1 other), and 14 (25.5%) showed no effect (5 academic, 6 behavioral, 3 social). In the group study, of 11 dependent variables, eight showed significant improvement (total interactions, focus student contributions, peers contacted, academic engagement, social goal attainment, class participation, friendship gains, and gone from classroom), while three showed no differences across groups (proximity to direct support, social skills ratings, and academic goal attainment). Maintenance was measured in 21 studies from 1 week to more than 1 month after intervention, and all of them showed positive results. Among the single-case design studies, only four studies met all of the relevant quality indicators, while the group design study met all but three relevant quality indicators for its design.

Conclusion: Although the impact of the interventions on most outcomes was positive, the methodological quality of these studies was somewhat varied. The authors recommend strengthening the availability and implementation of effective interventions within inclusive secondary school classes.

Bias and quality assessment tools: Adaptation of the Council for Exceptional Children's standards of methodological quality.

Funding and conflict of interests: Partial support for this research was provided by a doctoral leadership grant from the U.S. Department of Education. The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Walton (2013), Journal of Autism & Developmental Disorders.

Title: Improving Social Skills in Adolescents and Adults with Autism and Severe to Profound Intellectual Disability: A Review of the Literature.

Objectives: To review the existing research on improving social skills in this population; to point out gaps and needs in the existing research; to highlight important clinical considerations for social interventions in this population; and to propose future directions for addressing the social needs of this population.

Included studies: 17 studies. 7 multiple baselines, 3 withdrawal, 2 uncontrolled pre-post, 2 non-randomized trials, 1 microanalysis, 1 alternating treatment, 1 multiple treatment.

Population: Adolescents and adults with autism and severe to profound ID. Sample: 148 participants. Age range 5y - early 60s.

Setting: Hospital, group home, educational setting, family home, institution, restaurant, retirement community

Intervention: Video modeling (1), developmental/relationship-based interventions (2), peer-mediated interventions (3), behavioral interventions (6), structured teaching (5).

Comparison: Direct assistance from teachers or baselines.

Outcomes: Number, duration, and type of social interactions, offering food, behavioral symptoms, incidental teaching, greeting, joint play, and skill range.

Results: All types of interventions positively affected social skills.

Conclusion: Despite early evidence of efficacy for improving positive social behaviors, more research is needed to confirm the efficacy of these interventions.

Bias and quality assessment tools: Not reported.

Funding and conflict of interests: Not reported.

AMSTAR 2: critically low confidence.

1.3. Pharmaco-dietary

Meta-analyses

Rueda (2015), Cochrane Database of Systematic Reviews.

Title: L-acetylcarnitine for treating fragile X syndrome.

Objectives: To review the efficacy and safety of L-acetylcarnitine in improving the psychological, intellectual, and social performance of people with FXS.

Included studies: 2 trials.

Population: Children and adolescents with fragile X syndrome. Sample: 83 male participants. Age range: 6 to 13 y.

Setting: Outpatients and extra care at home and at school. Europe.

Intervention: L-acetylcarnitine (LAC).

Comparison: Placebo.

Outcomes: Psychological and learning capabilities, behavior or social performance, safety, caregiver burden.

Results: No important differences between treatment and placebo (verbal and non-verbal intellectual functioning using the Wechsler Intelligence Scale for Children - Revised). No clear evidence concerning the impact of the treatment on hyperactive behavior using the Conners' Abbreviated Parent-Teacher Questionnaire. Parents' assessments favored LAC in only one study, though changes were not large enough to be considered clinically relevant. Regarding social skills, one study reported no clear evidence of a difference in Vineland Adaptive Behavior composite scores yet results in the socialization domain favored LAC (low-quality evidence). No side effects. Neither of the included studies assessed the secondary outcome of caregiver burden.

Conclusion: There is low-quality evidence showing that LAC has small or uncertain effects on psychological and learning capabilities and behavior performance outcomes of children with FXS. Results for social performance were inconsistent. Our findings did not support LAC as an effective treatment for children with FXS.

Bias and quality assessment tools: Cochrane Collaboration's tool for assessing risk of bias.

Funding and conflict of interests: No external sources of support. Authors declare no conflict of interest.

AMSTAR 2: high confidence.

Systematic reviews

Rueda (2009), BMC Neurology.

Title: Systematic review of pharmacological treatments in fragile X syndrome.

Objectives: To review the literature and summarize the evidence from clinical controlled trials that compared at least one pharmacological treatment with placebo or other treatment in individuals with diagnosis of FXS syndrome and assessed the efficacy and/or safety of the treatments.

Included studies: 14 clinical controlled trials.

Population: Individuals diagnosed with Fragile X Syndrome. Sample: 229 participants. Age range 1-54 y.

Setting: In-patients in 1 study. Not reported in the others.

Intervention: Pharmacological treatments.

Comparison: Placebo or control preparations.

Outcomes: Intelligence and behavioral, emotional and/or learning capabilities.

Results: 10 studies on folic acid (9 with crossover design, only 1 of them with good methodological quality and low risk of bias) did not find in general significant improvements. A small sample size trial assessed dextroamphetamine and methylphenidate in patients with an additional diagnosis of ADHD and found some improvements in those taking methylphenidate, but the length of follow-up was short. Two studies on L-acetylcarnitine, showed positive effects and no side effects in patients with an additional diagnosis of ADHD. One study on patients with an additional diagnosis of autism assessed ampakine compound CX516 and found no significant differences between treatment and placebo.

Conclusion: Currently there is no robust evidence to support recommendations on pharmacological treatments in patients with FXS in general or in those with an additional diagnosis of ADHD or autism.

Bias and quality assessment tools: Cochrane Collaboration criteria.

Funding and conflict of interests: Funded by a grant from the Spanish Ministry of Health. Authors supported by grants of the University of the Basque Country, and by GIRMOGEN (Spanish Research group on mental retardation of genetic origin). The authors declare no conflicting interests.

AMSTAR 2: critically low confidence.

Rueda (2011), Cochrane Database of Systematic Reviews.

Title: Folic acid for fragile X syndrome.

Objectives: To review the efficacy and safety of folic acid in the treatment of people with fragile X syndrome.

Included studies: 5 RCTs.

Population: Individuals diagnosed with fragile X syndrome. Sample: 67 male participants. Age range 1-54 y. Borderline to severe ID.

Setting: not stated.

Intervention: Folic acid administration.

Comparison: Placebo.

Outcomes: Psychological and learning capabilities (Stanford Binet, Leiter or Wechsler IQ tests, Peabody Picture Vocabulary Test Revised). Behavior or social performance (Autistic Descriptors Checklist, Autism Behavior Checklist, Childhood Autism Rating Scale or Conners' Parent and Teaching Rating Scales); activities of daily living (Katz ADL or Lawton IADL). Adverse effects (safety); caregiver burden.

Results: None of the individual studies found evidence of clinical benefit with the use of folic acid medication in fragile X syndrome patients on any of the areas of interest, either psychological and learning capabilities or behavior and social performance, as measured with standardized tools. Adverse effects of folic acid treatment were rare, not serious, and transient.

Conclusion: The quality of available evidence is low and not suitable for drawing conclusions about the effect of folic acid on fragile X syndrome patients. It consists of few studies with small samples of patients, all of them male, with little statistical power to detect anything other than huge effects.

Bias and quality assessment tools: Cochrane Collaboration's Risk of Bias Tool.

Funding and conflict of interests: Sources of support: University of the Basque Country UPV/EHU, Spain. Grant GIU07/07; Iberoamerican Cochrane Centre, Spain; Spanish Ministry of Health, Spain. Grant FIS ETES PI07/90180 D. Authors declare no conflict of interest.

AMSTAR 2: low confidence.

Salman (2002), European Journal of Paediatric Neurology.

Title: Systematic review of the effect of therapeutic dietary supplements and drugs on cognitive function in subjects with Down syndrome.

Objectives: To evaluate the effects of therapeutic dietary supplements and drugs on cognitive function in subjects with Down syndrome.

Included studies: 11 trials (RCTs or pseudo-RCTs).

Population: Individuals with Down Syndrome. Sample: 373 participants. Age range: 0-42 y.

Setting: Inpatients, outpatients, clinic, support groups, institutionalized subjects.

Intervention: Vitamin B6 (2); 5-hydroxytryptophan (1), vitamins and minerals (4), vasopressin (1), piracetam (1), niacin (1), pituitary extract (1), thiamine (1).

Comparison: No treatment or between treatments.

Outcomes: Developmental and IQ scales (Bayley (2), Vineland social maturity scale (1), WISC–R (2), WPPSI (1), Griffiths developmental scale (1), Stanford–Binet IQ scale (5), Word list and visual verbal paired associate task (1), Cognitive test battery, and questionnaire (1).

Results: None of the trials reported cognitive enhancing effects in subjects with Down syndrome. The quality of the trials was poor with few subjects and generally inadequate allocation concealment of the treatments given.

Conclusion: No positive evidence that the tested drugs, vitamins, and minerals enhance either cognitive function or psychomotor development in people with Down syndrome. However, because of the small number of subjects involved and the overall unsatisfactory quality of the trials, an effect cannot be excluded at this point. At present there is no justification for the use of such regimes outside the context of large well-designed trials.

Bias and quality assessment tools: A modification of Prendiville's criteria.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Tarrant (2018), Research in Developmental Disabilities.

Title: The effectiveness of methylphenidate in the management of attention deficit hyperactivity disorder (ADHD) in people with intellectual disabilities: A systematic review.

Objectives: To conduct a systematic review of all the RCTs in people with intellectual disabilities (ID) that assessed effectiveness of MPH on the core ADHD symptoms.

Included studies: 12 double blind RCTs that evaluated the effectiveness of methylphenidate (MPH) in the management of ADHD. Also included one study on adverse events and another one on long term follow up.

Population: Individuals diagnosed with both ID and ADHD. Sample: 315 participants with ID. Age range: 4-26 y.

Setting: Community.

Intervention: Methylphenidate.

Comparison: Placebo.

Outcomes: Clinical global impression scale (CGI); Aberrant behavior checklist (ABC); Conners rating scales; Adverse events; Revised Behavior Problem Checklist; Learning.

Results: On average in around 40%–50% of cases there was some improvement according to selected measures, but it was as high as 72% according to some measures in some of the smaller studies. The rate of adverse events from treatment with MPH in children with ID ranged somewhere between 12–24%, although in some studies the rate was higher - up to 40% for some adverse events.

Conclusion: Initial weak evidence to support a beneficial effect of MPH. Evidence is lacking to either support or refute its use in the long-term.

Bias and quality assessment tools: Cochrane Collaboration's tool for assessing risk of bias.

Funding and conflict of interests: National Institute of Health Research, UK. Conflict of interests not stated.

AMSTAR 2: critically low confidence.

Thomson (2009), Cochrane Database of Systematic Reviews.

Title: Amphetamine for attention deficit hyperactivity disorder in people with intellectual disabilities.

Objectives: To examine the effectiveness of amphetamine for the treatment of attention deficit hyperactivity disorder in people with intellectual disabilities.

Included studies: 1 crossover RCT.

Population: Children diagnosed with both Fragile X Syndrome and ADHD. Sample: 15 participants. Age range: 3-11 y.

Setting: Not reported.

Intervention: Methylphenidate and dextroamphetamine.

Comparison: Placebo.

Outcomes: ADHD symptoms (as measured by the Conners Abbreviated Parent-Teacher Questionnaire and behavioral observation), response as judged by parents and teachers, and side effects.

Results: Amphetamine had no significant effect on symptoms of ADHD compared to placebo, with increased reporting of adverse effects (the most frequent complaints were mood lability and irritability).

Conclusion: The current RCT evidence does not allow conclusions to be drawn about the efficacy or the risk-benefit profile of amphetamine for the treatment of ADHD in children or adults with ID. Prescribing amphetamine in this group can be based only on non-RCT studies and extrapolation of research in people without a diagnosis of ID.

Bias and quality assessment tools: Cochrane Collaboration Handbook.

Funding and conflict of interests: The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Van Der Vaart (2015), JAMA Neurology.

Title: Treatment of cognitive deficits in genetic disorders: A systematic review of clinical trials of diet and drug treatments.

Objectives: To evaluate clinical impact, strengths, and weaknesses of clinical trials of diet or drug treatments to improve cognitive function in patients with a genetic disorder.

Included studies: 169 studies. RCTs (107), clinical trials without a control group (34), and clinical trials with nonrandomized control groups (28).

Population: People with genetic disorders and cognitive deficits. Sample: 6571 individuals with genetic disorders (3159 Huntington's disease, 1580 DS, 842 phenylketonuria, 347 FXS, 252 Turner's Syndrome, 137 PWS, 32 Rett's Syndrome, 222 others). Age range not reported.

Setting: Not stated.

Intervention: Various dietary and pharmacological interventions.

Comparison: Mostly control groups but otherwise not specified.

Outcomes: Cognitive outcomes, such as thinking, problem solving, memory, and learning.

Results: Despite a growing number of trials and many reports presenting positive conclusions in the abstracts, only 2 treatments have established clinical impact, namely, dietary restriction for phenylketonuria and miglustat for Niemann-Pick disease type C.

Conclusion: In addition to enhancement of the quality of preclinical drug studies, improvement in clinical studies is needed to allow the successful translation of early findings and the establishment of new treatments. Current practice may lead to undesired off-label prescription or result in premature abandonment of promising drugs.

Bias and quality assessment tools: Jadad scale for quality of design and CONSORT checklist for quality of reporting and risk of bias in RCT. Non-RCT studies did not have their quality assessed.

Funding and conflict of interests: The authors received funding from The Netherlands Organization for Health Research and Development and declared no conflict of interest.

AMSTAR 2: critically low confidence.

1.4. Physical therapy, physical education or occupational therapy

Meta-analysis

Kapsal (2019), Journal of Physical Activity & Health.

Title: Effects of Physical Activity on the Physical and Psychosocial Health of Youth with Intellectual Disabilities: A Systematic Review and Meta-Analysis.

Objectives: To synthesize the literature and quantify the effects of physical activity on the physical and psychosocial health of youth with intellectual disabilities.

Included studies: 109. Pre/post controlled experimental designs (46.5%); single group pre/post experimental designs (36.1%), post-test-controlled designs (4.5%), involved cross-sectional or observational designs (12.9%).

Population: Youth diagnosed with ID. Sample: 4200 participants. Age range: 5-19.9 y. Levels of ID ranged from mild to severe.

Setting: Not stated.

Intervention: Aerobic training, resistance training, movement/sport skills, general physical activity/education balance/core stability.

Comparison: Control group or pretest-posttest.

Outcomes: Physical health, and psychological, cognitive, emotional, behavioral, social outcomes.

Results: Physical training had a large effect on physical health, and a moderately-large effect on psychosocial health. While resistance training shows the most physical benefits, teaching movement and sports skills appears to benefit physical and psychosocial health. These include specially those of social nature, that are performed in groups, such as basketball, soccer, ball throwing, judo, or table tennis training.

Conclusion: Practitioners seeking the greatest improvements in both the physical and psychosocial health of youth with intellectual disabilities may wish to focus interventions on playing sport and training sports/movement skills.

Bias and quality assessment tools: Downs and Black Quality index was used for risk of bias and quality assessment.

Funding and conflict of interests: Grants from the Australian Research Council and the Social Sciences and Humanities Research Council of Canada. The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Systematic reviews

Barton (2015), Research in Developmental Disabilities.

Title: A systematic review of sensory-based treatments for children with disabilities.

Objectives: To conduct a comprehensive and methodologically sound evaluation of the efficacy of sensory-based treatments for children with disabilities.

Included studies: 30 studies. 15 group research design studies (13 RCTs and 2 quasi-experimental), and 15 single case research studies.

Population: Individuals with disabilities. Sample: 856 participants (49 DS). Age range: 1-19 y.

Setting: Clinic (11), dark room (1), home (3), classroom (14), home and classroom (2), summer program (1), residential program (2), not reported (2).

Intervention: Comprehensive sensory integration (17), sensory perceptual motor training (7), special seating (3), sensory diet (3), massage (2), neurodevelopmental treatment (2), weighted vest (6), snoezelen (environmental alteration) (1).

Comparison: Another treatment (14) or control for group studies and multiple baselines, withdrawal or alternating treatments designs for single studies.

Outcomes: The outcomes were classified into 13 categories (academic, autism symptomatology, problematic behavior, stereotypies, motor, adaptive, attention/engagement, in-seat, sensory-related behaviors, language, goal attainment scaling, play, and other). Other outcomes included goal attainment scaling, language, play, stress, social behaviors, sleep, asking for a break, feeding, balance and walking, and self-esteem.

Results: Sensory-based interventions are prevalent and widely used with children who have developmental disabilities, especially ASD. Based on the author's analysis of this literature, sensory-based treatments are more likely to be ineffective than effective for children with disabilities.

Conclusion: Given the substantial number of rigorous studies, future research on sensory-based treatments might not be necessary until positive neurological effects of sensory-based treatments are effectively and efficiently measured and documented.

Bias and quality assessment tools: Adaptation of Cochrane Collaboration's risk of bias tool; What Works Clearinghouse group design standards.

Funding and conflict of interests: Authors declare no funding or conflict of interests.

AMSTAR 2: critically low confidence.

Charry-Sánchez (2018), Journal of Developmental and Behavioral Pediatrics.

Title: Effectiveness of Animal-Assisted Therapy in the Pediatric Population: Systematic Review and Meta-Analysis of Controlled Studies.

Objectives: To synthesize the results of controlled studies evaluating the effectiveness of animal-assisted therapy (AAT) in children.

Included studies: 26 controlled studies in total. 2 studies with Down syndrome children as the population.

Population: Individuals with Down syndrome. Sample: 61 participants. Age range: 6-12 y.

Setting: Therapeutic setting.

Intervention: Animal-Assisted Therapy. Interventions of both studies were unorthodox, using elephants and dolphins as the therapy animals.

Comparison: Regular school activities; swimming therapy (no animal) or no intervention.

Outcomes: Matson Evaluation of Social Skills for Individuals with Severe Retardation or Bruininks- Oseretsky Test of Motor Proficiency.

Results: 8 participants with Down syndrome were submitted to Elephant-assisted therapy, and motor outcomes were measured using the Bruininks-Oseretsky Test of Motor Proficiency, a postural control record form, and the Beery's Visual-Motor Integration (VMI) Test. No significant differences were observed in outcomes pertaining to balance or postural control. Beery's VMI scores were significantly higher after therapy in the experimental group vs control group (n = 8). 18 participants with DS were submitted to Dolphin-assisted therapy on social skills and outcomes compared with controls of swimming therapy without the animal (n = 10) and a waitlist (n = 17). Significant time-group interactions were observed before and after treatment on the Matson Evaluation of Social Skills for Individuals with Severe Retardation subscales of verbalization and recognition of persons. In follow-up measurements, only the effect on the verbalization subscale remained

Conclusion: Although results are in general positive for the management of children with DS, the diversity of scales used to measure outcomes makes it difficult to establish true effectiveness.

Bias and quality assessment tools: Cochrane Risk of Bias Tool.

Funding and conflict of interests: No funding stated. The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Maber-Aleksandrowicz (2016). Research in Developmental Disabilities.

Title: A Systematic Review of Animal-Assisted Therapy on Psychosocial Outcomes in People with Intellectual Disability.

Objectives: To review the literature on animal assisted therapy (AAT) in people with intellectual disabilities (ID) measuring psychosocial outcomes (behavioral, social, cognitive and emotional).

Included studies: 10 quantitative studies: before and after design (8), alternating treatment (1), controlled clinical trial (1).

Population: People with intellectual disability. Sample: 100 participants. Age range: 5y-40s.

Setting: Rehabilitation center (1), classroom (4), residential facility (1), clinic (1), riding center (2), not reported (1).

Intervention: Animal-assisted therapy. Horse (3), donkey (1), dog (6), guinea pig (1).

Comparison: Before and after, alternating treatment or control.

Outcomes: Psychosocial outcomes: cognitive, social, emotional, or behavioral functioning.

Results: Social Outcomes. 9/10 studies examining social outcomes found that participants that had the animal interventions had increased social interaction/socialization and an improvement in communication (both verbal and non-verbal). Cognitive Outcomes: 7/10 studies examining cognitive outcomes found that participants that had the animal intervention showed improvement in cognitive functions, concentration, or attention. Emotional Outcomes: 2/10 studies found participants that had the animal intervention had a positive impact on their emotional well-being. 1 study reported significant improvements compared to baseline in mood and ability to relax. Behavior Outcomes: 4/10 studies found participants that had the animal intervention showed improvement in behavior.

Conclusion: Current evidence shows that AAT may be a potentially useful supportive intervention in improving quality of life in persons with ID, but good quality research is lacking. Therefore, it merits further exploration within this population. Future research should strive to address the methodological limitations of existing work if AAT is to be considered a clinical and cost-effective adjunct in services for people with ID.

Bias and quality assessment tools: EPHPP, Quality Assessment Tool for Quantitative Studies, 2009.

Funding and conflict of interests: No funding reported. Authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Serwacki (2012). International Journal of Yoga Therapy.

Title: Yoga in the Schools: A Systematic Review of the Literature.

Objectives: To examine the evidence for delivering yoga-based interventions in schools.

Included studies: 12 studies: 4 quasi-experimental controlled studies, 1 single-cohort, 2 randomized matched pairs, 3 experimental studies, 1 within-groups counterbalanced design, and 1 within-groups multiple-baseline design with comparison group.

Population: School children. Sample: 348 participants (90 ID). Age range: 5-15 y. Other diagnoses included ASD, ID, specific learning disability or emotional disturbance, and severe educational problems.

Setting: Public or alternative schools in the United States, India, England, and Germany.

Intervention: Yoga programs: poses, breathing, meditation, relaxation, reflection, role-playing, guided imagery, discussion, music, visual aids, internal cleansing, mindfulness practice, massage, media literacy, interactive discourse, and imagination journeys.

Comparison: Control groups (with gross motor activities, such as games and physical activity) and multiple baselines.

Outcomes: academic, cognitive (attention, concentration, planning time, execution time, intelligence), and psychosocial (self-worth, physical wellbeing, body dissatisfaction, teacher and parent ratings, pulse rate, muscle tone, breathing, stress, social and self-confidence, communication and interaction abilities, self-control, emotional symptoms, conduct problems, hyperactivity, and peer relationship problems, eating-disordered behavior) benefits of using yoga in schools to promote health and wellness.

Results: Although effects of participating in school-based yoga programs appeared to be beneficial for the most part, methodological limitations, including lack of randomization, small samples, limited detail regarding the intervention, and statistical ambiguities curtailed the ability to provide definitive conclusions or recommendations.

Conclusion: Findings speak to the need for greater methodological rigor and an increased understanding of the mechanisms of success for school-based yoga interventions.

Bias and quality assessment tools: Sackett's levels of evidence strategy.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Tint (2017), Journal of Intellectual Disability Research.

Title: A systematic literature review of the physical and psychosocial correlates of Special Olympics participation among individuals with intellectual disability.

Objectives: To synthesize key findings regarding the physical, psychological/emotional, social and/or intellectual/cognitive correlates of SO participation for individuals with ID and to highlight limitations in the extant research as well as directions for future research.

Included studies: 46 studies (5 RCT, 7 NRCT, 14 cross-sectional, 11 case series, 5 cohort, 11 qualitative, with overlap)

Population: Individuals with intellectual disability. Sample: 3931 participants with mild to severe ID. Age range: 3-65 y.

Setting: Sport settings.

Intervention: Special Olympics programs with an existing research base: Traditional SO sports (15), Unified Sports (11), Healthy Athletes and others (4), or not specified (16).

Comparison: For the controlled studies (31), individuals with ID in different SO programs (1), non-SO interventions (11), and/or no intervention (21); individuals without ID in SO programs and/or no intervention (6). 12 studies used more than one type of control.

Outcomes: Psychological/emotional (e.g., self-worth) (24), physical (e.g. physical fitness, sport/motor skill development) (23), social (e.g. perceived social acceptance, social skills, and community involvement) (14), and cognitive/intellectual development (perceived competence in academic activities such as reading and spelling) (1).

Results: Psychological/emotional measures varied greatly by study and sport. 9 studies credited SO with positive changes in participants' self-perception and general self-worth. The number of total competitions and SO involvement seem to predict positive results in this area. There seems to be an increase in adaptive behavior and a decrease in maladaptive behavior, but results were mixed. Participation in SO has been associated with increased positive emotions (e.g., fun and enjoyment). 13 studies highlighted improved athletic and motor skills as a positive outcome of participation for SO athletes with ID across a variety of SO programs, while other studies found no additional benefit for the SO programming despite positive results in athletic skill across physically active groups. Mixed results were reported for physical fitness measures. 17 studies show SO participation led to increased social skills and friendships. SO participation was also associated with greater community participation and social inclusion. There is, however, conflicting evidence in this area. In the single study evaluating cognitive development, positive results were found but were more likely due to changes in physical competency and social acceptance sub-scales. Overall, most studies (14) were of low quality with a high risk of bias.

Conclusion: While studies generally indicated positive associations with SO participation, there is insufficient evidence in the extant literature to make firm conclusions.

Bias and quality assessment tools: Scottish Intercollegiate Guidelines Network methodology checklists.

Funding and conflict of interests: The authors received funding from the Social Sciences and Humanities Research Council and Department of Canadian Heritage (Sport Canada), as well as from the Chair in Autism Spectrum Disorders Treatment and Care Research.

AMSTAR 2: critically low confidence.

1.5. Technology

Systematic reviews

Boulton (2018), Journal of Intellectual Disabilities.

Title: Could participant-produced photography augment therapeutic interventions for people with intellectual disabilities? A systematic review of the available evidence.

Objectives: To determine the evidence base underpinning the use of participant-produced photography within therapeutic settings.

Included studies: 13 experimental studies, 2 including ID.

Population: Individuals who participated in therapeutic photography programs. Sample: 194 participants (28 ID). Age range 3-70 y. Age range of ID participants: ~16-60 y. Other participants were diagnosed with obesity, mental illnesses, chemical dependency, or cancer.

Setting: School (5), playschool (1), special school (1), summer school (1) community residential/day centers/rehab centers for people with ID (2), hospital (2), outpatient (1).

Intervention: Participant-produced photography.

Comparison: Pretest/posttest.

Outcomes: change at an emotional, educational or behavioral level, measured by photographs (3) and PowerPoint presentation (1), student self-reflection rubric (1), description of pre-school day (1), meal quality and dietary diversity (1), self-reported nutritional intake (1), food diary (1), weight, BMI and physical health (1) multidimensional self- esteem inventory (1), trauma symptom inventory (1), researcher designed abuse questionnaire (1), Ryff Scale of Psychological Well- being (1), The Empowerment Scale, Community Integration Measure (1), psychological distress and health related quality of life (1), measure of self-concept (1).

Results: The intervention was considered a tool for construction of meaning, information gleaning, transcendence of language barriers, improving self-esteem and sense of empowerment, enhancement of learning, sense of inquisitiveness and wonder, increasing of level of focused attention on the surrounding environment, improvement of communication and description of activities, enabling initial responses to be non-verbal, providing participants with a tangible object from which to speak, and promoting personal development and growth. Methodological limitations require caution in accepting these findings.

Conclusion: Encouragingly, results suggest that programs using inexpensive, simple photographic equipment, combined with targeted discussion of photographs can facilitate a shift from 'learner' to 'expert' status in the therapy room, facilitating a sense of value, empowerment, and improved self-esteem.

Bias and quality assessment tools: Critical Appraisal Skills Program Checklist (CASP, 2014).

Funding and conflict of interests: The authors declare no financial support or conflict of interest.

AMSTAR 2: critically low confidence.

den Brok (2015), Disability and Rehabilitation: Assistive Technology.

Title: Self-controlled technologies to support skill attainment in persons with an autism spectrum disorder and/or an intellectual disability: a systematic literature review.

Objectives: To analyze the literature regarding mobile devices (e.g. iPod, and iPad), robots, and virtual reality (VR) technologies in relation to learning and independence for people with ASD and/or ID.

Included studies: 28 studies. Single case design (27), between-subjects design study (1).

Population: Individuals with ID, ASD, or both. Sample: 197 participants (120 ID, 72 ASD, 5 both). Age range for ID participants: 5-70 y. Age range for ASD participants: 3-27 y. Age range for participants with both conditions: 1-14 y.

Setting: Not specified.

Intervention: Self-controlled technologies: learning by prompting (13), Learning through interaction with robots (6), learning by practicing in the present, in a real life or virtual daily living situation (8). The used technologies were videos on computers, videos on handheld devices, pictures on handheld devices, robots, and virtual reality.

Comparison: Usually no intervention, multiple-baseline, pre-post, or not specified.

Outcomes: Daily living skills, vocational skills, transitioning within tasks, transitioning between tasks, engagement, and safety, social convention skills, time perception, and imagination.

Results: There is evidence that technology supports learning of activities of daily living and cognitive concepts in children and adults with ASD and/or moderate to mild ID. Results showed that videos on computers or handheld devices (or pictures on handheld devices) were commonly used to prompt and were mainly used to teach activities of daily living such as daily living skills, vocational skills, and transitioning within and between tasks. Children may need more intrusive prompts for more difficult tasks. Learning through interaction with robots was commonly used to stimulate play and engagement. VR was mainly used to create an environment to practice in the present, most commonly to support learning cognitive concepts such as time perception, imagination, or the emotions of others.

Conclusion: This review has found promising advances in using technology to support attaining new daily living skills and cognitive concepts (e.g., time perception, imagination) in persons with ASD and/or moderate to mild ID. Only a few reports were found on using technology to stimulate the understanding of emotions. More research on using technology to teach skills with an affective component to persons with ASD and/or ID may support their participation in society and add to their overall well-being.

Bias and quality assessment tools: Not stated.

Funding and conflict of interests: The authors received funding from ZonMW-InSight, a Dutch association promoting application-oriented research to support the needs of people with a visual impairment.

AMSTAR 2: critically low confidence.

Kagohara (2013), Research in Developmental Disabilities.

Title: Using iPods® and iPads® in teaching programs for individuals with developmental disabilities: A systematic review.

Objectives: To inform evidence-based practice in the use of iPod/iPad-based instruction for individuals with developmental disabilities.

Included studies: 15 studies.

Population: Individuals with developmental disabilities. Sample: 47 participants. Age range: 4-27 y. Diagnosis of ASD and/or ID.

Setting: School settings.

Intervention: The use of iPods® and iPads® in teaching programs.

Comparison: Multiple probe; multiple baselines.

Outcomes: Academic, communication, leisure, employment skills, and transitioning skills.

Results: The results of these 15 studies were largely positive, suggesting that iPods, iPod Nano, iPod Touch, iPads, and iPhones are viable technological aids for individuals with developmental disabilities. The results of these 15 studies also suggest that individuals with developmental disabilities can be taught to use such devices for a variety of purposes: specifically for enhancement of academic, communication, leisure, employment skills, and transitioning skills.

Conclusion: The tentative conclusion is that careful implementation of applied behavior analysis-based instructional procedures can make iPod, the iPodTouch, iPhones, and iPads viable technological aids for individuals with developmental disabilities.

Bias and quality assessment tools: None.

Funding and conflict of interests: Support from New Zealand Government through the Marsden Fund Council, Victoria University of Wellington, The University of Canterbury, and The New Zealand Institute of Language, Brain & Behavior.

AMSTAR 2: critically low confidence.

Kim (2017). Journal of Computer Assisted Learning.

Title: The quality of evidence in tablet-assisted interventions for students with disabilities.

Objectives: The purpose of this review is to weigh the evidence of the effectiveness of tablet-assisted instructions (TAIs) at improving academic outcomes of students with disabilities.

Included studies: 17. Single-case (4), within-subjects design with repeated measures (2), multiple-baseline (4), multiple-probe (3), alternating treatment (1), quasi-experimental (1).

Population: Students with disabilities. Sample: 112 participants (~12 ID). Age range: young children, students at the K-12 level and young adults were included. Other disabilities included ASD, ID, DD, learning difficulties, and hearing impairments.

Setting: Schools in EUA (15), Taiwan (1), South Korea (1).

Intervention: Tablet-assisted instruction (TAI).

Comparison: Multiple-baseline, repeated measures, multiple-probe.

Outcomes: Academic outcomes, including reading, spelling, writing, listening, math, science, and general performance.

Results: Evidence suggests that tablet-based interventions are popular in special education classrooms and may have the potential to enhance learning for students with disabilities and learning difficulties.

Conclusion: Regardless of several studies that addressed the effects of TAI for students with disabilities, this review revealed that there remains more to be established for TAI to be considered promising and evidence based.

Bias and quality assessment tools: Operationalized quality indicators (QIs) developed by Jitendra, Burgess and Gajria (2011).

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Oudshoorn (2020), Journal of Applied Research in Intellectual Disabilities.

Title: eHealth in the support of people with mild intellectual disability in daily life: A systematic review.

Objectives: To provide an overview of the use of eHealth as it relates to supporting people with a mild intellectual disability in daily life.

Included studies: 46 studies. Group design (10), and single-case design (36).

Population: Individuals with mild ID. Sample: 346 participants (210 mild ID). Age range: 5-70 y.

Setting: Home, community, educational context, vocational context, daycare, or unclear. United States (39), Netherlands (2), Turkey (2), Australia (1), France (1) and Spain (1).

Intervention: Technology as temporary aid in facilitation of learning practical daily living skills (e.g., purchasing groceries), permanent aid to support independent task completion in home or vocational settings, and facilitator for remote professional support to carry out daily activities (e.g. remote coaching). Evidence-based instructional practices, such as a system of least prompting, most-to-least and least-to-most prompting, constant and progressive time delay prompting, and model-lead test format, were used to achieve independent use of eHealth devices. Devices included portable applications (such as smartphones) (13), iPad/iPod tablets (frequently combined with apps, specific software, videos, Bluetooth earpieces and e-books) (16), computers/laptops (in combination with specific software, showing step-by-step pictures or videos of target skills) (11), virtual reality (1), and augmented reality (1).

Comparison: Control, multiple baselines, multiple probing, alternating treatment.

Outcomes: Daily-living skills.

Results: Most of the 46 studies included were small-scale case studies and focused on using eHealth to acquire daily living skills and vocational skills. In addition, several studies focused on eHealth use for self-support in daily living, and three studies focused on remote professional support. Various eHealth applications can be successfully implemented following structured training using behavioral therapeutic principles for people with mild intellectual disability. eHealth can contribute to the expansion of opportunities to support people with mild intellectual disability in various domains of their daily lives and their participation in the community.

Conclusion: eHealth offers opportunities to support people with mild intellectual disability in various contexts of daily life. Scientific research on this topic is in its early stage, and further high-quality research is needed.

Bias and quality assessment tools: Evaluative Method for Determining Evidence-Based Practice (EMDEBP).

Funding and conflict of interests: Funding by ASVZ. Authors declare no conflict of interest.

Ramdoss (2011), Journal of Developmental and Physical Disabilities.

Title: Use of Computer-Based Interventions to Promote Daily Living Skills in Individuals with Intellectual Disabilities: A Systematic Review

Objectives: To evaluate and synthesize the evidence-base regarding computer-based interventions (CBI) for teaching daily living skills to individuals with intellectual disability.

Included studies: 11 studies.

Population: Individuals with ID. Sample: 42 participants. Age range: 7-58. 3 participants were also diagnosed with DS, and others, with ASD or PDD.

Setting: Baselines and outcome measurements were taken in outcome-related settings (grocery stores, bus stops, or restaurants), while the interventions were implemented in instructional settings (home or school)..

Intervention: Six studies used software programs that were specifically designed for the purposes of their intervention: Project Shop (3), I can! Daily Living and Community Skills (2), or ATM Sim (1) (these software were no longer commercially available at the time of the review); Five studies used commercially available multi-authoring tools such as Hyper Studio, Microsoft PowerPoint, and Windows Movie Maker.

Comparison: Baselines.

Outcomes: Some aspects of grocery purchasing (reading grocery aisle signs, grocery item selection, purchasing sequence, and determining how much money to hand the checkout person) (5); setting the table and simple meals (2); using a debit card (1); using an ATM machine (1); placing an order in fast-food restaurants (1); and using public bus transportation (1).

Results: Thirty-nine of the 42 participants (93%) acquired the targeted daily living skill via CBI. Out of the 11 studies, 8 reported positive outcomes for all participants, and three reported mixed outcomes. Six of the included studies assessed the maintenance of acquired skills via follow-up probes, which showed that target skills were maintained at similar levels to the final intervention phase.

Conclusion: CBI is a promising intervention strategy for teaching daily living skills to individuals with moderate ID.

Bias and quality assessment tools: None.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Roche (2015). AAC: Augmentative and Alternative Communication.

Title: Microswitch Technology for Enabling Self-Determined Responding in Children with Profound and Multiple Disabilities: A Systematic Review.

Objectives: To review studies reporting on the use of microswitch technology to enable self-determined responding in children with profound and multiple disabilities.

Included studies: 18 single-case experimental studies (9 multiple probes, 6 ABAB reversal, 3 alternating treatments, 2 multiple baseline)

Population: Individuals with S/PID and other concomitant disabilities. Sample: 45 participants. Age range: 4-18 y. Concomitant disabilities included cerebropathy, epilepsy, visual impairments, and spastic tetraparesis.

Setting: Classrooms and others.

Intervention: Microswitches (microphone switches activated by vocalizations; vibration switches activated by tapping/hitting the table surface on which the switch had been placed; lever switches activated by chin movement; optic microswitches activated by forehead movements) to enable participants to control access to preferred stimulation (e.g., music, songs, vibration, and/or lights). The children were trained with response prompting, prompt fading, and contingent reinforcement.

Comparison: Baseline.

Outcomes: Making choices, recruiting attention, social interaction.

Results: The results of these studies were consistently positive and support the use of microswitch technology in educational programs for children with profound and multiple disabilities to impact their environment and interact with others.

Conclusion: There is sufficient empirical evidence to support and justify the use of microswitch technology in educational programs for these children.

Bias and quality assessment tools: Not stated.

Funding and conflict of interests: Authors received royalties for publication of books referenced in the paper.

AMSTAR 2: critically low confidence.

Tsikinas (2019), Journal of Computer Assisted Learning.

Title: Studying the effects of computer serious games on people with intellectual disabilities or autism spectrum disorder: A systematic literature review.

Objectives: To identify the state-of-the-art research on serious games (SGs) for people with ID or ASD.

Included studies: 58 (36 from journals and 22 from conferences) in total. For ID, 9 pretest-posttest, 6 in-game evaluation, and 2 observational. For ASD, 21 pretest-posttest, 11 in-game evaluation, 3 observations, and 2 interviews).

Population: Individuals with ID or ASD. Sample: not stated. Age range for ID participants: ~8-25. Age range for ASD participants: ~6-18. Age ranges are not comprehensive, few ages were cited.

Setting: Non-formal context (33) and formal learning process of special education institutions (17).

Intervention: Serious games (SG): computer games (46), console games (4) and mobile games (7).

For ID (17 studies), intellectual functioning games focused on attention and understanding, working memory, punctuation and comprehension, and cognitive skills. Conceptual games focused on language and literacy, money, time, and numbers. Practical games focused largely on daily living and work-related skills.

Comparison: Baseline, typically developing children, or not specified control participants.

Outcomes: For ID studies, adaptive behavior: conceptual skills (4) (language and literacy (1), money (1), time (1), numbers (1)) and practical skills (7) (daily living (2), work related (3), healthcare (1), travel/transportation (1)); Intellectual functioning/cognitive skills (6).

Results: 14 studies showed positive effects and 3 studies showed neutral results. Positive effects were improvements in social interaction, motor skills, attention and understanding, working memory, punctuation and comprehension, and cognitive skills. The SGs for practical skills are mainly focused on daily living and work-related skills.

Conclusion: The results indicated that SGs for people with ID and people with ASD could improve practical, conceptual, cognitive, and social skills. Thus, developing SGs for people with ID and people with ASD could be used to enhance the learning process.

Bias and quality assessment tools: SLR protocol.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

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2. Fine motor-adaptive

2.1. Physical therapy, physical education, or occupational therapy

Systematic reviews

Houwen (2014), Research in Developmental Disabilities Review.

Title: A systematic review of the effects of motor interventions to improve motor, cognitive, and/or social functioning in people with severe or profound intellectual disabilities.

Objectives: To systematically review the evidence related to motor interventions designed to improve motor, cognitive, and/or social outcomes in people with severe or profound ID.

Included studies: 45. Single-subject designs (40): variation of a multiple baseline or probe design (16), reversal (15), alternating treatment (2), ABA design (2), AB design (3), case reports (4). Group design (5): quasi-experimental pretest posttest (2); pretest posttest design without control group (3).

Population: Individuals with Severe or profound ID. Sample: 204 participants. Age range: 2-60 y.

Setting: Multiple settings. Not stated.

Intervention: Motor interventions: behavioral techniques, behavioral techniques with assistive technology, motor learning strategies, sensory stimulation techniques, and specific interventions.

Comparison: multiple baselines, reversal, alternating treatment; control in some group studies.

Outcomes: Functional hand use, ambulation, stereotypical movements, hand skill, balance, gait, hyperactivity, communication, social interaction, independent walking, motor scales, dancing, ball-hitting.

Results: Thirty-eight articles reported improvement in basic motor skills and eight articles reported improvement in recreational or more specialist motor skills. None of the articles reported negative effects due to motor interventions. This review suggests that sessions of 10–15 min, 3-5 times a week may result in significant functional improvements in motor functioning. The effects of motor activity on cognitive and social functioning remain unknown.

Conclusion: Further research is required to determine which motor interventions are the most effective in improving motor outcomes and/or cognitive and social outcomes, and on the longer-term effects of these interventions.

Bias and quality assessment tools: evaluated based on the level of evidence (according to Sackett (1989)), with minor modifications supported by research from the American Academy for Cerebral Palsy and Developmental Medicine) and the internal and external validity. Rating system based on Siebes, Wijnroks, and Vermeer (2002).

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

2.2. Technology

Systematic reviews

Hickman (2017), Developmental Medicine and Child Neurology.

Title: Use of active video gaming in children with neuromotor dysfunction: a systematic review.

Objectives: To examine current evidence on use of active video gaming (AVG) to improve motor function in children with movement disorders including cerebral palsy, developmental coordination disorder, and Down syndrome.

Included studies: 2 (1 RCT and 1 quasi-experimental study) with Down syndrome (DS) population. 20 in total.

Population: Children with neuromotor dysfunctions. Sample: 606 participants (185 DS). Age range: 3-20 y. Age range for ID children (7-13 y). Other conditions included cerebral palsy, developmental coordination disorder (DCD), developmental delay, progressive spinocerebellar ataxia, and acquired brain injury.

Setting: Clinic, supervised.

Intervention: Active video gaming (Nintendo Wii Sports; Wii Fit with Wii balance board; games: Football Heading, Tightrope Walk; Penguin Slide Game).

Comparison: Standard care.

Outcomes: Fine motor precision and integration, manual dexterity, bilateral coordination, balance, running speed and agility, upper limb coordination, and strength (Bruininks-Oseretsky Test); Visual-Motor Integration (VMI); Test of Sensory Integration Function (TSIF).

Results: The children's scores for the Bruininks-Oseretsky Test improved after AVG training. The remaining two outcome measures evaluated visual perception, motor coordination, postural movement control, and various sensory and behavioral abilities. There was improvement in the scores of these two tests as well.

Conclusion: There was clear support for use of AVG to improve general motor function in children with neuromotor conditions. However, the literature did not support use of AVG as a standalone intervention capable of creating permanent neuromotor improvements.

Bias and quality assessment tools: Centre for Evidence-based Medicine and GRADE.

Funding and conflict of interest: The authors received the 2015 UNLVPT Student Opportunity Research Grant but reported no conflict of interest.

AMSTAR 2: critically low confidence.

Kokol (2020), Current Pediatric Reviews.

Title: Serious Game-based Intervention for Children with Developmental Disabilities.

Objectives: To review clinical studies which investigated the application of serious games in children with developmental disabilities.

Included studies: 145 clinical studies. Of those, 4 RCTs concerning ID.

Population: Individuals with developmental disabilities from between 2 and 24 years of age. Sample: not reported. Age range: not reported. Other conditions included ASD, ADHD, developmental coordination disorder (DCD), disabilities affecting intellectual abilities and CP.

Setting: Home or clinical setting.

Intervention: Serious games: Wii-Fit games, other Wii games, and JECRIPE (the game addresses aspects of interaction, communication and stimulates cognitive abilities).

Comparison: Traditional physical therapy for the Wii-Fit games group and not stated for other studies.

Outcomes: Motor skills.

Results: For DS, a program of three Wii-Fit games in addition to the traditional physical therapy program yielded a significant improvement of balance in the study group. In another study, playing Wii resulted in improvements in the child's postural stability, limits of stability, and balance, upper-limb coordination, manual dexterity, and running speed and agility standard scores.

Conclusion: Studies on the use of serious games in children with neurodevelopmental disorders appeared to be used principally in children with ASD, ADHD, developmental coordination disorder, and disabilities affecting intellectual abilities. For DS individuals, serious games seem to improve motor skills.

Bias and quality assessment tools: None.

Funding and conflict of interests: No funding or conflict of interests declared.

AMSTAR 2: critically low confidence.

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3. Language

3.1. Caregiving

Meta-analysis

Liao (2020). Review Journal of Autism and Developmental Disorders.

Title: Caregiver Involvement in Communication Skills for Individuals with ASD and IDD: a Meta-analytic Review of Single-Case Research on the English, Chinese, and Japanese Literature.

Objectives: To determine the effects of caregiver involvement for promoting communication skills of children with ASD and IDD.

Included studies: 43 single case studies. 149 caregiver-child dyads/triads were included

Population: Individuals with ASD and IDD and their caregiver. Sample: 149 caregiver-child dyads/triads. Age range: 1-7+ (not otherwise specified).

Setting: Home (87), clinic or center settings (36), multiple settings (22).

Intervention: Caregiver involvement for promoting communication skills of children, defined as: "an individual-centered connection between caregivers at home and professionals in school settings (e.g., teachers, therapists, and service providers) who share responsibility for the development of individuals with ASD and IDD".

Comparison: Pairwise comparisons.

Outcomes: Communication skills (not specified).

Results: Caregiver involvement has shown improvement in communication skills of individuals with ASD and ID.

Conclusion: Caregiver involvement promotes reduction in caregivers' stress and anxiety and improvement in caregiver-child interaction, parental quality of life, the understanding of ASD and ID, and in communication skills.

Bias and quality assessment tools: A combination of quality criteria for single-case experimental study proposed by previous studies (Council for Exceptional Children 2014; Ganz and Ayres 2018; Horner et al. 2005, Kratochwill et al. 2013, Maggin et al. 2013; Reichow et al. 2008; U.S. Department of Education, Institute of Education Sciences, What Works Clearinghouse 2016).

Funding and conflict of interests: Funding not stated. Authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Systematic reviews

O'Toole (2018), Cochrane Database of Systematic Reviews.

Title: Parent-mediated interventions for promoting communication and language development in young children with Down syndrome (Review).

Objectives: To assess the effects of parent-mediated interventions for improving communication and language development in young children with Down syndrome. Other outcomes are parental behavior and responsivity, parental stress and satisfaction, and children's non-verbal means of communicating, socialization, and behavior.

Included studies: 3. 2 RCTs and 1 quasi-RCTs.

Population: Children with Down Syndrome. Sample: 45 participants. Age range: 2-6 y.

Setting: Home, clinic, or both; interventions were delivered through group or one-to-one sessions.

Intervention: parent-mediated interventions (Hanen Parent Program; Responsive teaching (RT); Enhanced milieu teaching (EMT)).

Comparison: Treatment as usual, therapist-only control.

Outcomes: Expressive language (number of (different) target words); receptive language (total language; standard scores); changes in parental behavior/responsivity; socialization; language attrition; adherence to treatment; parental stress.

Results: Two studies found no differences in children's language ability. One study reported gains on general measures of overall language ability. Most strategies were retained by the intervention group 12 months later. One study reported increases in socialization skills.

Conclusion: Very low certainty for all outcomes, with very little confidence in the effect estimate. The true effect is likely to be substantially different from the estimate of effect.

Bias and quality assessment tools: Cochrane Collaboration's Risk of Bias Tool.

Funding and conflict of interests: Authors received funding from universities, charities, and the Cochrane Fellowship. They are or have been also involved in other projects, such as books, intervention centers, universities, courses, charities, and other research, regarding language and disabilities.

AMSTAR 2: high confidence.

3.2. Educational

Meta-analyses

Neil (2016), Developmental Neurorehabilitation.

Title: Communication intervention for individuals with Down syndrome: Systematic review and meta-analysis

Objectives: To identify effective intervention strategies for communication in individuals with Down syndrome.

Included studies: 37. Multiple baseline (22); RCT (6); alternating treatments (4); reversal (4); both multiple baseline and alternating treatment (1);

Population: Individuals with Down Syndrome. Sample: 225 participants. Age range: 0-54 y.

Setting: Mostly schools.

Intervention: Most studies used prompting and reinforcement strategy (73%). Other strategies were morphosyntactic grammar intervention; manualized reading and language intervention; speech recasting; Milieu teaching; mirroring and responding.

Comparison: multiple-baselines, alternating treatments, reversal, treatment as usual.

Outcomes: Expressive behaviors: spoken responses (or adapted for use with alternative communication systems), and receptive behaviors: nonvocal responses to a teacher's spoken instructions.

Results: The results suggest that intervention for expressive communication for individuals with Down syndrome is effective. Effect sizes for group studies ranged from trivial to moderate.

Conclusion: The results suggest that behavior analytic strategies are a promising approach, and future research should focus on replicating the effects of these interventions with greater methodological rigor.

Bias and quality assessment tools: No specific tool reported. Authors used quality indicators (maintenance, generalization, integrity, and reliability).

Funding and conflict of interests: The authors have not reported funding but they reported no conflict of interest.

AMSTAR 2: critically low confidence.

Reichow (2019), Cochrane Database of Systematic Reviews.

Title: Beginning reading interventions for children and adolescents with intellectual disability.

Objectives: To assess the effectiveness of interventions for teaching beginning reading skills to children and adolescents with intellectual disabilities.

Included studies: 7 RCTs.

Population: Children and adolescents with ID. Sample: 352 participants. Age range: 5-18 y.

Setting: School.

Intervention: Beginning reading instruction (teaching students to identify printed letters and words).

Comparison: Treatment as usual.

Outcomes: Primary: phonological awareness, sound-symbol correspondence, word reading, decoding, and adverse events. Secondary: supplemental measures of literacy (vocabulary, comprehension, spelling, writing, oral reading fluency, or a combination of these); supplemental measures of language skills, treatment acceptability, attitudes towards reading, self-efficacy related to reading, and behavioral outcomes.

Results: Medium effect sizes in favor of the beginning reading interventions for the primary outcomes of phonological awareness, word reading, both with moderate-quality evidence and decoding with low-quality evidence. Moderate effect for the secondary outcomes of oral reading fluency, with low-quality evidence, and language skills, with moderate-quality evidence. No adverse events.

Conclusion: Beginning reading interventions that include elements of phonological awareness, letter-sound instruction, and decoding, are associated with small-to-moderate improvements in phonological awareness, word reading, decoding, expressive and receptive language, and oral reading fluency.

Bias and quality assessment tools: Cochrane's risk of bias tool.

Funding and conflict of interests: All authors received funding from different universities in the USA, and three of them also received funding from the Department of Education of USA, honoraria from not-for-profit organizations, for publications, lectures, and advisor and editorial duties on scientific journals. However, all authors declare no conflict of interest.

AMSTAR 2: high confidence.

Systematic Reviews

Colenbrander (2019). Language, Speech, and Hearing Services in Schools.

Title: To See or Not to See: How Does Seeing Spellings Support Vocabulary Learning?

Objectives: To determine when, why and how the presence of a word's written form during instruction aids vocabulary learning (a process known as orthographic facilitation).

Included studies: 23 experimental studies from 19 papers. Only 1 study including ID children (Down syndrome) Study designs not stated.

Population: Individuals learning vocabulary. Sample: 1050 individuals (17 DS). Age range: 5-64 y. Age range for ID participants: 7-16 y.

Setting: Not stated.

Intervention: Orthographic facilitation, with the presence of the written form during word learning.

Comparison: Word learning with no orthography.

Outcomes: Learning of orthographic, phonological, and semantic information.

Results: The findings suggest that the presence of the written form during word learning is likely to be beneficial for beginning readers and English language learners, as well as children with DLD, reading difficulties, ASD and Down Syndrome. There is evidence that orthographic facilitation can support vocabulary acquisition in children with Down Syndrome.

Conclusion: A small number of studies have shown that the presence of a word's written form benefits vocabulary learning in children with Developmental Language Disorder, autism, Down Syndrome and reading difficulties. However, further research into the effects of orthographic facilitation in special populations is needed.

Bias and quality assessment tools: None.

Funding and conflict of interests: The authors declare no competing interests.

AMSTAR 2: critically low confidence.

Joseph (2009), Research in Developmental Disabilities.

Title: Teaching students with intellectual or developmental disabilities to write: A review of the literature.

Objectives: To identify effective methods for teaching writing to students with intellectual disabilities.

Included studies: 9. Single-subject experimental designs (6) and pre-experimental designs (3).

Population: Children with intellectual or developmental disabilities. Sample: 31 participants with ID. Age range: 6-18 y. Some participants were also diagnosed with ASD.

Setting: school (6); university computer room (2); child development institute (1).

Intervention: Strategy instruction (4): self-regulated strategy development; modified cognitive strategy instruction in writing (1); Computer-based instruction (3); one-on-one instructional sessions (1); Four Blocks literacy approach (1).

Comparison: multiple-probe (3), multiple-baseline (2), reversal design (1), and pre-test/post-test (3).

Outcomes: writing quality/accuracy (8), productivity (5), pre-writing (planning) (2), and emergent/alternative writing skills (computer-assisted sentence construction) (2).

Results: Among the types of instruction reviewed, strategy instruction yielded the strongest writing performance outcomes for students with intellectual or developmental disabilities, and SRSD was the most frequently used.

Conclusion: Students with intellectual disabilities can benefit from writing instruction and can be taught learning strategies to help them improve the quantity and quality of their written expression.

Bias and quality assessment tools: None.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Machalicek (2010), Journal of Developmental and Physical Disabilities.

Title: Literacy Interventions for Students with Physical and Developmental Disabilities Who Use Aided AAC Devices: A Systematic Review

Objective: To guide and inform practitioners in evidence-based literacy instruction for students with physical and developmental disabilities who use aided AAC. An additional purpose is to identify gaps in existing literature to stimulate future research efforts aimed at developing new and more effective intervention strategies.

Included studies: 18 single-subject studies. 11 were non-experimental (5 AB, 2 non-concurrent multiple-baseline, 4 ABCD or multiple probe ABC) and 7 were experimental (4 multiple-baseline, 3 multiple probes or multielement)
Population: Students with physical and developmental disabilities. Sample: 41 individuals (9 ID, 2 DS, 10 RS). Age range 2-14 y. Age range for ID participants: 6-14 y. Age range for DS participants: 9-14 y. Age range for RS participants: 3-7 y.
Setting: Family homes (13 participants), segregated special education classrooms (5 participants), separate rooms outside of classrooms (8 participants), an inclusive classroom (1 participant), unspecified school setting (2 participants), both home and school (1 participant), outpatient language and speech clinic (1 participant), not described (8 participants). Not reported by the review (2 participants).
Intervention: direct instruction, scaffolding of communicative attempts (9), least-to-most prompting with time delay, writing workshop, and story mapping
Comparison: Baselines.
Outcomes: Frequency, variety, or complexity of AAC messages during literacy activities (9), letter-sound correspondence (4), participation in literacy activities (1), letter identification and spelling (1), decoding (1), spelling and print awareness (1) phonemic awareness, phonics, vocabulary, text comprehension, and
Results: Improvements were reported for 37% of participants, and positive participant outcomes appear to be related to the use of evidence-based systematic instructional procedures such as direct instruction, least to most prompting, and scaffolding of child communication. However, the certainty of evidence for an intervention effect was rated as inconclusive for 12 of the 18 reviewed studies, and therefore, positive results must be interpreted with caution.
Conclusion: Systematic instruction including scaffolding, direct instruction, and least-to-most prompting with time delay may be the most effective strategies to teach literacy skills to this population.
Bias and quality assessment tools: Millar et al. (2006)'s measures of certainty of evidence.
Funding and conflict of interests: Not reported.
AMSTAR 2: critically low confidence.

3.3. Technology

Meta-analyses

Ganz (2017), Augmentative and Alternative Communication.
Title: High-technology augmentative and alternative communication for individuals with intellectual and developmental disabilities and complex communication needs: a meta-analysis.
Objectives: To report the social-communication outcome results of a meta-analysis of single-case experimental research on the use of high-tech AAC, including mobile devices, by individuals with intellectual and developmental disabilities, including autism spectrum disorder.
Included studies: 24 single-case design studies.

Population: Individuals with intellectual and developmental disabilities and complex communication needs. Sample: 56 participants. Age range: pre-school to secondary school participants.

Setting: Natural and didactic settings.

Intervention: High-technology augmentative and alternative communication (e.g., voice-output communication aids, speech-generating devices, mobile devices with AAC apps).

Comparison: Contrast between phases of the studies.

Outcomes: Communication outcomes.

Results: All communicative functions evaluated produced significant effects, and communication of wants and needs was found to be significantly more improved compared to social closeness, though no other comparisons had significant differences. Unfortunately, most of the authors evaluated expression of wants and needs and few evaluated the impact of more social and more complex communicative behaviors, thus, these results must be interpreted with caution and indicate a gap in the literature.

Conclusion: Implementation of high-tech AAC was effective and likely to be effective under most conditions and for most participants with intellectual and developmental disabilities to improve communication skills.

Bias and quality assessment tools: What Works Clearinghouse.

Funding and conflict of interests: The research was supported in part by the Preparation of Leaders in Autism Across the Lifespan grant awarded by the U.S. Department of Education, Office of Special Education Programs. The authors declare no conflict of interests.

AMSTAR 2: critically low confidence.

Kent-Walsh (2015), Augmentative and Alternative Communication.

Title: Effects of Communication Partner Instruction on the Communication of Individuals using AAC: A Meta-Analysis.

Objectives: To conduct a systematic review and meta-analysis of the augmentative and alternative communication (AAC) partner instruction intervention literature to determine the overall effects of partner interventions on the communication of individuals using AAC, and any possible moderating variables relating to participant, intervention, or outcome characteristics.

Included studies: 17 single-case design studies.

Population: Individuals with complex communication needs. Sample: 53 participants (17 ID or DD). Age range: 2-26 y. Other conditions included ASD, CP, and others.

Setting: Not stated.

Intervention: Partner instruction on the communication of individuals using AAC, with strategy instruction or individual skill training.

Comparison: Multiple baseline or multiple probes.

Outcomes: Pragmatic: social interaction, proportion of communicative turns and initiations, frequency of communication acts; Morpho-syntactic: tracking of multi-symbol messages; Pragmatic and semantic: frequency of both multimodal communicative turns and different semantic concepts.

Results: A body of evidence consistently indicates that communication partner instruction has positive effects on communication performance of individuals using AAC.

Conclusion: Including communication partner instruction within AAC intervention plans will likely assist in yielding improvements in the communication skills of individuals with complex communication needs.

Bias and quality assessment tools: Certainty of evidence ratings based on design/internal validity, reliability of the dependent variable, and procedural integrity (Schlosser & Wendt, 2008).

Funding and conflict of interests: The second and third authors report no conflicts of interest. Some of the first and fourth authors' research is reviewed in the present article. However, procedural precautions were taken to minimize conflict of interest.

AMSTAR 2: critically low confidence.

Millar (2006), Journal of Speech, Language and Hearing Research.

Title: The impact of augmentative and alternative communication intervention on the speech production of individuals with developmental disabilities: a research review.

Objectives: To determine the effect of augmentative and alternative communication (AAC) on the speech production of individuals with developmental disabilities.

Included studies: 23. Descriptive case studies (8); single-participant (14); pretest–posttest design (1). After best-evidence analysis, 6 studies remained.

Population: Individuals with developmental disabilities. Sample: 67 participants (40% ID, 31% ASD) before best-evidence analysis and 27 participants (44% ID, 15% ASD). Age range: 2-60 y. Age range for ID participants: 5-60 y. Age range for ASD participants: 3-12 y.

Setting: Clinic.

Intervention: unaided AAC interventions using manual signs (5); aided AAC systems without speech output (1).

Comparison: Alternating treatments; multiple baselines; pretest-posttest; withdrawal design.

Outcomes: Speech production during or after the intervention.

Results: In the 6 best-evidence studies, 94% of the participants showed improvements in speech production which held true in different instructional approaches. There was an increase of 1-52 spoken words (mean = 13) and 4-7 two-word phrases (mean = 6). In 21% of the cases, the speech gains were not immediate. None of the studies showed conclusive evidence.

Conclusion: The results of the best evidence analysis should assuage the fears of parents and professionals about the potential negative impact of AAC intervention on speech production. Clinicians and parents should not hesitate to introduce AAC interventions to individuals with developmental disabilities whose speech is inadequate to meet their communication needs.

Bias and quality assessment tools: Certainty of evidence according to Simeonsson & Bailey, 1991 and N. L. Smith, 1981

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

Systematic reviews

Barbosa (2018), BMC Pediatrics.

Title: Augmentative and alternative communication in children with Down's syndrome: a systematic review.

Objectives: To analyze research findings regarding the different instruments of 'augmentative and alternative communication' used in children with Down's syndrome.

Included studies: 13 studies. 6 experimental studies, 3 longitudinal, 1 cross-sectional, 1 observational, 2 case reports.

Population: Children with Down Syndrome. Sample: 120 participants. Age range: 1-29 y.

Setting: Home, school, therapy, or not specified.

Intervention: Twelve instruments that provided significant aid to the process of communication and socialization of children with DS were identified: Speech-generating devices (SGDs), Picture Exchange Communication System (PECS), Sign language system (MAKATON), Picture communication symbols (PCS), Computer-generated pictographs (COMPIC), Web-based survey (joystick), Modified ride-on car, Picture-based strategy, Core vocabulary, Input techniques, Language signals system, and Digital interactive board. The instruments most used by the studies were speech-generating devices (SGDs) and the Picture Exchange Communication System (PECS).

Comparison: Intragroup, between-group, or compared to control groups.

Outcomes: Communication and associated outcomes: speech and core vocabulary improvement, cognition, learning processes, socializing, quality of life, autonomy, and individual self-esteem.

Results: Improvements were noted for every modality.

Conclusion: Beyond aid in communication, these instruments appear to increase the interaction between individuals, contributing to their quality of life and self-esteem.

Bias and quality assessment tools: PEDro Scale.

Funding and conflict of interests: The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Chung (2012), Research and Practice for Persons with Severe Disabilities.

Title: A Systematic Review of Interventions to Increase Peer Interactions for Students with Complex Communication Challenges.

Objectives: To identify and examine intervention approaches aimed at improving peer interaction outcomes for school-aged children with complex communication challenges who regularly used or might benefit from augmentative and alternative communication (AAC).

Included studies: 31 single-case studies (22 multiple-baseline, 7 reversals, and 2 alternating treatment). 29 of those were experimental (at least 3 demonstrations of intervention effects).

Population: Children with complex communication challenges. Sample: 89 students (58 ID, 9 ASD, 11 both). Age range: 6-22 y.

Setting: Schools.

Intervention: Intervention packages incorporating the use of AAC (introduction of new AAC systems, modification to students' existing AAC systems, or discrete teaching of skills related to the use of the AAC systems) or intervention packages involving students who were regular users of AAC (introducing new supports to students already using AAC without providing new AAC-related skill training or making changes in their existing AAC system). The approaches included peer training, adult facilitation, and environmental arrangements. Communication modes included speech, gestures, symbols, speech-generating device, vocalizations, facial expressions, pictures, signs, movements, communication book, written words, eye gaze, alphabet board, electronic device, switch, voice scanning system, switch-activated tape recorder, reaching, no formal communication, bliss books, affect, DynaWrite, head movements, Step-by-Step, computer, and visual tracking.

Comparison: Repeated measures during baseline.

Outcomes: Social interaction with TD peers, use of AAC during interactions, number of peers participating in each social contact, new peers met, types of interactive behavior, perceived quality of interactions, and conversation topics.

Results: Peer training is a component that should not be overlooked when designing interventions targeting peer interaction outcomes for students with significant disabilities. In 12 studies, overall positive results were reported from interviews or surveys with students, their peers without disabilities, and/or teachers. In 6 studies, teachers who were involved in interventions or delivered instructions all considered the interventions as practical and/or cost-effective.

Conclusion: Although the configurations of these intervention packages varied, most were found to be effective at increasing measures of peer interaction.

Bias and quality assessment tools: Adaptation of Horner et al. (2005).

Funding and conflict of interests: Not reported.

AMSTAR 2: critically low confidence.

Mandak (2018). AAC: Augmentative and Alternative Communication.

Title: The effects of literacy interventions on single word reading for individuals who use aided AAC: a systematic review.

Objectives: To investigate the effects of instruction on single word reading of individuals who use aided augmentative and alternative communication (AAC).

Included studies: 9 single-case studies.

Population: Individuals who use aided AAC. Sample: 24 participants (2 ID, 2 DS, 11 ASD, 1 RS). Age range: 6-22 y. Age range for ID participants: 11-14 y. Age range for DS participants: 9-10 y. Age range for ASD participants: 7-18 y. Age for the RS participant: 14.

Setting: One-on-one setting.

Intervention: Literacy interventions on single-word reading: phonological (4), sight-word (4), or both (1) approaches. Instructional strategies included pre-response: stimulus adaptations (changing the field size (1),

stimulus fading (1)), response prompts (physical guidance (1), modelling (7), and time delay (3)); and post-response: reinforcement (9), error correction (7), and error analysis (6).

Comparison: Multiple baseline/multiple probes or alternating treatments.

Outcomes: Percentage or number of words read correctly.

Results: Instruction had positive effects on reading at the single-word level for individuals across ages and diagnostic categories. The studies revealed that these effects were consistent across a range of participant, intervention, and outcome measure characteristics. Phonological approaches, sight-word approaches, and a combination of these two approaches yielded very large effects.

Conclusion: The findings must be viewed with caution due to limitations in the number of studies and participants and limitations in the reporting of detailed participant and intervention characteristics across the studies.

Bias and quality assessment tools: Quality markers offered by Horner et al. (2005) for single-case designs.

Funding and conflict of interests: Funding: Penn State AAC Leadership Project and Rehabilitation Engineering Research Center on Augmentative and Alternative Communication (The RERC on AAC). The authors report no conflicts of interest.

AMSTAR 2: critically low confidence.

Morin (2018), AAC: Augmentative and Alternative Communication.

Title: A systematic quality review of high-tech AAC interventions as an evidence-based practice.

Objectives: To evaluate the quality of single-case experimental design research on the use of high-tech AAC to teach social-communication skills to individuals with autism spectrum disorder or intellectual disabilities who have complex communication needs, and to determine if this intervention approach meets the criteria for evidence-based practices as outlined by the What Works Clearinghouse.

Included studies: 23 studies. Multiple probe (MPD), multiple baseline (MBD), and alternating treatment designs (ATD).

Population: Individuals with complex communication needs. Sample: 30 participants (2 ID, 28 ASD). Age range: 3-15 y. Age range for ID participants: 3-15 y. Age range for ASD participants: 3-12 y.

Setting: Not stated.

Intervention: AAC: PE = picture exchange; PECS = Picture Exchange Communication System; SGD = speech-generating device; manual signs. Instructional strategies included PECS training protocol, modified PECS protocol, discrete trial format, least to most prompting, time delay, functional communication training, error correction, full-physical prompt.

Comparison: Multiple baselines, multiple probe and alternating treatments.

Outcomes: Communication: requesting, positive facial expression; receptive identification, vocalization, response to intraverbal statements, labeling, discrimination; AAC preference; challenging behavior.

Results: High-tech AAC to teach social-communication skills to individuals with autism spectrum disorder or intellectual disabilities and complex communication needs can be considered an evidence-based practice,

although the review of comparison (i.e., alternating treatment) design studies did not indicate that high-tech AAC is significantly better than low-tech AAC.

Conclusion: The current study found evidence supporting the efficacy of high-tech AAC interventions, but because the literature base has primarily evaluated use of high-tech AAC with younger individuals, with more introductory communicative functions, and in more highly controlled research contexts, it cannot be stated with confidence that these results can generalize beyond these contexts.

Bias and quality assessment tools: WWC Procedures and Standards Handbook.

Funding and conflict of interests: The research was supported by the Institute of Education Sciences, US Department of Education and by the Office of Special Education Programs, U.S. Department of Education. No conflict of interest was declared.

AMSTAR 2: critically low confidence.

Roche (2014). AAC: Augmentative and Alternative Communication.

Title: Tangible Symbols as an AAC Option for Individuals with Developmental Disabilities: A Systematic Review of Intervention Studies

Objectives: To evaluate the effectiveness of tangible symbols (real objects representing ideas or situations) as a form of augmentative and alternative communication (AAC) for individuals with developmental disabilities.

Included studies: 9 studies, including 3 pre-experimental (2 pre-post and 1 B-only), 2 quasi-experimental (1 single-case ABCDE and 1 controlled trial), and 4 experimental (3 multiple baseline, 1 multiple baseline with alternating treatment)

Population: Students with developmental disabilities. Sample: 129 participants. Age range: 3-20 y. Almost all participants had ID, and other conditions included hearing and/or vision impairment, ASD, DD, among others.

Setting: Pre-, primary, or secondary schools.

Intervention: Tangible symbols were defined as real whole objects, miniature objects, or parts of the actual referent (e.g., a real spoon to represent lunchroom, a small plate to represent food). The instructional procedures used were response prompting, time delay, differential reinforcement, a modified picture exchange communication system (PECS) protocol, or a manualized intervention protocol.

Comparison: Baselines, alternating treatments, or control groups.

Outcomes: Requesting, choice making, naming, transitioning, and directing the actions of others.

Results: 54% of the participants (70) showed positive results, which were described as learning to use at least one tangible symbol for communicative purposes. Only 10 out of all participants were enrolled in experimental studies, which weakens the results.

Conclusion: Tangible symbols are

promising AAC option for individuals with developmental disabilities, perhaps especially for people who are also visually impaired.

Bias and quality assessment tools: None.

Funding and conflict of interests: The authors report no conflicts of interests. This review was supported by grants from the New Zealand Government through the Marsden Fund Council, by Victoria University of Wellington, The University of Canterbury, and The New Zealand Institute of Language, Brain & Behavior.

AMSTAR 2: critically low confidence.

Snyder (2019). American Journal on Intellectual and Developmental Disabilities.

Title: Computer Assisted Instruction to Teach Academic Content to Students with Intellectual Disability: A Review of the Literature.

Objectives: To explore the nature of academic content taught to students with ID using CAI, the CAI programs used to provide instruction, research methodology, and student learning outcomes associated with CAI.

Included studies: 22. Group studies (3 pretest/posttest), 1 of those a RCT; single-case-design (19): multiple-probe (7), multiple-baseline (6), alternating treatment (6).

Population: Students with intellectual disability. Sample: 188 participants. Age range: 5-22 y.

Setting: School.

Intervention: Computer Assisted Instruction (CAI) for academic content (literacy, math, written expression, social studies, or science), in most cases associated with systematic instruction (such as constant time delay, simultaneous prompting, system-of-least-prompt, explicit instruction, response prompts, stimulus prompts, error correction, reinforcement).

Comparison: Participants' baselines and/or non-CAI instructional procedures.

Outcomes: Literacy (9): sight word acquisition (5), early literacy skills and vocabulary development (4); Math (5): math computation problems (2), money skills (1), number identification (1), place value (1), word problems (1); Written expression (4): writing at sentence level or higher (3), writing single words (1), correct spelling of single words (1); Social studies (1): map reading skills (1); Science (1): ability to read and match science content words to science pictures and definitions (1).

Results: Across all studies CAI interventions were effective for teaching the targeted academic skills. Five studies including 12 students with ID (11 with moderate ID) found that CAI was effective for teaching students to identify sight words. All four studies comparing CAI to teacher-led instruction showed either no difference between the instructional approaches or mixed results across participants. Researchers reported generally positive maintenance results, indicating that skills taught via CAI maintained up to 2 months. 8 out of 12 studies which assessed generalization of skills yielded positive results. Teachers generally reported liking CAI interventions and thought CAI was effective for teaching academic skills.

Conclusion: Results from the 22 included studies suggest that CAI can be an effective means to teach academic content to students with ID.

Bias and quality assessment tools: Not stated.

Funding and conflict of interests: Not stated.

AMSTAR 2: critically low confidence.

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4. Gross motor

4.1. Physical therapy, physical education, or occupational therapy

Meta-analyses

Jeng (2016), Disability and Health Journal.

Title: Exercise training on skill-related physical fitness in adolescents with intellectual disability: A systematic review and meta-analysis.

Objectives: This study synthesized the results from the reviewed studies and determined whether exercise training improves SRF in adolescents with ID.

Included studies: 18 in total. 14 included in the meta-analysis. RCTs (9); cohort (6); follow-up (3).

Population: Adolescents with intellectual disability. Sample: 518 participants. Age range: 13-18 y.

Setting: Not stated.

Intervention: Exercise-training interventions: circuit training, muscle strengthening, unified sports, hippotherapy, Swiss ball exercise, traditional Greek dancing, treadmill and Wii Sports game training, sensorimotor training, device-assisted training, water aerobic exercise, low-intensity run/walk training, aerobic dance, walking exercise, Taekwondo, adapted dancing, road-running training, and horseback-riding machine training. The training period varied from 6 to 28 weeks (average 11.4 weeks), 1 to 3 sessions per week, and 30-90 min per session.

Comparison: Physical education, regular activity, no activity, or not mentioned.

Outcomes: Agility, power, coordination, reaction time, activity speed, balance.

Results: The results supported positive exercise training effects on agility, power, coordination, reaction time, and speed, but not balance in adolescents with ID.

Conclusion: Limited number of studies exhibited high quality evidence. Therefore, the results of this systematic review and meta-analyses should be interpreted with caution.

Bias and quality assessment tools: For risk of bias, authors evaluated each study for recruitment processes, sample size, methods, measurement, and study outcomes, such as effect estimates for intervention studies and confidence intervals (CIs) for intervention studies when possible. For quality, PEDro scores.

Funding and conflict of interests: Financial support: Chang Gung Memorial Hospital, Taiwan. The authors declare no conflict of interests.

AMSTAR 2: critically low confidence.

May (2019), Disability and Rehabilitation.

Title: Physical, cognitive, psychological and social effects of dance in children with disabilities: systematic review and meta-analysis.

Objectives: To collate and synthesize the findings of research conducted to date that has explored the psychological, cognitive, social and physical effects of dance programs for children with disabilities; to evaluate the quality of the studies conducted; and to provide recommendations for future research.

Included studies: 19 group studies (3 quasi-experiments, 4 uncontrolled trials, 9 controlled trials, 3 RCT).

Population: Children with disabilities. Sample: 521 participants (304 ID or DS, 80 ASD, 40 DD). Age range: 3-18 y. Age range for ID, DS, and DD participants: 6-18 y. Age range for ASD patients: ~3-18 y. Other conditions included hearing, speech, language, visual, and learning impairments.

Setting: School (17) or community (2) settings.

Intervention: Recreational dance classes or programs (creative, adapted, aerobic, community, data-based, or traditional Greek dances, dance exercises, or general dance education focused on basic movements and postures).

Comparison: Not specified.

Outcomes: physical outcomes: jumping (7), balance/coordination (7), running/sprinting/walking (5), sit-ups (4), perceptual motor skills (4), dance skills (4), flexibility (3), movement/rhythm skills (2) and BMI (1).

Cognitive outcomes: attention (2), organization skills (1), and serial learning (1). Psychological outcomes: creativity (3), self-concept (3), attitude toward dance (1), and body image (1). Social outcomes: social competence/peer interaction (3), peer play disconnection and disruption (1), social contact with peers and overcoming autistic symptoms of aloofness and rigidity (1).

Results: All papers that measured dance skills found significant improvements, and physical fitness gains were observed in 7/8 studies. Other physical health areas that had positive effects were balance/coordination (5/7), flexibility/agility (2/3), perceptual motor skills (2/4) and movement/rhythm skills (1/2). No significant change was found in BMI. Jumping and balance had medium and large effects, respectively. For cognitive outcomes, attention (2/2) and organization skills (1/1), but not learning (0/1) skills showed improvement. For psychological outcomes, creativity (3/3), self-concept (1/3), and body image (1/1) showed improvements, unlike attitude toward dance (0/1). For social outcomes, all studies showed improvements.

For methodological quality, 16 studies received a weak rating, 2 received a moderate rating and 1 received a strong rating.

Conclusion: Existing literature is heterogeneous and of poor quality but indicates dance may have physical, cognitive and psychosocial benefits for children with disabilities.

Bias and quality assessment tools: Effective Public Health Practice Project (EPHPP) Quality assessment tool for quantitative studies.

Funding and conflict of interests: The study was funded as part of a National Disability Insurance Scheme Information, Linkages and Capacity building grant. One author received funding from the Ferrero Group Australia, Moose Toys and the Australian Football League. He has previously received speaker honorarium from Novartis (2002), Pfzier (2006) and Nutricia (2007) and is also a Director of the Amaze Board (Autism Victoria).

AMSTAR 2: critically low confidence.

Sugimoto (2016), Research in Developmental Disabilities.

Title: Effects of Neuromuscular Training on Children and Young Adults with Down Syndrome: Systematic Review and Meta-Analysis.

Objectives: To synthesize existing research evidence and examine effects of neuromuscular training on general strength, maximal strength, and functional mobility tasks in children and young adults with Down syndrome.

Included studies: 7 controlled studies (5 randomized and 2 non-randomized).

Population: Children and young adults with Down Syndrome. Sample: 309 participants. Age range: 8-29 y.

Setting: Not stated.

Intervention: Neuromuscular training: strength/resistance (5), balance/bicycle riding training (2), vibration training (1), functional mobility tasks/grocery shelving and timed up- and-down stairs (2).

Comparison: regular activities.

Outcomes: General strength (knee extension and flexion strength), maximal strength (one-repetition maximum values of leg press and chest press), functional mobility performance (grocery shelving tasks and timed up-and-down stairs).

Results: Neuromuscular training could be considered as an effective intervention to promote general and maximal muscular strength enhancement in children and young adults with Down syndrome. A small effect was obtained in functional mobility tasks. In the current analysis, five of the seven reviewed studies were rated as level I (high quality, individual randomized control trial) while 2 studies were rated as level II (lower quality, clinical trial and cohort study).

Conclusion: Although there were limited studies, the results showed that neuromuscular training could be used as an effective intervention in children and young adults with Down syndrome. Conducting further randomized controlled trials with specific strength, balance, and functional performance measurements is warranted in order to develop an effective exercise prescription for children, young adults, and adults with Down syndrome. Improvement in motor proficiency likely helps individuals with Down syndrome to engage with greater physical adaptation, which further leads them to participate in a more physically active lifestyle. The more physically active lifestyle may bring a better quality of life in individuals with Down syndrome.

Bias and quality assessment tools: PEDro scale.

Funding and conflict of interests: The authors declare no funding or conflict of interests.

AMSTAR 2: critically low confidence.

Systematic reviews

Fonzo (2020), Brain Sciences.

Title: Evidence-Based Physical Therapy for Individuals with Rett Syndrome: A Systematic Review.

Objectives: To analyze the most recent evidence concerning the role of physical therapy in the management of individuals with Rett syndrome.

Included studies: 22. case reports (14), case series (2), multiple baseline studies (2), single-case A-B-A-B design study (1), single-case AB design study (1), case-control study (1), and modified individually randomized stepped wedge trial (1).

Population: Individuals with Rett syndrome. Sample: 63 participants. Age range: 3-36 y.

Setting: Not stated.

Intervention: applied behavior analysis, conductive education, environmental enrichment, traditional physical therapy with or without aids, hydrotherapy, treadmill, music therapy, computerized systems, and sensory-based treatment (Snoezelen).

Comparison: No physical therapy.

Outcomes: Motor skills, functioning and quality of life.

Results: Regardless of the type of intervention practiced, all studies included in this review demonstrated that physical therapy improved the quality of life in patients diagnosed with RTT, mainly helping to preserve autonomy. Interventions should be provided early, individualized, and must include appropriate involvement of the patient's family or caregivers.

Conclusion: A physical therapy program should be regularly recommended to patients with RTT, in order to preserve and restore movement and physical function threatened by the disease, preserve autonomy, improve quality of life, and support family caregivers

Bias and quality assessment tools: Oxford Centre for Evidence-Based Medicine (OCEBM)—Levels of Evidence guide.

Funding and conflict of interests: The authors declare no external funding and no conflict of interest.

AMSTAR 2: critically low confidence.

Hocking (2016), International Journal of Evidence-Based Healthcare.

Title: Physical therapy interventions for gross motor skills in people with an intellectual disability aged 6 years and over.

Objectives: To investigate the effectiveness of physical therapy interventions for improving gross motor skills (GMSs) in people with an intellectual disability aged 6 years and older. There is a lack of physical therapy research for GMSs in this population, and no prior systematic review.

Included studies: 7. RCTs (2), pseudo-RCTs (2), repeated measures studies (2), and case report (1).

Population: Children with mild to severe ID. Sample: 82 participants (42 DS, 22 PWS, 13 CP, 2 RS). Mean age range: 10-33 y. Age range for DS participants: ~11-42 y. Age range for PWS participants: ~25-42 y. Age range for CP participants: ~6-26 y. Age range for RS participants: ~6-26 y.

Setting: Community gym, home, rehabilitation hospital, special school, or not reported.

Intervention: Physical therapy interventions: progressive resistance training using weight machines; strength exercises for limbs; completion of log book; partial body weight-supported treadmill training; Home Exercise Program (HEP); Body weight support (BWS); modified-judo training; Nintendo Wii games.

Comparison: No physical therapy.

Outcomes: Gait, standing balance, and composite assessments of gross motor function.

Results: Interventions were well tolerated with negligible adverse effects. In spite of the low overall quality of evidence, results suggest that task-specific training may be useful. The available evidence provides initial support for the consideration of physical therapy interventions, particularly lower limb exercises for cadence, partial body-weight-supported gait training for cadence and nondimensionalized gait velocity, and adapted judo training.

Conclusion: Improved gross motor skills in people with intellectual disability may potentiate greater functional independence for people with an intellectual disability.

Bias and quality assessment tools: Critical appraisal instruments from the Joanna Briggs Institute Meta-Analysis of Statistics Assessment and Review Instrument.

Funding and conflict of interests: A $500 stipend was provided by The Joanna Briggs Institute to the primary author. The authors report no conflict of interest.

AMSTAR 2: critically low confidence.

Maïano (2019), Journal of Intellectual Disability Research.

Title: Effects of motor skill interventions on fundamental movement skills in children and adolescents with intellectual disabilities: a systematic review.

Objectives: To summarize the findings from studies pertaining to the effects of motor skill interventions designed to improve fundamental movement skills (FMS) in children and adolescents with ID.

Included studies: 14. Experimental (9), quasi-experimental (3). and pre-experimental (2).

Population: Children and adolescents with ID. Sample: 464 participants, with mainly mild ID. Age range: 5-22 y.

Setting: Regular and special schools and care institutions. Five studies were conducted in America, four in the Eastern Mediterranean area, four in Europe and one in South-East Asia.

Intervention: Balance and/or strength exercises, adapted play training, handball and hemsball techniques, computerized games, developmental physical education, therapeutic sensorimotor training, physical development program, intensive motor skills training, physical therapy programs, and vestibular stimulation exercises.

Comparison: Conventional physical therapy program, regular school schedule/usual activities or a physical education program.

Outcomes: Balance skills measured with either the Berg Balance Scale, the Pediatric Balance Scale, the gross motor function measure, the Bruininks-Oseretsky Test of Motor Proficiency, and the Lincoln-Oseretsky Motor Development Scale or Test of Motor Proficiency. Overall fundamental movement skills were measured with the basic skills test, or the Frostig Movement Skills Test Battery.

Results: In all but one study the intervention groups have significantly higher post-test balance skills, and overall fundamental motor skills than the control groups. Findings on locomotor skills were mixed. 5 studies were rated as high-quality.

Conclusion: Given the relatively small number of studies and their limitations, the present findings need to be interpreted with caution, and further rigorous studies are necessary.

Bias and quality assessment tools: PEDro scale.

Funding and conflict of interests: Grants from the Social Sciences and Humanities Research Council of Canada. The authors declare no conflict of interest.

AMSTAR 2: critically low confidence.

Appendix 3- Excluded studies with reasons

Not systematic reviews or meta-analyses

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Non-interventional studies

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Wrong population

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7. CONSIDERAÇÕES FINAIS

A sensação de incapacidade, desconhecimento de intervenções possíveis, ou percepção de incerteza do efeito das intervenções conhecidas pode gerar desânimo nos profissionais de saúde, educação e cuidadores de crianças e adolescentes com DI. Recomendações baseadas em evidências podem guiar os envolvidos em direção a um cuidado eficiente.

Esta revisão de revisões fornece substrato científico para a formulação de diretrizes de potencialização da aprendizagem de crianças e adolescentes com DI. As estratégias aqui discutidas, podem ser instituídas de maneira gradual em cenários clínicos e de educação, observando-se sua aplicabilidade à população e contexto brasileiro, aceitação pelas crianças e familiares, de acordo com seus valores e preferências.

As recomendações geradas são, em sua maioria, baseadas em estudos de qualidade limitada, e sintetizadas por revisões com importantes falhas metodológicas. Novos estudos podem avaliar com maior rigor metodológico cada uma das intervenções expostas, de maneira a aumentar a confiança nos efeitos estimados.

Apesar destas limitações, a maioria das intervenções são factíveis, pois são de baixo custo, fácil treinamento do interventor e de fácil aplicação, tendo algum potencial, mesmo que incerto, de benefício, o que na visão dos familiares e pacientes pode ser suficiente.

Para concluir este trabalho, é oportuno citar a conclusão expressiva de Mirenda (2014)[1] em seu estudo intitulado "*Revisiting the Mosaic of Supports Required for Including People with Severe Intellectual or Developmental Disabilities in their Communities"*, no qual ela faz uma avaliação da evolução das tecnologias de assistência à comunicação para indivíduos portadores de deficiência nos 20 anos anteriores.

> *"We were in the throes of suggesting the probable, demonstrating the possible, explaining the processes, and documenting the effective. Now,*

we have made much progress toward each of these goals by working together and by keeping our eyes on the prize. Now, we are challenged to implement the routine and evaluate the systems we devise to accomplish that. If we do this, the lives of people with IDD will be changed again, 20 years from now. We can do this; we must do this".

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