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Highlights in disability, rehabilitation, and inclusion 2021/22

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REVIEW
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A Scoping Review of Evidence-Informed Recommendations for Designing Inclusive Playgrounds

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Background: Playgrounds provide children with many sensory, motor, and socioemotional experiences that are critical to child development. Unfortunately, playgrounds also represent an environment where children with disabilities experience barriers to accessing play. Structures and materials that are prominently found in almost all playground designs (e.g., swings, slides, sand) can present as obstacles for many children with disabilities to engage in independent play.

Aims: This scoping review engaged in the empirical literature to address the research question, "What are the evidence-informed recommendations for designing inclusive playgrounds to enable participation for children with disabilities?" Consideration was given not only to the physical design of playgrounds, but also the playgrounds' surrounding built and social environments.

Methods: A systematic search of Medline, PsycINFO, CINAHL, EMBase, ERIC and Scopus was conducted. Only peer-reviewed literature published in English between January 1990 and January 2021, with a primary focus on inclusive playground structure design related to any type of disability were included. Data extraction included the study author(s), year of publication, country of origin, purpose, disability types considered, methods, sample characteristics and key findings. Key findings were synthesized into evidence-informed recommendations, which were later collated, using inductive content analysis, into five broader thematically congruent groups.

Results: Thirty-five studies were included using case study ($n = 17$); observational ($n = 6$); survey ($n = 5$); experimental ($n = 4$); and multiple study ($n = 3$) designs. Thirteen evidence-based recommendations and one promising practice were categorized into five broad playground elements: entry points; surfacing and paths; features to foster inclusive play; staffing/supervision; and design process.

Conclusion: These recommendations build upon previous design-based best-practices that focused exclusively on the physical design of the playground. Our recommendations have implications for how future playgrounds should be designed to maximize usability and inclusiveness and the overall playground experiences for children with disabilities.

Keywords: inclusive playgrounds, playground design, childhood disability, play, accessibility

INTRODUCTION

It has been over 30 years since the United Nations Convention on the Rights of the Child formalized play as a fundamental human right of all children (1). The more recent United Nations Convention on the Rights of Persons with Disabilities offers further support for children with disabilities regarding equal access to play (2). Despite these declarations of human rights, the United Nations has acknowledged that the unique needs, interests, and rights of children with disabilities have continued to be overlooked, including those concerning equal access to play opportunities (3). This oversight is troubling given that play is integral to children's cognitive, physical, and social development, and to their emotional well-being (4–7).

Schoolyards and parks are an integral part of the larger experience of play within children's communities. Playgrounds—defined in this article as constructed play areas that contain traditional play equipment (e.g., swings, slides, merry-go-rounds) on the ground as well as structures built with paths to and between elevated play equipment (8)—are omnipresent within the landscape design of these spaces and constitute a significant part of the overall play experience available to children. Unfortunately, playgrounds also represent an environment where children with disabilities experience barriers to accessing play (9–16). Although playgrounds are designed to provide children with an array of opportunities to engage in different types of play, the designs are frequently informed by normative understandings of children's bodies, mobilities, and abilities that do not adequately account for the presence of childhood disability. The resultant playground designs can create inequitable access to play opportunities and may cause children with disabilities to experience exclusion (17–19).

While children with disabilities continue to experience barriers to accessing play opportunities across playgrounds, their needs are beginning to be recognized in playground design research and practice as the concept of inclusive play continues to develop (20). Inclusive play and, correspondingly, inclusive playgrounds, are intended to remove physical and social barriers to participation through designs that provide an environment where all children can play together using the same equipment (20). We conducted a scoping review of the nascent literature on inclusive playground design to contribute an updated, comprehensive analysis that can inform scholars and practitioners in designing playgrounds to enable and include children with disabilities. Our review engaged the following research question: "What are the evidence-informed recommendations for designing inclusive playgrounds to enable participation for children with disabilities?" By engaging this question, our scoping review aims to identify key playground design factors that have been found to improve play equipment usability and overall playground experiences for children with disabilities. Prior to presenting our scoping review process, results, and discussion, we briefly discuss three topics to provide necessary context: (i) playground experiences for children with disabilities; (ii) playground design standards; and (iii) past playground reviews.

Playground Experiences for Children With Disabilities

Considering playground play is largely unstructured, it gives children opportunities to advance their imagination, self-awareness, risk perception, and identity, as well as their social and motor skills (17, 18). The diverse play opportunities that can emerge within playgrounds make these spaces a unique setting where children can choose when, where (i.e., in relation to specific equipment), and how to interact with others while playing together or sharing the play space. Although children with disabilities value playgrounds as play spaces (9, 17, 21), playground designs often do not provide them with equal and equitable access to play opportunities (9, 10, 12, 13, 15, 16). Hence, children with disabilities can face numerous barriers when trying to access playground play. Sometimes, these barriers are encountered at playground entrances (e.g., raised borders). This can make it difficult or impossible to access the playground space—let alone its play equipment—without assistance from caregivers (19, 22, 23). Further, the absence of ramps from elevated play structures can restrict some children with disabilities from accessing and moving freely on the structures (10, 13, 19). In the rare cases where elevated play structures include ramps, the structures' accessible routes can terminate at dead ends that do not provide play opportunities, access, or egress (21, 24).

In addition to issues concerning playground surfaces and elevated play structures, the play components (e.g., slides, merry-go-rounds) themselves are often inaccessible. In fact, adapted play components that enable children with physical disabilities to fully and safely engage in playground play (e.g., slides that can be accessed via ramps, wheel-on merry-go-rounds) remain largely absent from playgrounds (9, 10, 14, 15, 17). Many playgrounds also lack sensory-based play components that may promote active engagement among children with developmental disabilities, such as tactile play components that offer different textures to touch and manipulate, or musical play components that produce a variety of sounds and vibrations (25, 26). These ongoing playground design issues may explain why playgrounds have been identified as landscapes where children with disabilities can end up feeling isolated, excluded from peer interaction, or excluded from the play space entirely (17–19).

Playground Design Standards

In North America and beyond, there are various accessibility standards that apply to playgrounds (e.g., Americans with Disabilities Act Standards for Accessible Design, Canadian Standards Association Standard for Children's Playspaces and Equipment, and Australian Standards' "AS 4685 Playground Equipment and Surfacing" and "AS 1428 Design for Access and Mobility"). These standards support the implementation of access ramps to elevated components, the provision of accessible play components that are at ground level and elevated, and the removal of barriers from playground entrances and pathways (27). Although the presence of standards represents a shift toward improving access to play opportunities for children with disabilities, the standards include limitations and do not necessarily ensure inclusive play opportunities.

One notable limitation is that playground accessibility standards are largely informed by the opinions of playground designers that have outpaced (and are thus no longer informed by) scientific evidence (28). Additionally, playground accessibility standards have often focused on addressing barriers for children with mobility impairments more so than barriers for children with sensory or developmental disabilities. As a result, playground designs often prioritize play for children with mobility impairments (25) and disregard the play of children with sensory or developmental disabilities. More empirical research, including the voices of parents and children experiencing disability, on accessible and inclusive playground design that will inform playground standards is sorely needed.

How practitioners engage and treat accessibility standards is another key concern. For example, by treating minimum accessibility standards uncritically as fixed accessibility standards (i.e., by not carefully assessing if minimum standards should be exceeded to suit a specific site and its users' needs), practitioners may produce technically accessible landscapes that meet legal requirements that are functionally inaccessible to some (29). This uncritical treatment of standards remains a concern as municipal employees who work in or in relation to parks and playgrounds have expressed having limited knowledge about inclusive design beyond addressing accessibility (e.g., adding ramps where required) and that they have no available standards for reference (10).

Past Playground-Related Reviews

Our review builds upon two past reviews by Moore and Lynch (28) and Fernelius and Christensen (20). Moore and Lynch (28) conducted a scoping review of 14 studies that explored the accessibility and usability of playgrounds for children of all abilities. Their overarching recommendation was that the Principles of Universal Design (Center for Universal Design, 1997) should be considered when designing playgrounds to promote inclusion through equal and equitable access to play options for all children, including those with disabilities. The suggested principles would help to ensure: (i) equitable use, (ii) flexibility in use, (iii) intuitive use, (iv) provision of perceptible information, (v) tolerance for error, (vi) minimal physical effort to access, and (vii) appropriate size and space for approach and use (30).

Fernelius and Christensen's (20) review of 22 studies identified 10 specific physical design elements to improve playground play for children with disabilities. These design elements support the use of: (i) circular playground design, (ii) common and recognizable objects, (iii) loose parts, (iv) accessible surfacing and sufficient space, (v) elevated and ground level components, (vi) multi-niche settings, (vii) equipment that provides appropriate levels of challenge and risk, (viii) observation points, (ix) comfortable places, and (x) sensory stimulus.

While these two past reviews have identified ways to improve a playground's physical design in order to create play opportunities for children with disabilities, their focus has primarily been on the playground structure itself, therefore there may be additional evidence-informed recommendations to consider. For example, since playgrounds are inherently a social experience for children, and are not experienced in isolation from their surroundings, it is sensible to consider ways in which a playground's surrounding built and social environments can enhance playground experiences for children with disabilities. We engage this gap in this scoping review by considering playgrounds' physical designs, social environments, and surrounding built environments. Our intent in expanding our scope in this way is to help readers begin to move past creating play opportunities for children with disabilities through just physical design elements within the borders of the playground toward a more comprehensive approach focused on ensuring children with disabilities and their families experience inclusion during playground visits.

MATERIALS AND METHODS

Scoping reviews are a rigorous and transparent approach for synthesizing evidence when the purpose is to capture the relevant literature on a topic, regardless of the study design (31, 32). Our review follows the five recommended stages identified within existing frameworks for conducting a scoping review (31–35): (i) identifying the research question; (ii) identifying relevant studies; (iii) study selection; (iv) charting the data; and (v) collating, summarizing, and reporting the results. The following sections provide further details on stages ii–v of this review process.

Identifying Relevant Studies

The research team developed the initial search strategy in consultation with an academic librarian. The search strategy was intentionally broad to maximize coverage of all relevant studies (35). This involved using search terms related to playgrounds: "playground*," "playspace*," "play space*," "playscape*," "play component*," "play area*," "play structure*," "play park*," and "play environment*." For this review, the term "playground" refers to play areas built as part of schoolyards or parks that contain traditional play equipment (e.g., swings, slides) at ground level and structures built with paths to and between elevated play equipment (8). Disability theory and research, and hence the concepts and terms used to describe and understand disability, have evolved considerably over the past three decades. Only using playground-related search terms circumvented the potential to exclude articles that have used a variety of terms to describe different disabilities (e.g., blind vs. visually impaired) and disability design (e.g., accessible, inclusive, universal, barrier-free).

After identifying journal databases in consultation with an academic librarian, the first author conducted literature searches across Medline, PsycINFO, CINAHL, EMBase, ERIC, and Scopus databases, from January 1990 to July 2019, which was later updated in March 2020 and again in January 2021. All captured search records were exported into an online review management system that identified and removed duplicate records from the database (Covidence, Veritas Health Innovation, Melbourne, Australia). The first author completed a subsequent search of all included articles' reference lists after the full-text screening stage and the reference lists of excluded position papers and reviews to identify any additional relevant articles.

Study Selection

Study inclusion criteria are provided in **Table 1**. Given that scoping reviews involve an iterative process rather than the linear process adopted by systematic reviews (31), the four-person study selection team (DB, KAN, TR, JL) regularly discussed criteria during the search process and modified them (i.e., to the criteria in **Table 1**) as the nature of the literature became apparent. Manufactured play structures were focused on due to the inherent differences that exist between manufactured and natural playgrounds in terms of their affordances and play opportunities (36).

The study selection team began screening by applying inclusion/exclusion criteria to the titles and abstracts of 100 randomly selected records (∼1%). Once completed, they discussed decision discrepancies to support reliability during the screening process (37). Next, the four reviewers independently screened the titles and abstracts of the remaining 99% of records. Each record was screened by two reviewers and inconsistent decisions were resolved by a third reviewer. Upon completing the title and abstract screening stage, the first author retrieved the full-text articles for all records that met inclusion and exclusion criteria. Each full-text article was screened by two reviewers (DB, KAN) to further determine whether it should be included for review. Again, inconsistent decisions were resolved by a third reviewer (JL). Reasons for exclusion were recorded at the full-text screening stage.

Charting the Data

The research team collectively determined which attributes of the articles to extract for summary and analysis after piloting the Microsoft Excel-based data charting form with a representative sample of the studies to be reviewed. The finalized data charting form was developed for extraction of the following study attributes: author, year of publication, country of origin, purpose, disability types considered, methods, sample and key findings. The first author independently extracted and charted the data from each article. The senior author (KAN) checked the extractions and updated the data charting form in an iterative process.

Collating, Summarizing, and Reporting the Results

The research team summarized and reported the key findings that emerged from the charting process. The first author then synthesized the key findings into potential evidence-informed recommendations using an inductive content analysis approach (38). This approach involved applying codes to the key findings to reduce and group data into mutually exclusive concepts (recommendations). Next, the research team reviewed and revised the coding for the potential evidence-informed recommendations and further reduced and grouped the data, which were later collated into five broader thematically congruent groups (i.e., playground elements). Given the broad range of key findings identified in each study, each article could be mapped to multiple recommendations based on its contents. Next, three authors (DB, KAN, JL) independently reviewed the recommendations and playground elements prior to discussing as a team until consensus was achieved. Through team discussion, the recommendation with only one study for support was relabelled as a promising practice and identified as an area for future research.

RESULTS

Selection of Studies

The search yielded 16,261 records which was reduced to 10,360 after duplicates were removed. After screening the title and abstract of each record using the inclusion/exclusion criteria, 163 articles remained and their full texts were obtained. Full-text screening for inclusion was completed independently by two reviewers, resulting in 139 records being removed and 24 records being selected for inclusion. Reference lists of these 24 articles were screened for missed records, resulting in an additional five articles. Updated searches using the original search strategy were conducted in March 2020 and January 2021, providing an additional six articles to be included (three articles in each respective update). Overall, a total of 35 articles were selected for full review (see **Figure 1** for PRISMA flow chart).

Study Characteristics

The country of origin, purpose, disability types considered, methods, sample and key findings for each study are presented in **Supplementary Table 1**. Two studies were published in the 1990s (1996, $n = 1$; 1999, $n = 1$), six studies were published from 2000 to 2009 (2000, $n = 1$; 2001, $n = 2$; 2006, $n = 2$; 2007, $n = 1$), and the remaining 27 studies were published from 2010 to 2020 (2010, $n = 3$; 2011, $n = 2$; 2012, $n = 4$; 2015, $n = 2$; 2016, $n = 2$; 2017, $n = 3$; 2018, $n = 5$; 2019, $n = 1$; 2020, $n = 5$). Research was conducted in Australia ($n = 4$), Brazil ($n = 1$), Canada ($n = 3$), Hong Kong

TABLE 1 | Scoping review study inclusion and exclusion criteria.

Inclusion criterion	Exclusion criterion
(1) Primary focus on inclusive playground structure design;	(1) Not a primary data collection study (e.g., position paper, review);
(2) Focus on disability (any type);	(2) The full text could not be obtained;
(3) Primary peer-reviewed studies of qualitative, quantitative, or mixed study design in order to consider different findings that have the potential to inform inclusive playground design practices;	(3) Playground was defined in an alternative context (e.g., an environmental playground of bacteria);
(4) Written in English;	(4) Focused on natural playground design (e.g., garden, forest);
(5) Published since 1990*	(5) Focused on playground injury epidemiology;
	(6) Focused strictly on playground design for safety

**A January 1990 search start date inclusion criterion was used to acknowledge (and to capture changes in best practice recommendations since) the passing of the Americans with Disabilities Act (ADA).*

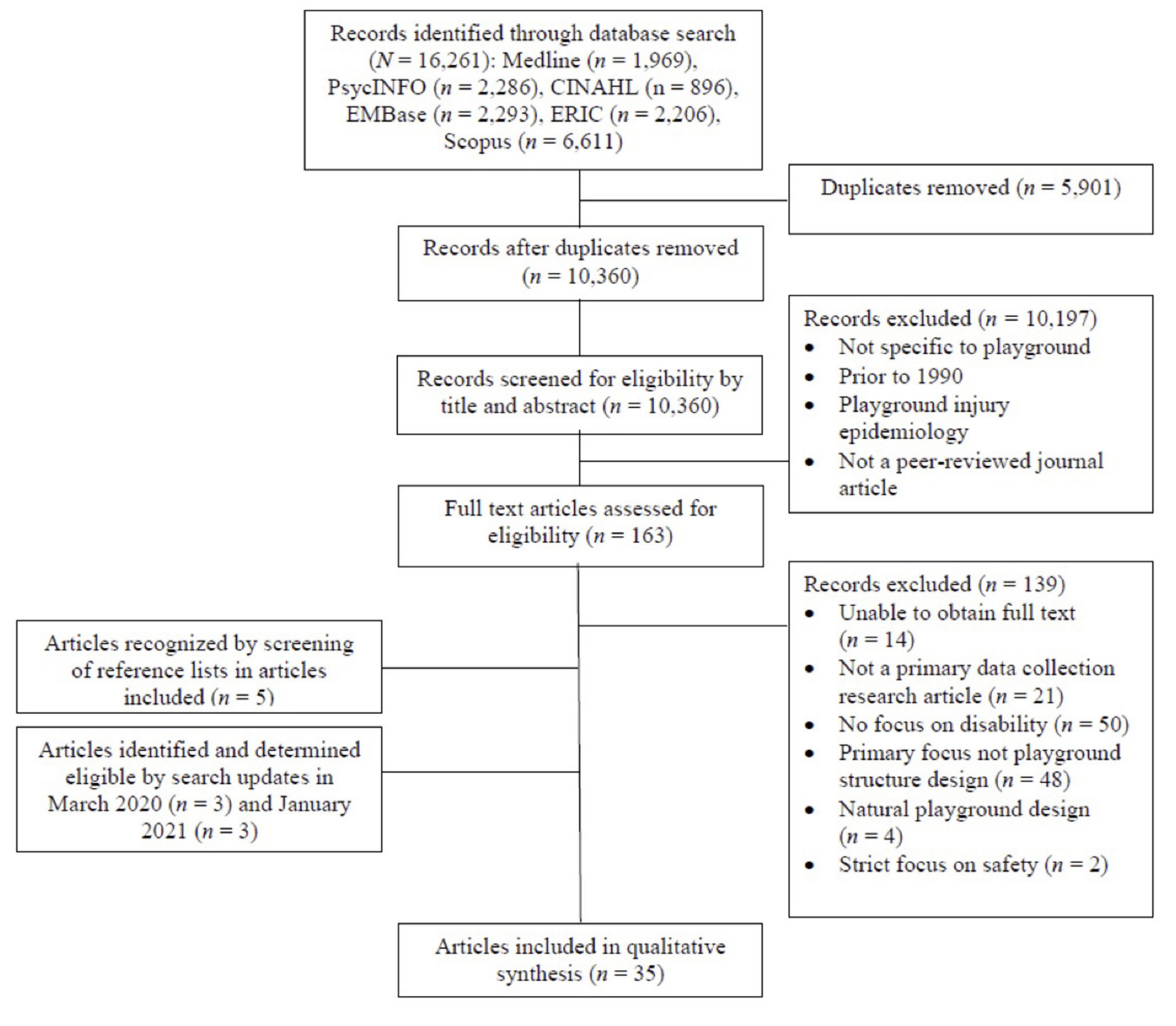

FIGURE 1 | PRISMA flow chat.

($n = 1$), Israel ($n = 1$), the Netherlands ($n = 1$), New Zealand ($n = 1$), Poland, ($n = 1$), Sweden ($n = 4$), Switzerland ($n = 1$), Turkey ($n = 2$), United Kingdom ($n = 3$), and the USA ($n = 12$). The studies used a range of different designs including case studies ($n = 17$), observational ($n = 6$), survey ($n = 5$), experimental ($n = 4$) and multiple study designs ($n = 3$).

TABLE 2 | Summary of the evidence-informed recommendations and supporting evidence for designing inclusive playgrounds for children with disabilities.

Playground element	Recommendation	Supporting evidence
1. Entry points		
	1.1. Entrance to the playground space is wide and free of any obstacles	(12–14, 16, 19, 25, 39, 40)
	1.2. Wide, flat and firm pathways from the entrance to the playground	(10–15, 22, 39, 41, 42)
	1.3. Enclosing the playground to prevent children from straying (*Promising Practice*)	(12)
2. Surfacing and paths		
	2.1. A flat uniform surface that consists of material that is moderately firm and stable	(11–19, 21, 23, 24, 39, 41, 43–47)
	2.2. Ramps that provide access to and between elevated play components	(10–13, 19, 21, 24–26, 39–42, 45, 48, 49)
3. Features to foster inclusive play		
	3.1. Play equipment accessible to all children	(9, 10, 12–17, 21, 22, 24–26, 39–42, 47, 48, 50–53)
	3.2. Variety of play equipment that provides appropriate challenges for children of all ages and abilities	(10, 17, 19, 21, 24, 39, 42, 46–48, 50, 53)
	3.3. Different types of sensory play components that are spread out within the play space to reduce overstimulation	(12, 21, 25, 26, 39, 41, 42, 53, 54)
	3.4. Solitary play components for escaping overstimulation	(12, 17, 39, 41, 42)
	3.5. Play components shaped in recognizable designs that allow for creative and imaginative pursuits	(17, 25, 50)
	3.6. Informational features to aid with spatial orientation, communication and guidance on proper use of equipment	(10, 12, 17, 25, 26, 39–42)
	3.7. Shaded spaces to aid with temperature regulation	(11, 21)
4. Staffing/Supervision		
	4.1. Trained staff present in the play space to support play for all children	(9, 14, 24, 40, 48, 55, 56)
5. Design process		
	5.1. User involvement (families of children with disabilities and representatives from disability organizations) in the design process	(13, 14, 21, 46, 47, 52, 55)

Sample Characteristics

Twenty-five studies involved data collection with human participants, with sample sizes ranging from one to 667 participants. These studies included children with and without disabilities, parents/caregivers of children with and without disabilities, school staff, municipal employees, playground designers, and not-for-profit organization representatives. Studies primarily focused on children with varied disabilities ($n = 15$) and physical disabilities ($n = 8$), although one study focused on children diagnosed with autism spectrum disorder (ASD; $n = 1$) and two studies did not specify the disability focus ($n = 2$). Specific impairments represented in the 25 studies included physical (e.g., lower extremity prosthesis, cerebral palsy, mobility impairments), learning, developmental, sensory processing (e.g., ASD), visual (e.g., congenital blindness, severe visual impairment), auditory, and intellectual impairments/disorders. Among the 11 studies focused on playground audits or testing of playground surfaces, sample sizes ranged from one to 355. These studies focused on playground surfaces ($n = 2$) and audits of playgrounds at schools or parks ($n = 6$), boundless playgrounds ($n = 1$), adapted playgrounds ($n = 1$) and playgrounds at schools for children with disabilities ($n = 1$). One study involved data collection with human participants and an audit on the infrastructure of the playground which explains why 36 studies are described in this section.

Evidence-Based Recommendations

Our analysis of each study's key findings identified 13 evidence-based recommendations for designing inclusive playgrounds for children with disabilities. One additional promising practice was identified, based on findings from one study. These recommendations and the promising practice were classified into five broad playground elements: entry points; surfacing and paths; features to foster inclusive play; staffing/supervision; and design process. A summary of the recommendations for each playground element is provided below. **Table 2** provides an overview of these evidence-informed recommendations.

Entry Points

This playground element concerns the playground's perimeter, including any entrance, and the paths that provide access to the playground surface. Two evidence-informed recommendations were identified for entry points: (i) ensure playground entrances are wide and free of obstacles; and (ii) provide wide, flat and firm pathways leading to the playground. An additional promising practice (based on findings from one study) was identified for the entry points playground element–enclose the playground to prevent children from straying.

Wide, Flat and Firm Pathways Leading to The Playground

The importance of having wide, flat and firm pathways leading to a playground was highlighted in 10 of the 35 studies (10–15, 22, 39, 41, 42). Interviews with playground users and municipal playground personnel have shown children with disabilities, particularly those with mobility impairments, often cannot access the playground without assistance (13). Obstacles limiting movement into the playground include paths that have irregularities (13, 14, 22) and/or unstable ground cover, such as sand or pea gravel (12–14). To improve access to playgrounds, all paths should be wheelchair accessible (10, 41, 42) and wide enough to allow unobstructed movement into the play space (41, 42). Paths wide enough for two wheelchairs to move side-by-side have been recommended (42). While existing accessibility standards (e.g., ADA Standards for Accessible Design) mandate the availability of accessible pathways, playground audits indicate accessible pathways are uncommon (11, 15, 39), and in cases where accessible paths are present, they are often too narrow (12).

Playground Entrances That Are Wide and Obstacle-Free

Having a wide playground entrance that is clear of any obstacles was also found to be important [eight of 35 studies; (12–14, 16, 19, 25, 39, 40)]. Although studies have suggested that these entrances should be wide enough to accommodate those using mobility devices (25, 40), playground users and municipal playground personnel have indicated via interviews and surveys that this is often not the case (13, 14, 39). Raised playground borders (12, 19) and bollards positioned in the middle of entrances are two examples of obstacles known to make entering playgrounds more difficult for children who use mobility devices (16).

Enclosed Playground Space to Prevent Children From Straying

One study (12) highlighted the importance of enclosing the playground space to prevent children from straying. In a study examining the accessibility and usability of 21 parks in New Zealand, only two of the playgrounds observed had appropriate fencing (> 1.2 m high) (12). The authors suggested that enclosing the playground with fencing may help keep children prone to straying, such as those with autism spectrum disorder (ASD), within the play space and away from potential hazards (e.g., open water, roads).

Surfacing and Paths

This playground element refers to the surface on which the play components are installed and the paths onto and between elevated play structures. Two evidence-informed recommendations emerged for surfacing and paths: (i) use a flat, uniform surface that consists of material that is moderately firm and stable; and (ii) incorporate ramps that provide access to and between elevated play components.

Flat, Uniform Playground Surface

Of the 35 studies, 19 (11–19, 21, 23, 24, 39, 41, 43–47) highlighted the importance of using a flat, uniform surface constructed from material that is moderately firm and stable. Children with mobility impairments have expressed that many playground surfaces impede their access to play equipment (18). Using unstable or overly soft ground cover such as pea gravel, engineered wood fiber, and sand may pose the greatest barrier to playground use for children with mobility impairments (14, 16, 17, 19, 21, 23, 24, 45–47). Poor surface material choices may limit independence, with children using mobility devices requiring assistance from a teacher to join their peers at the playground equipment (24). Playground audits in different countries have revealed most playgrounds have surfacing that fails to provide accessible routes to and between play equipment (11, 12, 15, 19, 39). Parents of children who use a lower extremity prothesis have reported that firm, flat surfaces are the easiest for their child to navigate (45). In fact, two experimental studies comparing different types of playground surfaces have demonstrated that poured-in-place rubber may be the ideal surface, as its moderately firm and stable profile promotes safety and accessibility (43, 44), thus enhancing usability and participation. Caregivers and children with disabilities have reiterated the importance of using a firm, shock-absorbent playground surface that provides an ideal blend of accessibility and safety (41).

Ramps That Provide Access to and Between Elevated Play Components

The second recommendation concerns the importance of having ramps that provide access to and between elevated play components [16 of 35 studies; (10–13, 19, 21, 24–26, 39–42, 45, 48, 49)]. A study involving parents of children with disabilities revealed that their children view being on the elevated play structure as more fun than ground-level activities (26). Aside from playgrounds purposely designed with accessibility in mind (25), most playgrounds lack ramp systems that provide access to elevated play structures (10–13, 19, 39). This is problematic given that stairs and ladders—equipment commonly used to provide access to elevated components—may be inaccessible and unsafe for children with mobility and visual impairments (40, 45). In addition to providing ramp access to elevated play structures (21, 41, 42), playground designers should do more to ensure there are inclusive play opportunities available on the accessible elevated play structures, as some have been found to provide no such options and, sometimes, they simply lead to dead ends (21, 24). Incorporating looped paths into accessible elevated play structures may facilitate play by enabling children to move continuously throughout the structure (48, 49).

Features to Foster Inclusive Play

This playground element refers to play components that account for the variety of needs, abilities, and desires of children with disabilities to facilitate quality play experiences and overall participation in playgrounds. We identified seven recommendations for features that foster inclusive play: (i) implement play equipment that is accessible to all children; (ii) ensure a variety of play equipment that provides appropriate challenges for children of all ages and abilities; (iii) provide and spread out different types of sensory play components across the play space to reduce overstimulation; (iv) offer solitary play components for escaping overstimulation; (v) implement play components shaped in recognizable designs that allow for creative and imaginative pursuits; (vi) incorporate features to aid spatial orientation, communication and guidance for using play space; and (vii) provide shaded spaces to aid body temperature regulation.

Play Equipment Accessible to All Children

Having play equipment that is accessible to all children was highlighted most often among features to foster inclusive play [23 of 35 studies; (9, 10, 12–17, 21, 22, 24–26, 39–42, 47, 48, 50–53)], as widely accessible equipment promotes inclusion (9, 52) and fosters interaction between children with and without disabilities (50, 53). Children with disabilities and parents of children with disabilities both voiced a desire for playgrounds that offer adapted equipment often not found at conventional playgrounds that meets their child's needs, abilities and interests (10, 47). However, studies have found a lack of specialized equipment (e.g., wheelchair accessible swings) for children with disabilities across playgrounds (12, 13, 15, 22, 24, 26, 39, 40, 52, 53). Traditional types of playground equipment (e.g., slides, swings) are generally inaccessible to some children with disabilities without caregiver assistance (14), and the work of physically transferring a child becomes increasingly difficulty as children grow older and heavier (10, 16). Shapiro (42) highlighted the importance of designing play components that can be accessed independently or with minimal transfer work, but evidence suggests that such equipment remains rare on playgrounds (51). Research also indicates that intuitive, easy-to-use playground equipment may be enabling to children with developmental disabilities (17). Multiple studies have recognized the importance of designing playground equipment and layouts such that they offer adequate space for children using mobility devices to maneuver onto and use equipment with ease (17, 25, 39, 40). Having a playground design that can accommodate the presence of adults can be helpful for having assistance readily available (48).

Some examples of the adapted equipment in the literature include raised sandboxes that accommodate children using wheelchairs (25, 39, 41), merry-go-rounds with ramped or flush surface access (15), swings with full body support (12), and static-free roller slides that can be used by children with cochlear implants without discomfort (42). Children with disabilities have identified swings as a favorite piece of play equipment (17, 21). Playground designers should be encouraged to include various swing types and sizes to support the inclusion of children with specific needs (e.g., wheel-on swings, full body support swings) and children or youth who are larger (17).

Variety of Play Equipment That Provides Appropriate Challenges

Twelve studies (10, 17, 19, 21, 24, 39, 42, 46–48, 50, 53) recognized the importance of having a variety of play components that provide appropriate challenges for children of all ages and abilities. Having a diverse range of play components provides children with opportunities to self-select activities that match their abilities and interests (42, 48), while also helping to promote several important aspects of healthy development [i.e., social emotional, perceptual motor, physical, intellectual, sensory; (39)]. Notably, one study found school playgrounds have limited diversity in terms of the play opportunities available to children with physical disabilities due to a lack of accessible equipment (19). Although some play components may have limited inaccessibility, children with disabilities have also voiced their enjoyment of observing other children challenge themselves when using play equipment such as climbing walls (47). For families of children with varying abilities, these findings have important implications pertaining to whether or not these families can go to playgrounds to play together. Playgrounds have also been found to lack developmentally appropriate play components for older children with disabilities (10, 21) and children without disabilities in general (10, 17, 24, 46, 53).

Different Types of Sensory-Based Play Components

Nine studies (12, 21, 25, 26, 39, 41, 42, 53, 54) highlighted the importance of having different sensory-based play components within playgrounds, including musical elements (12, 25, 26, 39, 41, 42, 54), tactile play components (21, 25, 26, 39, 42), and visual stimuli (39, 42). Including sensory elements is important for engaging children with sensory processing disorders and visual impairments on playgrounds (26, 53, 54). These sensory-based components should be spread throughout a playground to help prevent experiences of overstimulation among children with sensory processing disorders (42).

Solitary Play Components for Escaping Overstimulation

Five studies (12, 17, 39, 41, 42) called for solitary play components to offer escapes from overstimulation. These solitary play components are helpful to those children with disabilities who desire quiet, private places within a playground where they can relax away from adults (17, 41). Shapiro (42) proposed that areas of solitude may also provide children with disabilities with a sense of security. Despite the importance of areas for escaping overstimulation, play components that provide solitary spaces are rarely, if ever present at playgrounds (12, 39).

Play Components Shaped in Recognizable Designs

The importance of having play components shaped in recognizable designs (e.g., like a car) that foster creative and imaginative pursuits was evident in three studies (17, 25, 50). Two studies involving children with varied impairments revealed a desire for play equipment shaped in recognizable designs such as cars or houses (17, 50), although playground designers should ensure that these imaginative play components are spacious and wheelchair accessible (25).

Informational Features to Aid With Spatial Orientation, Communication and Guidance on Equipment Use

Nine studies (10, 12, 17, 25, 26, 39–42) highlighted the importance of informational features that aid in spatial orientation, communication, and guidance on equipment use to enhance the play experiences of children with disabilities. For instance, having a central auditory or visual cue such as a waterfall may assist with spatial orientation (42). Relief maps can provide important three-dimensional information for wheelchair users about the playground landscape (39). Despite evidence suggesting a lack of use (10, 12, 17), contrasting colors can also be used to help with spatial orientation by demarcating changes in surfacing (e.g., stairs) and potentially dangerous zones (e.g., areas around swings). An audit of playgrounds in Hong Kong revealed limited informative signage (40), although site maps and sign walls were desired to help children with disabilities understand the play components and to allow them to express their interests (41, 42). Signage should be presented in multiple formats (e.g., photos/diagrams, braille description of play component usage) to meet the diverse needs of children with disabilities (41), and to assist parents in supporting their child with navigating the playground space (17). Braille play elements can be invaluable for children with visual impairments and should be incorporated into future playgrounds (25, 26).

Shaded Spaces

Of the 35 studies, only two recognized the importance of providing shaded spaces on the playground for children who have difficulty regulating their body temperature (11, 21). Although shaded spaces can enhance the inclusion of children with disabilities on playgrounds, an audit of 57 playgrounds revealed only 14% of them offered some type of accessible shade (11).

Staffing/Supervision

This playground element refers to the presence of trained staff who can supervise and assist children on playgrounds. Seven of the 35 reviewed studies recognized the importance of having trained staff present to promote playground inclusion (9, 14, 24, 40, 48, 55, 56). The presence of trained staff was acknowledged as being important to initiating play (56), as staff can help to facilitate integrated play among children with and without disabilities (55), and provide children with disabilities with physical or instructional assistance to use equipment (9, 14, 24, 48). Interviews conducted with school staff have revealed that having trained staff present is helpful for modeling appropriate behavior and supporting children in managing their emotions (56).

Design Process

This playground element refers to the stages involved in determining how a playground should be designed with a focus on meeting the needs of children with disabilities. Including playground users in the design process was a key recommendation that emerged for this playground element [7 out of 35 studies; (13, 14, 21, 46, 47, 52, 55)]. Despite research indicating that municipal playground personnel and the construction industry have inadequate knowledge regarding the needs of children with disabilities (14, 52), there has been a lack of representation from individuals with lived experience (e.g., children with disabilities and their caregivers) or those who work closely with children with disabilities (e.g., disability organizations, occupational therapists) when designing playgrounds (13, 14, 52). Five studies have argued that the involvement of families of children with disabilities in the design process is critical for advancing accessibility and inclusion within playgrounds (21, 46, 47, 52, 55). Gaining insight from children with disabilities about their experiences with different play opportunities available on playgrounds can provide important feedback for future design as well as retroactive adaptations aiming to optimize inclusion (52).

DISCUSSION

The lack of evidence-informed recommendations in inclusive playground design is a significant practice gap that municipalities and families of children with disabilities continue to call upon for further action (52). Our scoping review addresses this call to action by synthesizing the empirical evidence on inclusive playground design recommendations that consider not only the physical design of the playground, but also the surrounding built and social environments of playgrounds for children with disabilities. Thirteen evidence-informed recommendations and one promising practice emerged from our analysis of findings from the 35 reviewed studies. These recommendations consider entry into the playground space, play components to foster inclusive play, the role of trained staff within playgrounds for facilitating social inclusion, and the involvement of families of children with disabilities, in addition to rehabilitation professionals and disability organization representatives, in the playground design process. Our recommendations have implications for how future playgrounds should be designed to maximize playground usability and inclusiveness for all children.

The design of an inclusive playground's surrounding built environment should be considered by playground designers and municipalities as they work toward providing children with disabilities with opportunities to fully participate in outdoor play (2). If the built environment surrounding an inclusive playground is excluded from design considerations, the result may be that exclusionary surroundings render the playground inaccessible to many children and caregivers alike. This may of course deter families from wanting or even being able to visit the playground in the first place. Findings from this paper and others (20, 28, 57) indicate that the environmental design of entry points and surfacing and paths warrant as much care and attention as the playground itself. Barrier-free entry points are necessary to ensure easy access to the playground, particularly for children with mobility-related impairments [e.g., (13, 25, 40)]. While deemed as a promising practice, enclosing the playground space to prevent children from straying highlights the additional consideration of the playground site's proximity to hazards such as road traffic and open water (e.g., rivers, drainage ditches). Proximity to a hazard does not necessarily make a site unsuitable for an inclusive playground, as the hazard can be mitigated via design interventions (e.g., fencing, signage) or, possibly, the removal of a hazard. With many playgrounds located within schoolyards and parks, it would be useful to have the importance of playgrounds' accessible surrounding environments acknowledged among those designing, building, and servicing areas around the playground. This may help to avoid creating barriers to the playground and, perhaps, find ways to enhance a playground's surrounding environment (e.g., via well-designed and serviced parking and pathways, building nearby washroom facilities).

Once children can fully access the playground space, there are essential play component features within the playground that could enable meaningful play for children with disabilities. Existing accessibility standards and guidelines provide technical accessibility requirements or guidelines that must be followed or, at least considered, depending on the policy context. These guidelines or standards often equip designers with "general levels of usability" (58) to help ensure that children with disabilities can access play components within the designated play space. However, as was found in several of the studies included in our review, these standards and guidelines are not often engaged critically, with little consideration given to exceeding minimum requirements to promote play opportunities in playgrounds that offer equal and equitable play opportunities to *all* children, regardless of ability (2, 3). For example, under the ADA Accessibility Guidelines (see subsection 15.6.3), which was applied in several studies included in this review [e.g., (11, 39, 49)], at least 50% of elevated play components, if provided, are required to be located on an accessible route. This may limit some children using mobility devices from accessing elevated play structures and, in turn, the learning/play opportunities and peer interactions associated with such structures.

Several studies found that children with mobility impairments required assistance from adults (e.g., caregivers, teachers) to access elevated structures and their play components so that they could join peers in play [e.g., (13, 24)]. Ramping to elevated play components, as per our surfacing and path recommendation (Recommendation 2.2 in **Table 2**), is one way to enhance access for those children who use mobility devices, and thus lessen the labor undertaken by parents or other caregivers to facilitate play (59). Yet even with ramping, children might require transfers from mobility aids to equipment—and so having sufficient space for transfers, and play equipment that can accommodate more than one body simultaneously, should be considered. Our evidence-informed recommendations encourage developers to critically question going beyond minimum requirements as they prepare their designs by considering the quality of playground experiences for all children as well as their families. Focusing on playground designs that intentionally foster quality play experiences (e.g., autonomy, belonging, challenge, engagement) (60, 61) and create settings that enhance dignity in play (62) may create more meaningful participation for all children, not only those with disabilities.

In addition to physical design elements, our review highlights two social design elements (staffing/supervision and user engagement in design process) that may help with addressing physical and social barriers that may challenge and exclude children with disabilities from playgrounds. While the involvement of families of children with disabilities and occupational therapists in the playground design process was acknowledged by Moore and Lynch (28), we have explicitly outlined the importance of this element as a design recommendation. Having representation from individuals who are aware of the unique needs and interests of children with disabilities (e.g., the children themselves and their parents) can help with filling playground designers' knowledge gaps about experiences of childhood disability on the playground (13, 14, 21, 46, 47, 52, 55). However, as noted by van Melik and Althuizen (52), the involvement of families of children with disabilities must be balanced with the limited time, resources, and added pressures that families of children with disabilities face. Several studies also suggested that rehabilitation professionals working with children with disabilities (e.g., occupational therapists) be considered in the playground design process (14, 52). Such professionals possess the medical knowledge about different disabilities as well as the knowledge about activities that occur within a playground and what supports the children might need. Of all our recommendations, end user involvement in the design process should be considered a vital first step toward ensuring the end result is a more inclusive play space. Such involvement should go beyond simply soliciting families' playground experiences; rather, they should be encouraged to provide critical perspectives on proposed designs and how they can be improved using child-friendly practices; in other words, ask them what they want, and what they need (e.g., drawings) (63).

Our recommendation related to staffing and supervision highlights the interaction between the built environment and social inclusion. Many physical aspects of playground design, such as swings, sand, and elevated play components, assume children with disabilities will have caregiver support to aid with transfers. Yet, there tends to be an overall lack of support available at playgrounds for children with disabilities (40). Trained staff can play a critical role in filling this void through providing children with disabilities with several types of support as well as facilitating social interactions among children with and without disabilities alike. For example, trained staff can provide physical or instructional assistance to use equipment (9, 14, 24, 48), while also helping to initiate social interactions with other children through playground play (55, 56). Our recommendation for playground staffing and supervision is focused on playground programming where trained staff are knowledgeable about strategies to foster inclusive play and are cognizant of where and how liability issues can be properly addressed. Research is needed to understand how best to train and support staff, according to the play needs and preferences of children with and without disabilities, to deliver quality supervised programming that does not exclude smaller playgrounds in neighborhoods that could be very important for inclusion with peers.

While this study represents a particularly comprehensive synthesis of the peer-reviewed literature examining playground design for children with disabilities, it is not without limitations. First, studies were only included if they were published in 1990 or later. While we recognize our search was not exhaustive in that it did not capture earlier research [e.g., (64, 65)], findings prior to 1990—the year in which the ADA became law—may not reflect how this monumental change in regulations may have impacted playground design, in the United States at least. Second, we only included published studies with empirical evidence and as a result, academic literature involving clinical suggestions [e.g., for adapted swing design (66)] and unpublished work (e.g., white papers) were excluded. As such, our findings do not represent all of the available literature that may have implications for the design of playgrounds for children with disabilities. Third, our search was restricted to studies published in English, meaning that we most likely did not capture some of the inclusive playground design literature from scholars publishing in other languages, or in other parts of the world. As a result, our recommendations are largely informed by findings from research conducted in high-income countries. Fourth, very few of the included studies incorporated disability theory [e.g., social model of disability; (50, 55)] in their analyses, which limited our ability to connect our identified recommendations to the theoretical disability perspectives that may have produced them. Fifth, our scoping review was not prospectively registered in a database. Registration can help to avoid unplanned duplication and allow for comparison of reported review methods against what was planned in the protocol. Lastly, consistent with the scoping review methodology, the quality of the included studies was not appraised. While each of the included studies went through the peer review process, we recognize that there may be bias in our findings.

Overall, 13 evidence-informed recommendations and one promising practice for designing inclusive playgrounds for children with disabilities emerged from our scoping review of 35 peer-reviewed studies. Our evidence-informed recommendations are not exclusive to the playground design itself; they also recognize the importance of ensuring easy access into playground spaces, how the presence of trained staff within playgrounds may improve accessibility and inclusion, and how involving children with disabilities and their families in playground design processes can help with producing playgrounds that account for their needs, abilities, and desires. Building playgrounds that include families with children with disabilities in the design process will help to create play spaces in the community that are welcoming and inclusive for all. We anticipate this, along with exploring the role of trained staff will help communities better support all members with disabilities, thus enabling children and parents to experience play and the associated benefits of health and well-being.

AUTHOR CONTRIBUTIONS

DB: methodology, investigation, data curation, formal analysis, writing–original draft, and project administration. TR: methodology, investigation, and writing–original draft. JL: methodology, investigation, validation, and writing–original draft. RB: funding acquisition and writing–original draft. CS and AL-C: funding acquisition and writing–review and editing. KA-N: conceptualization, funding acquisition, investigation, validation, writing–original draft, and supervision. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

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Motivation for Rehabilitation in Patients With Subacute Stroke: A Qualitative Study

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Background: Motivation is essential for patients with subacute stroke undergoing intensive rehabilitation. Although it is known that motivation induces behavioral changes toward rehabilitation, detailed description has been lacking. Motivation can be intrinsic or extrinsic; however, it is unclear which type of factors mainly motivates patients' daily rehabilitation.

Purpose: This study aimed to examine the factors influencing patients' motivation and to explore the behavioral changes induced by motivation, especially age-related differences.

Method: Twenty participants (mean age 65.8 years [standard deviation 13.7]) who had a subacute stroke and underwent rehabilitation at a convalescent hospital were recruited using convenience sampling. Semi-structured interviews were conducted by an occupational therapist with an interview topic guide regarding factors influencing motivation and how it affects behavioral change. Interviews were recorded, transcribed to text, and analyzed by three occupational therapists using thematic analysis. The participants were divided into two groups: aged patients (aged $\geq$ 65 years) and middle-aged patients (aged $<$ 65 years), and data were analyzed according to the groups. This study was conducted according to the consolidated criteria for reporting qualitative research.

Results: Seven core categories were identified as factors influencing patients' motivation: patients' goals, experiences of success and failure, physical condition and cognitive function, resilience, influence of rehabilitation professionals, relationships between patients, and patients' supporters. The first four and last three core categories were further classified as personal and social-relationship factors, respectively. The categories related to intrinsic motivation such as enjoyment of rehabilitation itself were not derived. In both age-groups, motivation affected the frequency of self-training and activity in daily lives. In some aged patients, however, high motivation restrained their self-training to conserve their physical strength for rehabilitation by professionals. Some aged patients do not express their high motivation through their facial expressions and conversations compared to middle-aged patients; therefore, motivation is not always observable in aged patients.

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Conclusions: Interventions tailored to extrinsic factors are important for maintaining patients' motivation. Observational evaluation may lead to mislabeling of their motivation, especially for aged patients. Rehabilitation professionals should use validated evaluation scales or patients' narratives to assess patients' motivation.

Keywords: motivation, stroke, rehabilitation, qualitative study, age

INTRODUCTION

Stroke patients undergoing rehabilitation are required to be active participants in their treatment, and motivation is an essential factor for active participation. Self-determination theory (SDT) is a commonly known motivational theory (1, 2) and is applied in rehabilitation fields (3, 4). Conventionally, motivation is divided into extrinsic motivation and intrinsic motivation. Extrinsic motivation involves doing something because it leads to a separable consequence; that is, the goal is separate from the activity itself. In contrast, intrinsic motivation involves doing something because it is interesting and enjoyable in itself (1, 2). According to this classification, rewards such as functional improvement and praise from medical staff and family members can be categorized as extrinsic motivation, and enjoyment of rehabilitation itself can be considered intrinsic motivation. In previous studies, motivation for rehabilitation was reported to be affected by extrinsic factors such as environmental factors (5–7), individual beliefs (6–10), and support from medical staff and family (7, 10–13). Since the majority of stroke patients in the convalescent phase attend rehabilitation to return to their before-stroke-onset lives, a form of extrinsic motivation, we do not expect that intrinsic motivation is a major part of their commitments. Especially subacute stroke patients who are in convalescent phase are considered to be more affected by extrinsic motivation than chronic patients, and since their physical functions are relatively more changeable than that of chronic stroke patients, it is easier for them to earn rewards, i.e., functional recovery. Thus, we hypothesize that subacute stroke patients undergo rehabilitation based mostly on extrinsic factors such as functional improvement.

Motivation can induce behavioral changes. In rehabilitation, motivation is suggested as a predictor of functional outcomes (14). The motivation for rehabilitation is reported to influence physical activity (8, 15) and participation in training (16, 17). Thus, motivation can be expressed as physical activity and attitude toward rehabilitation. Therefore, medical staff tend to label the patients' motivation based on the activity in their daily lives, their attitude, and compliance with rehabilitation (7). However, a previous qualitative study pointed out a mismatch between motivation and activity and warned that observational evaluation of patients' motivation by medical staff may sometimes lead to mislabeling (7). Stroke patients in convalescent rehabilitation hospitals are required to pursue intensive rehabilitation training. Since the physical strength and capacity of aged patients tend to be less than that of younger patients (18), they may not have enough capacity to actively engage in physical activities or express a positive attitude toward rehabilitation. If medical staff label patients' motivation according to daily activity or attitude, those patients are mislabeled as being unmotivated. To correctly evaluate these patients, a detailed description of behavioral change, especially, age-related differences in their behavior, is required.

The purpose of this study is as follows: first, we clarified the factors influencing patients' motivation for rehabilitation in the convalescent rehabilitation hospital. In particular, this study aimed to examine which type of factors, extrinsic or intrinsic, mainly motivates patients' daily rehabilitation. Second, we hypothesized that the behavioral changes influenced by motivation may differ in aged patients and middle-aged patients; thus, we analyzed the behavioral changes among different age-groups.

Although several evaluation scales of motivation for rehabilitation have been reported (19–22), no validated evaluation scale for rehabilitation explicitly contains items related to intrinsic motivation. Furthermore, questionnaires cannot provide detailed descriptions about the patients' behavioral changes in daily life. Thus, we adopted a qualitative method. Qualitative methods are appropriate for investigating topics that are poorly understood (23). Semi-structured interviews are based around open-ended questions that define the area to be explored; then, the interviewer and interviewee pursue an idea in more detail (23). In order to investigate the factors influencing motivation and patients' behavioral changes in detail, it is necessary to understand patients' narrative. Therefore, we employed semi-structured interviews to elucidate participants' motivation for rehabilitation. We also adopted thematic analysis, since it provides a more detailed and nuanced account of themes within the data (24, 25).

MATERIALS AND METHODS

This study was conducted according to the consolidated criteria for reporting qualitative research [COREQ; (26)]. The study protocol was approved by the Ethics Committee of Tokyo Bay Rehabilitation Hospital (No. 144) and the Ethics Committee of Waseda University (No. 2019-059). All participants provided written and verbal informed consent before participating in this study.

Sample

Participants were recruited from those who were admitted to the Tokyo Bay Rehabilitation Hospital in Japan, which had three convalescent rehabilitation wards with 160 beds. The inclusion criteria were as follows: a Mini-Mental State Examination score of 24 or more and having had their first-ever hemiparetic stroke in the subacute phase (27). Patients who were diagnosed by their doctors as having aphasia, depression, or cognitive impairments, and therefore would have difficulty participating in the study, were excluded. Twenty-two Japanese patients who met the inclusion criteria were recruited through convenience sampling; of these, 20 agreed to participate (10 aged < 65 years). All participants were assessed with the Functional Independence Measure (FIM) (28) by trained nurses to evaluate their functional status. Sample characteristics are presented in **Table 1**.

Study Setting

This study was conducted at convalescent rehabilitation wards called Kaifukuki Rehabilitation Wards (KRWs). KRW is the system for subacute rehabilitation in Japan and is covered by governmental medical insurance. In KRW, patients undergo one-on-one rehabilitation with therapists up to 3 h every day. A typical schedule was 1 h in the morning and 1 or 2 h in the afternoon. Patients engage in self-training out of rehabilitation sessions if indicated. The content of the self-training is usually supervised by the therapist or doctor in charge. The participants can usually adjust the frequency and the intensity of self-training based on their own conditions and schedules. Interviews were conducted only once during the hospitalization and did not interrupt the participants' schedule of rehabilitation, meals, bathing, self-training, family visits, and meetings with doctors. It was mainly conducted in the afternoon between rehabilitation sessions and dinner. Interviews took place in the rehabilitation room or in the patient's room, where the content of the interview could not be overheard by others.

Interviews

The interviewers were three occupational therapists (TY, MK, and SK) at the hospital with a mean clinical experience of 6.7 ± 1.5 years. They had no personal relationships with the interviewees. All the interview contents were recorded using an IC recorder with pre-obtained permission from the interviewees. The interview topic guide (**Table 2**) was developed based on discussions between the three occupational therapists (TY, MK, and SK) and a rehabilitation doctor (YO) with more than 20 years of clinical experience. All interviews were performed according to the interview topic guide. The term "motivation" in the study was defined as "the impetus to initiate, sustain, orientate, enliven, and move forward a goal-oriented action," which was taken from the Japanese psychological dictionary (29). The definition was explained to participants prior to the interview to share a common understanding of the term. In addition, explanations for each question were prepared and shared among the interviewers; the meaning of the question was explained using these shared understandings prior to each question being asked and if the participant asked the interviewer about the meaning of the question. The interview was conducted by the occupational therapist who was not in charge of the patents' rehabilitation. Considering the psychological burden of the participants, the participants were told that the interview could be stopped immediately if a psychological burden arose during the interview.

Analyses

Recorded interview data were transcribed to text format and analyzed using thematic analysis (24, 25). The transcripts were read thoroughly to acquire meanings from the words of each sample. Close readings of transcripts generated sentences as the record units. Record units of each interview were categorized into codes by their meanings. The codes were then categorized into subcategories. These subcategories were then categorized into categories, which were further categorized into core categories. An occupational therapist (TY) made record units and codes, and three occupational therapists (TY, MK, and SK) made subcategories, categories, and core categories through repeated discussion until a consensus was reached. The personally identifiable data were not included in data analysis to preserve anonymity. These procedures were conducted separately for samples of participants aged < 65 and ≥ 65 years to consider age-group differences, since lifestyles may differ between these age-groups. Comparisons of the characteristics including demographic variables and FIM scores were performed between aged and middle-aged groups with Mann–Whitney *U*-test and chi-square test depending on the types of variables. Statistical analyses were performed with IBM SPSS Statistics 27 (IBM Corp., Armonk, NY). $P < 0.05$ were considered statistically significant.

RESULTS

Comparisons of Sample Characteristics Between Age-Groups

No significant differences were found in characteristics between age-groups, except for age and working status before onset (**Table 1**). Total FIM score ranged from 96 to 115 and 111 to 123 in aged and middle-aged groups, respectively. The participants in both age-groups had relatively high functional status.

Codes and Categories

The average interview time for participants aged ≥ 65 was 19 min 46 s (*SD* ±193 s), while time for participants aged < 65 was 17 min 17 s (*SD* ±238 s). From interviews with participants aged < 65 years, 462 record units, 191 codes, 37 subcategories, 21 categories, and eight core categories were derived. From interviews with participants aged ≥ 65 years, 490 record units, 176 codes, 36 subcategories, 19 categories, and eight core categories were derived (**Tables 3–5**). Although some differences between the two age-groups were found in some categories and subcategories, the same eight core categories were derived for both age-groups. Among these eight core categories, seven included factors influencing motivation for rehabilitation and one category concerned the effects of motivation on patients' behaviors (**Tables 3–5**).

Factors Influencing Motivation for Rehabilitation

The seven core categories for influencing factors of motivation for rehabilitation were patients' goals, experiences of success and failure, physical condition and cognitive function, resilience, influence of rehabilitation professionals, relationships between

TABLE 1 | Sample characteristics.

	Aged < 65 years (*n* = 10)	Aged ≥ 65 years (*n* = 10)	*P*-value
Gender, male/female, *n*	7/3	6/4	0.639
Age, mean (standard deviation)	53.5 (6.8)	78.0 (4.0)	<0.001†
Side of paresis (right/left/bilateral), *n*	5/5/0	4/5/1	0.574
Days from stroke onset to interview, mean (standard deviation)	84.7 (36.3)	74.5 (25.0)	0.597
Days from admission to interview, mean (standard deviation)	53.3 (31.0)	46.7 (24.4)	0.705
Total hospitalization days, mean (standard deviation)	86.9 (46.5)	86.9 (46.5)	0.472
Number of family members living together, median (interquartile range)	1 (0–2)	1 (1–1)	0.386
Functional Independence Measure total score, median (interquartile range)	117 (111–123)	109 (96–115)	0.088
Functional Independence Measure motor score, median (interquartile range)	83 (82–88)	80 (70–82)	0.103
Mini-Mental State Examination, median (interquartile range)	29 (29,30)	28 (28,29)	0.137
Living place before onset, home/not at home	10/0	10/0	
Working status before stroke, yes/no	9/1	3/7	0.006†

†*Significant difference between group aged < 65 years and aged ≥ 65 years.*

TABLE 2 | Interview topic guide.

1. When does your motivation for rehabilitation become high (low)?
2. What keeps your motivation high?
3. What makes your motivation low?
4. When your motivation becomes high (low), how does your behavior change?
5. What is your motivation for rehabilitation?

patients, and patients' supporters. The former four core categories were classified as personal factors (**Table 3**), and the latter three core categories were classified as social relationship factors (**Table 4**).

Personal Factors

Patients' Goals

Regardless of age-group, participants mentioned that their rehabilitation goal regarding individual goals of activities and functions, role as a family member, social participation such as work and hobby, and discharge to home influenced their motivation. In addition, one participant (aged < 65 years) mentioned the discrepancy between expected goal and reality. A participant (aged ≥ 65 years) noted that acquiring independence to reduce the burden on his family was a key motivation. A participant (aged < 65 years) mentioned being motivated to interact with friends, which was included in the "social participation" category.

Experiences of Success and Failure

Recovery from impairments and disabilities was reported as experiences of successes. A participant (aged ≥ 65 years) reported that the ability to have an expanded range of activities at the hospital was a motivator. The discrepancy between movement that participants expected and real task performance was reported as a failure, which decreased patients' motivation. Some participants (aged < 65 years) mentioned that recovery stagnation and repeated failures decreased their motivation.

Physical Condition and Cognitive Function

Participants in both age-groups commented that physical conditions including fatigue, physical weakness, and pain influenced their motivation. Some participants (aged ≥ 65 years) reported that it was vital that they have cognitive function to understand the purpose of rehabilitation, and some participants (aged < 65 years) reported that worsening numbness influenced their motivation.

Resilience

"Resilience" refers to the ability to adjust and adapt to varied situations and overcome challenges after adversity, although no universal definition exists (30). Participants in both age-groups reported that high resiliency played a key role in maintaining their motivation.

Social Relationship Factors

Influence of Rehabilitation Professionals

Relationships with professionals on the rehabilitation team—including therapists, nurses, and medical doctors—as well as exercise methods were classified into this core category.

Regardless of participants' age, they mentioned that professional positive feedback from therapists and nurses, reliance on therapists and nurses, and therapists' positive attitudes toward patients were motivating factors. Some participants (aged < 65 years) did not feel influenced by therapists' words, regardless of what they said; and some participants (aged ≥ 65 years) seemed to be motivated by their own beliefs—that they wanted to improve their body functions for the sake of their therapists. For some participants, doctors' explanations about medical conditions and prognosis also influenced their motivation. The explanation by a medical doctor that a patient could have sequelae and the percentage of improvement was lower than patient's expected reduced patients' motivation.

The variation and appropriateness of the exercises, sharing the goal setting, and understanding the purpose of the exercise positively influenced patients' motivation. Some advices and

TABLE 3 | Categories of personal factors derived from the thematic analysis.

Core category	Category	Subcategory	Patients' narratives
Patients' goals	Individual goals of activities and functions	Achievement goals pertaining to activities	"My goal is to recover my physical condition as soon as possible to get back to the same daily routine as before. Because of this, I can expend efforts on daily rehabilitation" (aged $\geq$ 65 years, Male 2).
		Goals for improvement of function and activity	"I want to be able to control my body freely again. Toward this end, I work hard to continue my rehabilitation" (aged $<$ 65 years, Male 5).
	Role as a family member	Role as a housewife	"I hope that I will return home and regain my role as a housewife after discharge. When thinking about this goal, I understand what is necessary for me to do now" (aged $<$ 65 years, Female 2).
		Role as a supporting family member	"Preparing meals has been my role. So, I am thinking now that I want to continue this role after discharged home" (aged $\geq$ 65 years, Female 1).
		Hope to acquire independence to reduce the burden on one's family[‡]	"If my walk is still unstable when I get discharged from hospital to home, it may be burdensome for my family. I don't want to bother my family" (aged $\geq$ 65 years, Male 7).
	Social participation	Hope to return to work	"My goal is returning to work. When I am thinking about that goal, I feel motivated and think that I should put much effort into my rehabilitation" (aged $<$ 65 years, Male 7). "My goal is to return to my previous work. That is my motivation for rehabilitation" (aged $\geq$ 65 years, Male 5).
		Hobby	"I really like to play golf. I have to do training [to] play that" (aged $\geq$ 65 years, Male 3).
		Interaction with friends[†]	"I love to drink with friends. So, I have to be able to pick up a beer mug with my hand. To go to drink with friends again, I know that I should do rehabilitation" (aged $<$ 65 years, Male 1).
	Discharge to home		"I want to go home and get back to my previous life as soon as possible. So, I should do what I have to do" (aged $<$ 65 years, Male 6).
	Discrepancy between expected goal and reality[†]		"I had expected that I would have been able to walk without a cane by this time and would be preparing for the discharge by now. But the reality is not what I had expected; I am still using a wheelchair. The discrepancy between the hospitalization length I expected and reality makes me lose motivation" (aged $<$ 65 years, Male 3).
Experiences of success and failure	Success experiences	Recovery from impairments and disability	"Motivation will be very high when I am able to do that [which] I couldn't do 1 week ago" (aged $<$ 65 years, Male 3).
		Expansion of permitted range of activities in the hospital[‡]	"I want to expand my activity space. Then, I could go to a shop in the hospital, for example" (aged $\geq$ 65 years, Male 7).
	Failure experiences	Discrepancy between expectations and real performance of the tasks	"Sometimes I became disappointed that I couldn't do an easy task that I could do before" (aged $\geq$ 65 years, Male 3).
		Stagnation of the recovery[†]	"I was depressed when my movement was worse than it was yesterday" (aged $<$ 65 years, Female 4).
		Repetitions of failure experiences[†]	"When I fail something repeatedly, [my] motivation [decreases]" (aged $<$ 65 years, Male 6).
Physical condition and cognitive function	Physical condition	Fatigue	"I make an effort to do more when I have more physical strength; but I want to lie down when I am tired" (aged $\geq$ 65 years, Female 1).
		Physical weakness	"I lose weight when I can't eat my meals. Then, my physical strength decreases. In such cases, I don't feel like participating in rehabilitation" (aged $\geq$ 65 years, Female 1).
		Pain	"Motivation decreases when I can't do any daily rehabilitation training due to pain" (aged $<$ 65 years, Male 3). "I'm worried that hard training may cause bad physical conditions such as muscle pain" (aged $<$ 65 years, Male 1).
		Worsening of numbness[†]	"Some changes such as pain or numbness in my body reduced my motivation" (aged $<$ 65 years, Male 9).
	Cognitive function[‡]		"I am here to do rehabilitation. We need to recognize why we are in this hospital. If I cannot understand the purpose of my training because of dementia, I cannot keep [my] motivation high" (aged $\geq$ 65 years, Female 10).
Resilience			"I recognize my disability. I need to live with this disability" (aged $<$ 65 years, Male 8). "When a doctor said to me that I could have sequelae, I realized that I must make a tremendous effort for my own rehabilitation. I will be in trouble if I can't walk" (aged $\geq$ 65 years, Female 9).

[†] *Only for participants aged $<$ 65 years;* [‡] *Only for participants aged $\geq$ 65 years.*

TABLE 4 | Categories of social relationship factors derived from the thematic analysis.

Core category	Category	Subcategory	Patients' narratives
Influence of rehabilitation professionals	Relationships with therapists	Feedback from therapists	"In my opinion, whether or not there is a certain improvement for me is very vague. When the therapists give me positive feedback from their professional viewpoints, it makes me realize that I have improved somehow" (aged ≥ 65 years, Female 4).
		Reliance on therapists	"I trust therapists and medical staffs with my rehabilitation. These good relationships could increase my motivation" (aged ≥ 65 years, Male 7).
		Therapists' attitude toward patients	"My motivation for rehabilitation decreased when my therapists repeatedly pointed out my mistakes, which made me feel that I could not do anything" (aged < 65 years, Male 6). "When I wanted to engage in self-exercise for my weak points, the therapist kindly devised a regimen for me. Since then, I trusted the therapist more, and my motivation for rehabilitation has increased significantly" (aged ≥ 65 years, Male 3).
		Not influenced by phrases of praise[†]	"Sometimes, I would doubt the therapists' positive feedbacks. I was pleased to hear some of those praises; but I could not detect any truth in them" (aged < 65 years, Male 1).
		To do training for the sake of therapists[‡]	"My therapist has made such a great effort for me. I wanted therapists to feel that I was worth teaching. I think I need to rehearse what I have learned and engage in self-training" (aged ≥ 65 years, Male 2).
	Relationship with nurses	Feedback from nurses	"I think it is very important for me to receive positive comments from nurses such as 'you have been improving and I know you are working hard on your rehabilitation"' (aged < 65 years, Female 10).
		Reliance on nurses	"I was very pleased when some nurses mentioned my improvement. That made me feel like they recognized my efforts" (aged < 65 years, female 10). "Some nurses are restless and that makes me hate them sometimes. I feel I am losing my motivation for rehabilitation when I am with them" (aged ≥ 65 years, Male 6).
	Relationship with medical doctors		"When a medical doctor in the acute hospital said that some impairments might remain after discharge, I was disappointed. But when another doctor in the same hospital said that they will make me improve within 2 months, that made me so motivated" (aged ≥ 65 years, Female 9).
	Exercise methods	Varied contents	"Exercises provided by my therapist are very good, of course, while other therapists would make me aware that other ideas for rehabilitation are also good" (aged < 65 years, Male 8).
		Shared goal setting and understanding of the purpose of the exercise	"I realized that becoming aware of the aim of the exercise is very important for increasing my motivation" (aged < 65 years, Male 9). "If goals are shared with therapists, motivation can be maintained even during hard training." (aged ≥ 65 years, Male 8).
		Appropriateness of the exercise contents	"I feel frustrated if I am given the next level of exercise when I am yet to perfect the current level. I would be more motivated when the therapist clarifies my rehabilitation goals step-by-step and changes the exercise method accordingly" (aged < 65 years, Female 2). "In daily rehabilitation, therapists provide feedback on my weak points, which I also recognize, and teach me specific exercise methods to improve, which makes me motivated continuously" (aged ≥ 65 years, Male 5).
Relationship between patients	Observation of other patients	Observation of other patients' efforts	"Just by watching others making a big effort motivates me. The important thing is that he is making his best effort by his own way, which has nothing to do with the severity of the disability" (aged ≥ 65 years, Male 3).
		Observation of aged patients' efforts[†]	"When I see the effort of the aged patient, I feel that I should be making more effort on my own rehabilitation" (aged < 65 years, Male 9).
		Comparison with other patients' impairments and disabilities[‡]	"Sometimes I compare my abilities or differences to other patients. If other patients can do some movement, I want to be able to do those" (aged ≥ 65 years, Female 10).
		No influences from other patients	"All I can do is do my best. Other patients have no relationship to me" (aged ≥ 65 years, Female 9).
	Communication with other patients	Interaction with other patients	"An interaction with other patients is very important for me. It makes me feel like having a mutual encouragement" (aged < 65 years, Female 2).
		Conflicts with patients[‡]	"My motivation decreases when I have trouble in relationships with other patients" (aged ≥ 65 years, Female 4).
Patients' supporters	Existence of supporters	Family	"My family work so hard for me, so I must not complain about anything. I want to give back to them" (aged ≥ 65 years, Female 1).
		Coworkers[†]	"Some of my coworkers say they will wait for me to return to work. This is such a great motivation" (aged < 65 years, Male 9).
		Friends[‡]	"When my friends visit me, I really appreciate that. If no one comes, I will be depressed" (aged ≥ 65 years, Female 10).

(Continued)

TABLE 4 | Continued

Core category	Category	Subcategory	Patients' narratives
	Relationship with supporters	Conversation with family	"My family member said to me that they were happy to see my abilities recovered, which makes me happy" (aged ≥ 65 years, Female 9).
		Encouragement†	"Keeping a high motivation is very tough. An encouragement from my friend helps me to maintain high motivation" (aged < 65 years, Female 10).
		Family visit‡	"When my family visits me, I get the feeling that I want to be discharged from here as soon as I improve significantly enough" (aged ≥ 65 years, Male 2).

†Only for participants aged < 65 years; ‡Only for participants aged ≥ 65 years.

TABLE 5 | Categories of effects on patients' behavior derived from the thematic analysis.

Core category	Category	Subcategory	Patients' narratives
Patients' behavior changes	Self-training	Extent of self-training	"When my motivation is high, I will walk through a corridor for self-training" (aged ≥ 65 years, Male 7).
		Cancelation of self-training for rehabilitation with therapists‡	"I never did self-training. If I did, I would be too tired to do rehabilitation with therapists" (aged ≥ 65 years, Female 1).
	Attitude	Attitude toward rehabilitation	"When my motivation is high, I demand my therapists to let me do harder training" (aged < 65 years, Male 6).
		Total time spent in a room	"I want to go shopping at the hospital store. When I have high motivation, I will not stay in my room" (aged < 65 years, Female 10).
		Attitude toward daily activities‡	"When my condition is good, I want to do what I can do in daily activities" (aged ≥ 65 years, Male 7).
	Expressions†	Facial expression	"I could smile when my motivation was high" (aged < 65 years, Female 2).
		Quality of expression	"When my motivation was high, I behaved cheerfully" (aged < 65 years, Female 2).
	Talking with other patients†		"I talked to others a lot when my motivation was high" (aged < 65 years, Female 10).
	No particular changes		"Motivation does not affect my activities. I just do what the therapist teaches me" (aged ≥ 65 years, Female 4).

†Only for participants aged < 65 years; ‡Only for participants aged ≥ 65 years.

viewpoints for rehabilitation not only from the therapist in charge but also from other therapists could positively influence patients' motivation. Furthermore, some participants commented that motivation was positively influenced by the appropriateness of exercise contents, including adjusting the difficulty of the exercises, setting a goal through shared decision-making step-by-step, and changing the exercise regimen appropriately.

Relationships Between Patients

Observations of other patients' efforts influenced some participants' motivation, regardless of the severity of other patients' disabilities. Motivation was greatly enhanced by observing other patients completing their training; however, some participants were not influenced by other patients. Some participants (aged < 65 years) reported that observing aged patients' efforts was influential, while some participants (aged ≥ 65 years) reported that comparing their abilities to those of other patients had a positive influence on their motivation. Motivation increased when mutual encouragement was obtained. Some participants (aged ≥ 65 years) reported that motivation decreased owing to the frustrations from the conflicts with other patients.

Patients' Supporters

Existence of supporters and their relationships devised this core category. Regardless of age-group, participants reported that having a family and the conversations with them influenced their motivation. Regarding the existence of supporters, participants in both age-groups commented about their coworkers and friends. One participant (aged < 65 years) talked about getting encouragement from her supporters, and some participants (aged ≥ 65 years) reported the influence of family visits.

Patients' Behavioral Change

The interviews revealed that patients' motivation had an effect on their behaviors in the hospital, although some participants reported that motivation had no influence on their behaviors (**Table 5**). Some participants reported on self-training and active participation for daily activities. Some other participants (aged $\geq$ 65 years) reported that they did not do self-training that made them tired when their motivation for rehabilitation was high because they wanted to display better performance during rehabilitation with therapists. For some participants (aged $<$ 65 years), the frequency of communication with other patients and changes in facial expressions were also reported.

DISCUSSION

This study investigated the factors influencing subacute stoke patients' motivation for rehabilitation and the behavioral changes induced by motivation at a convalescent rehabilitation hospital. The strength of the study was that it comprehensively elucidated the factors influencing patients' motivation for rehabilitation with the qualified study methodology according to COREQ. In addition, this study revealed that behavioral changes due to motivation differ depending on patients' age. Seven factors influencing motivation were identified, all of which were categorized as extrinsic motivation. No factor included a purely intrinsic description such as enjoyment of rehabilitation itself. Regarding behavioral change, we found that high motivation does not always enhance activity in aged stroke patients.

As for the core categories influencing patients' motivation, four personal factors (patients' goals, experiences of success and failure, physical condition and cognitive function, and resilience) and three social relationship factors (influence of rehabilitation professionals, relationship between patients, and patients' supporters) were derived. While all these core categories are classified as extrinsic motivation, a category such as joy for rehabilitation itself (intrinsic motivation) was not derived from patients' narratives. The same core categories were derived from both aged and middle-aged patients. Therefore, these core categories will affect the majority of stroke patients' motivation for rehabilitation. Previous studies have shown that the motivation for rehabilitation in patients who have had strokes or various other diseases is influenced by individual traits (8, 9) and social environmental and social relationship factors (5–7, 10–13). However, these studies have not discussed the existence of intrinsic motivation for rehabilitation. Our results indicated that motivation for rehabilitation is mainly based on extrinsic, rather than intrinsic, motivation, at least for subacute stroke patients in convalescent rehabilitation hospitals.

The results indicate that, unlike sports or studies where internal motivation is vital, it is relatively effective to intervene with extrinsic motivation (1, 2), rather than intrinsic motivation, in the rehabilitation fields. Some of the personal and social relationship factors elucidated in this study are modifiable in rehabilitation practice, such as patients' goal setting and relationships with medical staff. Our results will be useful to help optimize rehabilitation practice and planning.

If the goal is set too high, the rewards the patients receive, such as functional improvement, will be less than what they expected. Such reward prediction errors could reduce patients' motivation to reach their goals (31). To maintain stroke patients' motivation, it is important to set appropriate goals and provide a variety of training content that can be tailored according to the patient's situation, such as age.

Previous studies pointed out the importance of relationships with others. For example, overprotection from medical staff and family makes patients incapable (9, 32), while appropriate information from rehabilitation professionals helps maintain patients' motivation (9). Our results are consistent with these studies and demonstrate the importance of social relationships, such as communication with medical staff, other patients, and family for maintaining the motivation for rehabilitation.

A previous qualitative study targeting medical professionals reported that motivation is affected by both physical and human environments, such as a well-maintained room and group treatment sessions (7). In our study, however, only human environmental factors such as medical staff, other patients, and supporters were derived, not physical environmental factors. The participants of this study were all patients who were admitted to the convalescent rehabilitation hospital for the first time and were not able to compare it with other environments, which could be the reason physical environmental factors were not derived.

Although no differences in the core categories were found based on patients' age-groups, some differences appeared in subcategories. In the middle-aged patients, factors associated with improvement in physical function were derived, such as stagnation of recovery, repetitions of failure experiences, and discrepancy between expectations and reality. In contrast, among the aged patients, the subcategory associated with self-care was derived, such as hoping to acquire independence to reduce the burden on their family. Younger participants with disability may have higher expectations of what they can achieve compared to individuals in other age-groups (33). During inpatient rehabilitation, aged patients' main goals were regaining independence in self-care activities and going home (34). In other words, factors related to physical function and its disability may affect motivation among middle-aged patients, while factors related to independent self-care and its disability may affect motivation among aged patients.

Regarding the behavioral changes induced by motivation, the frequency of self-training, attitude toward rehabilitation, and total time spent in a room were derived regardless of the patient's age. The majority of stroke patients in the hospital engage in self-training, rehabilitation, and activities of daily living more actively when they are highly motivated. Some aged patients, however, intentionally did not participate in self-training when their motivation was high to conserve their physical strength. Some of them wanted to show better performance during rehabilitation with therapists, to live up to their expectations. Meanwhile, the category related to expression and frequency of communication with other patients was derived in only middle-aged patients. Therefore, motivation is not likely to be expressed as an observable behavior such as emotional expression and self-training in some aged patients. Medical professionals tend to label patients' motivation based on observable activities such as demeanor and their compliance with rehabilitation

(7). Our results indicate that if medical staff evaluate patients' motivation only from observational assessment, it may lead to mislabeling of motivation, especially in aged patients. Medical staff members need to understand that the differences in behavioral change are related to age. To assess patients' motivation for rehabilitation, it is desirable to evaluate their narrative or to use a validated evaluation scale for assessing patients' motivation.

There are several limitations in the study. Firstly, the sample size was relatively small, and the participants were enrolled with convenience sampling in a single institution. All the participants were Japanese and had relatively high functional level as measured with FIM. Since motivation may be influenced by the functional level as well as social factors, the generalizability of results obtained in this study should be done with caution. To examine the validity, a further study with a larger sample and participants with various functional levels using random sampling from multiple facilities in many countries should be conducted. Secondly, this study also involves the methodological limitations of qualitative studies. Although the study was conducted according to COREQ, the labeling could be different for another analyst. Some of the subcategories could be classified into multiple core categories. For example, hobby is classified as a subcategory in the category "social participation" but also could be allocated to the category "Individual goals of activities and functions." Thus, interpretation of classification needs to be considered given the analysts' characteristics. Lastly, we cannot eliminate the possible existence of intrinsic motivation because we did not ask the direct/specific questions about the existence of intrinsic motivation, e.g., whether the participants enjoy rehabilitation itself. Although it is difficult to prove the non-existence of intrinsic motivation, more in-depth interview research will help clarify this point.

The motivation of patients with subacute stroke in convalescent rehabilitation hospitals was mainly influenced by factors related to extrinsic motivation. The patients' goals, experiences of success and failure, physical condition and cognitive function, resilience, relationships with rehabilitation professionals, and family and supporters were derived as the core categories. It is important that rehabilitation professionals consider these factors when helping patients stay motivated. Regarding behavioral changes induced by motivation, some aged patients do not show motivation via activities observable by rehabilitation professionals. For accurate evaluation of patients' motivation, rehabilitation professionals should use not only observation but also validated evaluation scales or patients' narratives. The findings of this study can help facilitate rehabilitation professionals' understanding of patients' motivation, encourage motivation, and develop optimal treatment plans.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The protocol of this study was reviewed and approved by Ethic Committee of Tokyo Bay Rehabilitation Hospital and Ethic Committee of Waseda University. The patients provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

TY, YO, and JY contributed to the conception, design, and methodology of this study. TY, YO, MK, SK, and JY performed the formal analysis. YO, RO, and JY supervised this study. TY wrote the first draft of the manuscript. TY, YO, RO, and JY reviewed and edited the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.664758/full#supplementary-material

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BRIEF RESEARCH REPORT
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An Inclusive Early Childhood Intervention Program for Children With Disabilities: Possible Effects on Children and Nursery Teachers

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Inclusive early childhood intervention provides opportunities for children with disabilities to receive education with typically developing children. The present study examined the effects of the AI-AI STEP Program, which is designed to help nursery teachers learn the methods of inclusive early childhood intervention for children with disabilities. This study involved 37 managers of 37 nursery schools in Japan, 48 nursery teachers, and 48 children with disabilities. The school managers, who had previously learned about the program through a seminar we offered, provided the nursery teachers with guidance on the program. The guidance provided to the nursery teachers consisted of combined structured explanations with a manual and on-the-job training. The program was performed for 6 months, and changes in the children's development and behavior and the school nursery teachers' self-efficacy and state-trait anxiety, were examined before and after using the program. Multivariate analysis was used to assess factors that had an effect on the children's developmental gains through the program. The developmental quotient of children significantly improved. In addition, "emotional symptoms" and "peer problems" on the Strengths and Difficulties Questionnaire subscale markedly improved. The self-efficacy of nursery teachers significantly improved, and state anxiety decreased. There was a significant relationship between the improvement of the children's development quotient and a lower development quotient at baseline. The provision of inclusive early childhood intervention using the program promoted the children's development, and improved their behavior. Furthermore, it had a positive effect on the nursery teachers.

Keywords: inclusive early childhood intervention, children with disabilities, developmental quotient, self-efficacy, anxiety

INTRODUCTION

Early childhood intervention (ECI) is a provision of support for children with disabilities according to their developmental stages. ECI is one of a variety of evidences suggesting that early childhood intervention provides an important opportunity to alter children's development and children-parent interactions (1–3). Children with disabilities receiving education with typically developing children is an important aspect of inclusive early childhood intervention (IECI) (4–6). Communication among the children through daily life and play activities has a positive effect. Lindsay (7) and Rafferty et al. (8) reported that IECI promoted the language development of preschoolers with disabilities and improved their social adaptation ability. It also enhanced typically developing children's sociability and increased the numbers of their friends (9).

In Japan, IEI is defined as "education and care for infants with physical and/or intellectual disabilities provided in nursery schools." In 1974, the Ministry of Health and Welfare (currently, the Ministry of Health, Labor, and Welfare) launched a project for children with disabilities to receive education and care in nursery schools. It was an innovative attempt to incorporate nursery schools, in addition to rehabilitation centers, into Japan's IECI system for children with disabilities. Currently, 92.7% of all Japanese nursery schools provide IECI for such children (10). These schools are offered financial support to increase the number of nursery teachers or adopt other necessary measures; however, some nursery schools have insufficient financial resources, staff, and quality assurance.

IECI for children with disabilities in nursery schools is supported by experts through two types of service: on-site counseling and nursery school-visit services. The former is operated by municipalities that dispatch doctors, psychologists, or other experts to nursery schools, and provide guidance for nursery teachers. These experts directly advise nursery teachers about how to provide childcare to children with disabilities. In the latter, which started in April 2012, experts from the child developmental support centers visit nursery and elementary schools in communities to directly provide specialized support for children with disabilities.

The Japanese government has recently begun to incorporate IECI into its general policies for children with or without disabilities. In April 2015, it launched the Comprehensive Support System for Children and Childcare by three central government entities: the Cabinet Office; the Ministry of Education, Culture, Sports, Science and Technology; and the Ministry of Health, Labor and Welfare. Importance was attached to close collaboration to simultaneously support children with disabilities and their parents. It is necessary to further organize systems to provide IECI for children with disabilities in nursery schools, and improve their quality; however, there are limited assessments of IECI and evaluation of its impact on children in Japan.

Since 2004, the Aichi Children's Health and Medical Center has been training leaders of nursery teachers using the AI-AI STEP (Step TEaching Program) Program (11). The first "AI" represents the Japanese "word for love/ attachment" and the other "AI" is and abbreviation of "Aichi", the birthplace of the program. The program is designed to help nursery teachers learn the methods of IECI for target preschool children aged 2–6. Target children include those with suspected autism spectrum disorder, attention deficit hyperactivity disorder, learning disability, or other developmental disorders. When the program started, the recognition of developmental disorders was insufficient in Japan. In a large number of such cases, if the disorder had been identified and appropriately managed earlier, the situation may have been less stressful for both the child and the people around him/her. Many nursery teachers sought advice on individualized approaches from the experts of the Aichi Children's Health and Medical Center. As a result, the center began to provide opportunities for nursery teachers to learn about IECI, with the aim of enabling children with disability to perform group activities more comfortably at nursery schools, while enhancing nursery teachers' self-confidence in providing IECI. To date, more than 600 nursery teachers have been trained at the center using this program.

We examined the effects on target children and nursery teachers of the AI-AI STEP Program, and assessed factors that had an effect on the children's developmental gains through the program.

MATERIALS AND METHODS

Contents of the AI-AI STEP Program

The AI-AI STEP Program aims to support children's appropriate behaviors, focusing on their relationships with nursery teachers (11). The AI-AI STEP Program is a practice-based program based on behavioral science (12, 13) and social learning theory (14, 15). It is characterized by four points: (1) being useful for all nursery teachers; (2) being useful for all children, regardless of the characteristics of their disability; (3) facilitating practical verification; and (4) attaching importance to experience-based learning. The program integrates individualized and grouping approaches.

The individualized approaches correspond to a target child's three behavioral stages: (1) behaviors to be developed (to perform self-care), (2) behaviors to be adopted (to overcome difficulties or acquire new experiences), and (3) behaviors to be avoided (problematic or possibly problematic behaviors). The nursery teachers focus on these behaviors and select a behavior from stage 1 that they consider to be easier to correct. The process has six steps: (1) observing behaviors, (2) setting three support processes, (3) practicing (making commitments), (4) recording, (5) assessing (grading), and (6) summarizing. When the target child can perform the behavior properly, the nursery chooses a behavior from stage 2 and supports the child using the same six steps. A nursery teacher starts with an easier task (stage 1), and proceeds to the more difficult tasks (stage 2 or 3). These three stages and six steps enable nursery teachers to acquire and improve their objective observation, practical, and verification skills.

The grouping approaches are based on the idea that groups can be appropriately created by establishing favorable relationships between target children and nursery teachers, between target and other children, and between other children and nursery teachers, rather than by simply including target children in groups. Nursery teachers' behaviors are an exemplary for children at all times. The use of affirmations for target children by nursery teachers, such as praising them in front of other children, promotes the latter's affirmative behaviors toward the former. Such approaches are effective for other children to observe and develop appropriate attitudes toward the target children. They also promote affirmative behaviors and attitudes, such as 'praising', among all class members.

Targets and Study Design

The present study was a pre-post design quasi-experimental study. This study consisted of 37 managers of 37 nursery schools in the Aichi and Fukushima Prefectures, Japan; 48 nursery teachers; and 48 children with disabilities. The school managers, who had previously learned about the program through an intensive seminar we offered, provided the nursery teachers in charge of the children with disabilities with guidance on the program at their nursery schools. The managers' guidance consisted of combined structured explanations with a manual and on-the-job training. Subsequently, based on the program and with support from the school managers, the nursery teachers provided IECI for the children with disabilities for 6 months. Fidelity assessments of nursery teachers' IECI were conducted by managers using direct observation and reviews of the nursery teachers' self-reports that were completed in steps 4–6. Before and after IECI using the program, changes in the children's development and behaviors, as well as those in the school nursery teachers' self-efficacy and state-trait anxiety, were examined.

MEASURES

The children's development and behavior were assessed using the Kinder Infant Development Scale (KIDS) (16) and Strengths and Difficulties Questionnaire (SDQ) (17, 18), respectively, both of which are self-administered, registered questionnaires, which were filled out by the nursery teachers. The nursery teachers' self-efficacy and state-trait anxiety were evaluated using the General Self-Efficacy Scale (GSES) (19) and a new version of the State-Trait Anxiety Inventory (STAI) (20), respectively.

Kinder Infant Development Scale (KIDS)

KIDS is a parent/caregiver-rated questionnaire for developmental screening of children aged 1 month to 6 years and 11 months that has been used since 1989 all over Japan (16). There are four types of questionnaires for children of different ages. We used Type T, which is recommended for children aged 1 month to 6 years and 11 months with developmental delay. The questionnaire is composed of nine subscales with 282 items: Physical-Motor (37 items), Manipulation (37 items), Receptive Language (37 items), Expression Language (37 items), Language Concepts (25 items), Social Relationships with Children (25 items), Social Relationships with Adults (37 items), Discipline (25 items), and Feeding (22 items). For each item, the nursery teacher answered with a circle if the child could perform the behavior and a cross if the child could not. The raw score for the subscale was the number of circles, which were converted into developmental age (DA) by referring to a translation table in the KIDS manual. Development quota was calculated by dividing DA by the child's chronological age.

Strengths and Difficulties Questionnaire (SDQ)

We used the Japanese version of SDQ (21). The SDQ is a brief screening instrument to assess the positive and negative aspects of the behaviors of children and adolescents assessed by parents and teachers. The SDQ consists of five subscales with 25 items, four kinds of problem scores (emotional symptoms, conduct problems, hyperactivity/inattention, and peer problems), and a positive aspect of prosocial behavior. The SDQ had three item responses: 0 = not true, 1 = somewhat true, and 2 = certainly true. Higher scores on the four subscales reflect behavioral difficulties, whereas higher scores on the prosocial behavior subscale reflect strengths.

The General Self-Efficacy Scale (GSES)

The GSES was developed by Sakano and Tohjoh (19). It is an instrument for measuring an individual's strength of general self-efficacy across a variety of everyday life settings. Bandura (22), who originally proposed the concept, defined self-efficacy as a judgment of "how well-one can execute courses of action required to deal with prospective situations." The total score is calculated by the sum of all 16 items. We used standardized scores based on a conversion table indicating 50 average points and a standard deviation of 10. A higher score means more self-efficacy.

The State-Trait Anxiety Inventory (STAI)

STAI, which was developed by Spielberger et al. (20), is a psychological inventory based on a 4-point Likert scale and consists of 40 questions on a self-report basis. It measures two types of anxiety: state anxiety and trait anxiety. Each type of anxiety has its own scale of 20 different questions scored from 20 to 80. Higher scores indicate greater anxiety.

DATA ANALYSIS

Changes in these items after intervention with the AI-AI STEP Program were examined using a paired *t*-test. Furthermore, multivariate analysis was used to assess factors that had an effect on the children's developmental gains through the AI-AI STEP Program. The number of children was small; therefore, in the multivariate analysis, the children's and the nursery teachers' factors were selected based on correlation coefficients of associations between each factor and the children's developmental gains.

ETHICAL CONSIDERATION

We explained the study protocol to the school managers and the nursery teachers, then informed the parents of the children. The study was conducted with written informed consent from the school managers, the nursery teachers, and the parents of the children, and the approval of the ethics committee of the institution the first author belonged to (No. 599).

RESULTS

Table 1 shows the characteristics of the target children and nursery teachers. The mean ages of the school managers, nursery teachers, and children were 56, 37, and 5 years and 9 months, respectively. The number of children in each group was as follows: undiagnosed but suspected disorders (n = 26), autism

TABLE 1 | Characteristics of children and nursery teachers.

	***N* (%) or mean (SD)**	
Children ($N = 48$)		
Sex		
Male	37	(77.1)
Female	11	(22.9)
age (mean, SD)	53.8 months	(11.1)
two	4	(8.3)
three	9	(18.7)
four	20	(41.7)
five	12	(25.0)
six	3	(6.3)
diagnosis		
ASD	14	(29.2)
Suspicion of ASD	26	(54.1)
DS	2	(4.2)
ID	4	(8.3)
PD	2	(4.2)
Nursery teacher ($N = 48$)		
sex		
male	2	(4.2)
female	46	(95.8)
age	36.6	(9.7)
School Manager ($N = 37$)		
sex		
female	37	(100.0)
age	56.2	(3.4)

ASD, autistic spectrum disorder; DS, Down syndrome; ID, Intellectual disability; PD, physical disability.

spectrum disorder ($n = 10$), intellectual disorder ($n = 4$), Down syndrome ($n = 2$), and physical disabilities ($n = 2$).

Table 2 shows the pre-/post-program scores of the children's KIDS and SDQ, and the nursery teachers' GSES and STAI. After IECI using the AI-AI STEP Program, the developmental quotient significantly improved in the KIDS in all subscales. Furthermore, "emotional symptoms" and "peer problems" on the SDQ subscale markedly improved, regardless of the disorder. On comparing the female nursery teachers before and after the AI-AI STEP Program, GSES-standardized scores significantly improved, with decreases in scores related to state anxiety. The lower state anxiety of nursery teachers had a significant relationship with reduced anxiety (coefficient -0.49, $p < 0.0005$).

Improvement of the children's development quotient was significantly affected by a lower children's development quotient at baseline, the diagnosis of autism spectrum disorder, and higher state anxiety inventory at baseline of the children's nursery teacher (**Table 3**).

TABLE 2 | Pre/post scores of children's KIDS and SDQ, and nursery teacher's GSES and STAI.

	Pre-test scores		**Post-test scores**		
	Mean	**(S.E.)**	**Mean**	**(S.E.)**	***p*-value score**
Children					
Kinder infant development scale (KIDS)					
Physical-motor	37.4	(1.9)	43.5	(1.9)	<0.001
Manipulation	37.5	(2.8)	49.8	(2.8)	<0.001
Receptive language	37.6	(2.7)	45.5	(2.6)	<0.001
Expression language	32.7	(2.5)	39.0	(2.4)	<0.001
Language concepts	39.5	(2.6)	45.2	(2.3)	<0.001
Social relationships with children	33.7	(2.3)	39.9	(2.3)	<0.001
Social relationships with adults	28.8	(2.5)	33.8	(2.4)	0.02
Discipline	42.6	(2.2)	48.1	(2.3)	<0.001
Feeding	23.3	(1.6)	27.3	(1.2)	<0.001
Developmental quotient	63.6	(3.6)	70.0	(3.2)	0.01
Strengths and difficulties questionnaire (SDQ)					
prosocial behavior	2.3	(0.4)	2.5	(0.4)	0.24
hyperactivity/inattention	6.5	(0.4)	6.8	(0.3)	0.79
emotional symptoms	2.9	(0.3)	2.5	(0.3)	0.04
peer problems	5.0	(0.3)	4.4	(0.3)	0.04
conduct problems	2.6	(0.3)	2.8	(0.3)	0.80
total difficulties	16.9	(0.8)	16.4	(0.8)	0.20
Nursery teacher (N=48)					
General self-efficacy scale (GSES)					
standardized score	42.4	(1.3)	44.1	(1.3)	0.03
male ($N = 2$)	35.5	(4.5)	35.0	(1.0)	
female ($N = 46$)	42.7	(1.3)	44.5	(1.4)	0.03
State-trait anxiety inventory (STAI)					
State anxiety	46.2	(1.2)	42.3	(1.2)	0.001
Trait anxiety	46.6	(1.3)	45.1	(1.2)	0.059

TABLE 3 | Linear regression predicting child's development quotient gain by the program.

Variables	**Coefficient**	***p-value***	**95% Confidence interval**	
Child's development quotient at baseline	-0.39	<0.001	-0.56	-0.23
Total difficulties of strengths and difficulties questionnaire	0.41	0.29	-0.37	1.19
The diagnosis of autism spectrum disorder	13.5	0.02	2.2	24.8
State anxiety inventory at baseline	-0.63	0.02	-1.15	-1.01

DISCUSSION

The present study examined the effects of the AI-AI STEP Program to support IECI for children with disabilities on the children themselves and the nursery teachers in charge of them. The provision of IECI using the program increased the nursery teachers' self-efficacy and decreased anxiety. The nursery teachers had more confidence in their ability to address the needs of children with disabilities after the program. The program also promoted the children's development, and improved their behavior, in particular emotional symptoms and peer problems. The improvement of the children's development was larger for those children who had larger developmental delays or who had a diagnosis of autism spectrum disorder at baseline.

As the children's developmental index values increased even when adjusting for chronological age, it was confirmed that the program promoted children's development. Improvements in children's language development and social adaptation ability were also reported in some previous studies (8, 9). The present study adds to this research by comprehensively examining developmental changes using KIDS. However, because of the pre-post design of our quasi-experimental study, history effects, and the Hawthorne effect may have influenced our results. Further studies that include randomized controlled trials using a control group.

Among the nursery teachers, both state-related anxiety and general self-efficacy scores improved after starting to use the program. Most nursery teachers perceived providing IEI for children with disabilities to be challenging; therefore, the program may have reduced their anxiety through the learning of practical methods for education and care. In addition, the reduction in their state anxiety was more marked when their state anxiety was higher at baseline, suggesting that their anxiety was reduced by having school managers' support and realizing the positive effects of their own approaches on children. In a previous study that taught nursery teachers about health literacy (how to convey health-related messages to the target audience), we observed that participating teachers who used the learned skills continued to have confidence in conveying health messages even 1 year after the training (23). Both projects indicate that acquiring practical skills and their application in the field improve professional confidence.

This study has some limitations. First, the number of subjects was limited. Second, as the design of this study had a one-group pre-post design, it was difficult to clarify whether the effects had been achieved through intervention or as a result of child development. Furthermore, we were unable to ascertain whether the program directly influenced the nursery teachers' self-efficacy using their assessment results or whether other outside factors had an effect. Third, the children's outcomes may be overestimated because of differing perspectives among the teachers. Finally, the mechanism that led both the children and their teachers to change through the program remained unclear. The theoretical background and validation of the program were insufficient because it was a practice-based program. Thus, it may be necessary to use a control group in future studies or conduct randomized control studies with more detailed data collection from the both groups.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

AUTHOR CONTRIBUTIONS

KU, AG, TI, and YY: material preparation and data collection were performed. KU and AG: analysis was performed. KU: the first draft of the manuscript was written. All authors contributed to the study conception and design, commented on previous version of the manuscript, read, and approved the final manuscript.

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The Impact of Sub-maximal Exercise on Neuropathic Pain, Inflammation, and Affect Among Adults With Spinal Cord Injury: A Pilot Study

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Introduction: Persons with spinal cord injury (SCI) often report high levels of neuropathic pain (NP) and poor well-being, which may result from increased inflammation. This study examined the impact of sub-maximal aerobic exercise on NP, inflammation and psychological affect among adults with SCI.

Methods: Eight active adults with tetraplegia (*n*-4, AIS A-C) and paraplegia ($n = 4$, AIS A-C) performed 30-min of arm-crank aerobic exercise and reported their ratings of perceived exertion (RPE) each minute. Measures of NP, affect, and inflammatory cytokines (IL-6, IL-10, IL-1ra, TNF-α) were taken pre-(T_0), immediately post-(T_1), and 90-min post-exercise (T_2).

Results: NP decreased between T_0 and T_1 for tetraplegics (−60%, $d = 0.47$; CI = −0.32, 2.02) and paraplegics (−16%, $d = 0.15$; CI = −0.30, 0.90). Correlations between change in cytokines and change in NP were medium-to large for tetraplegics (*rs* ranged from −0.820 to 0.965) and paraplegics (*rs* ranged from −0.598 to 0.833). However, the pattern of correlations between change in cytokines and affect was inconsistent between groups. Lower baseline levels of IL-1ra predicted greater decreases in NP immediately post-exercise ($r = 0.83$, $p = 0.01$).

Conclusion: Sub-maximal exercise can positively impact NP for some persons with SCI. Further experimental research should identify the optimal exercise intensity to reduce NP for persons with SCI, in addition to understanding biomarkers which may predict changes in NP.

Clinical Trial Registration: www.ClinicalTrials.gov, identifier NCT03955523.

Keywords: neuropathic pain, exercise, disability, affect, inflammation

INTRODUCTION

Neuropathic pain is caused by a lesion or disease of the somatosensory nervous system (1) and commonly manifests as allodynia (pain resulting from a non-noxious stimulus) and hyperalgesia [heightened response from a noxious stimulus; (2)]. Approximately 75% of persons with spinal cord injury (SCI) experience neuropathic pain (3), with many reporting pain to be more debilitating than the injury itself. Individuals with SCI who experience greater pain also report more negative affect [instantaneous displeasure that lacks cognitive appraisal; (4)] and susceptibility to developing mood disorders, such as depression or anxiety (5).

Current treatment options for neuropathic pain are primarily pharmaceutical; however, in addition to eliciting debilitating side effects, pharmaceuticals result in just 50% pain reduction for only 30% of individuals with SCI-related chronic neuropathic pain (6). Consequently, many individuals who experience neuropathic pain prefer non-pharmaceutical alternatives (7). In a survey of 90 adults with SCI, 79% of those who reported on their preference for neuropathic pain management, preferred non-pharmaceutical treatments (8). Together, these data demonstrate the need to identify alternative treatment options to treat neuropathic pain among individuals with SCI.

One potential treatment to alleviate SCI neuropathic pain, improve affect, and potentially reduce risk for mood disorders (9, 10) is exercise. Exercise has been recognized as having positive effects on neuropathic pain among persons with SCI, despite the limited quantity and quality of evidence (11). However, evidence is emerging which indicates that exercise induced hypoalgesia may exist for persons with SCI. For example, in a case series, participants with SCI reported decreased neuropathic pain sensations following at least one of two bouts of self-selected, community-based exercise performed within a single week (9). The neuropathic pain-modulating benefits of acute exercise appear to persist with exercise training, as 10 weeks of aerobic exercise training led to decreased neuropathic pain for adults with SCI upon completion of the intervention (12). Various biological mechanisms underlying exercise induced hypoalgesia have been explored among able-bodied individuals, with evidence suggesting an interplay between the opioid, endocannabinoid, serotonergic and immune systems (13–15). However, the explanations for why exercise modulates neuropathic pain among persons with SCI are currently unknown.

The mechanisms underpinning neuropathic pain development are not well-understood. Various hypotheses exist in attempts to explain neuropathic pain development including structural damage within the central nervous system, and somatosensory cortex reorganization (16). However, the nociceptive environment also impacts the presence and severity of neuropathic pain (17). Pro-inflammatory cytokines such as interleukin-6 (IL-6), and tumor necrosis factor-alpha (TNF-α) have been shown to induce hyperalgesia, thereby increasing levels of neuropathic pain (18). In contrast, a reduction in these pro-inflammatory cytokines has been shown to prompt analgesic effects (17, 18). This relationship has also been demonstrated among persons with SCI using a dietary intervention. Relative to a control group, participants who consumed an anti-inflammatory diet for 12 weeks experienced changes in sensory neuropathic pain as a function of the change in pro-inflammatory cytokines (19). It is not known whether exercise can elicit the same effects in people with SCI.

In persons without SCI, research consistently shows that exercise can lead to increased acute levels of circulating IL-6, and subsequent rises in plasma concentrations of anti-inflammatory cytokines IL-1 receptor antagonist (IL-1ra), IL-10, and soluble TNF receptors (20, 21). However, exercise needs to be performed at a minimum intensity ($>50\%$ VO_2max) or duration (>30 min) for the anti-inflammatory response (21), analgesic effects (22) and improvements in affect (23) to occur. For persons with SCI above the 6th thoracic vertebrae (T6), the loss of somatic and autonomic control frequently results in a blunted cardiovascular response to exercise (24). Additionally, research evidence is inconclusive regarding the role and extent that the sympathetic nervous system plays in exercise-related changes in inflammation (25, 26) and subsequent analgesic effects. Although exercise can induce analgesia through upregulation of anti-inflammatory cytokines and reduced microglial activation in the central nervous system (27), it is unclear whether persons with SCI above T6 experience an attenuated analgesic response to exercise. Taken together, it is important to examine if any biomarkers can be used to predict who may experience exercise-related improvements in neuropathic pain following SCI.

The debilitating nature of neuropathic pain, coupled with the bidirectional relationship between pain and affect, highlight the need for understanding neuropathic pain as having both physiological and psychosocial contributors. Therefore, the primary purpose of this study was to test the effect of a single bout of sub-maximal aerobic exercise on inflammatory cytokines, neuropathic pain, and affect among individuals with SCI. Additionally, an exploratory aim of this study was to assess the relationship between baseline levels of cytokines and changes in neuropathic pain from pre- to post-exercise. Sub-maximal aerobic exercise was implemented in order to align with exercise guideline recommendations for people with SCI (28). It was hypothesized that a bout of sub-maximal aerobic exercise would lead to an acute increase in circulating levels of IL-6 and anti-inflammatory cytokines, decreased neuropathic pain, and improved affect from pre- to post-exercise among persons with SCI. Additionally, it was hypothesized that decreased neuropathic pain would be correlated with increased anti-inflammatory cytokines and improved affect.

METHODS

Participants

To participate in this study, individuals were required to: (1) have incurred an SCI > 12 months ago with an injury at the third cervical level or below (as long as diaphragmatic control and arm functioning allowed upper-body exercise); (2) experience chronic below-level of SCI neuropathic pain (>3 months) (at-level of SCI pain was excluded to minimize the risk of pain misidentification). Chronic neuropathic pain was an inclusion criterion given that neuropathic pain typically persists >3 months for persons with SCI, and to ensure that pain did not spontaneously resolve between testing sessions; (3) individuals were required to have the ability to read/write in English; and (4) routinely achieve at least the lowest level of the SCI exercise guidelines consisting of 20 min of moderate-to-vigorous intensity aerobic activity two times per week, and strength training two times per week, consisting of three sets of 8–10 repetitions of each exercise for each major muscle group (28). The latter criterion was instated because acute exercise participation by chronically inactive individuals may induce pain "flare-ups" (29) and impact inflammatory profiles.

Participants were recruited January–March, 2019 through advertisements emailed from community organizations from across British Columbia, and through emails directed toward individuals who had previously expressed interest in participating in SCI-studies. After screening, 10 active individuals with SCI of >1-year duration participated in this study. Ten individuals were the desired sample size based on previous case series data (9). This study carried the approval of the UBC Clinical Research Ethics Board (CREB; H18-03191), whereby all experiments were performed in accordance with CREB guidelines and regulations. Participants provided written, informed consent prior to enrolling in this study.

Procedural Overview

This study is a secondary analysis of case series data (clinicaltrials.gov registered: NCT03955523; 20/05/2019).

Measures

Inflammatory Cytokines

Blood samples were drawn by a trained phlebotomist from participants' most accessible antecubital vein. Samples were collected in the same clinical laboratory room as the exercise bout. All biosafety hazard protocols were followed, and JL's laboratory held a UBC-approved biosafety permit for this research space. Samples were placed in EDTA tubes and centrifuged at 2,000 g for 15 min at 4°C (Eppendorf, Hamburg, Germany), followed by a subsequent centrifuge at 10,000 g for 10 min at 4°C to remove platelets. The resultant supernatant was subsequently aliquoted and stored at −80°C. IL-6, IL-1ra and TNF-α were analyzed and quantified using the U-PLEX metabolic group 1 assay kits (K151ACL-1, LOT 289109, Mesoscale, Maryland, USA) according to manufacturers' instructions, and read using an MSD QuickPlex SQ120 plate reader (Mesoscale, Maryland, USA). All samples were analyzed in duplicate. The plate-specific intra-assay coefficients of variation (CV) were as follows: IL-10 = 10.4%; IL-1ra = 5.91%; IL-6 = 13.73%; TNF-α = 17.84%. The combined CV was 11.97%.

Neuropathic Pain Scale

Participants' neuropathic pain was measured using a modified version of the Neuropathic Pain Scale [NPS; (30)]. This 10-item scale (0 = nothing at all, 10 = most intense sensation imaginable) measures pain sensations common to neuropathic pain (e.g., "burning," "dull," "deep") in addition to measuring general pain qualities (i.e., "intensity" and "unpleasantness"). For each item on the NPS, participants were asked to verbally state how their pain sensation felt at that very instant. The first author (KRT) recorded each response. One question regarding the temporal nature of neuropathic pain was excluded, because it was not meaningful given the acute nature of the present study. Item scores were averaged to form a composite pain score at pre-, post- and 90-min post-exercise. The NPS has been shown to have sensitivity to detect acute treatment effects (30) and has been validated among people with various neuropathic pain syndromes (including SCI).

Affect

Hardy and Rejeski's (31) 11-point, single item Feeling Scale (FS) was used to measure participants' overall acute feeling of pleasure-displeasure (−5 = very bad, +5 = very good). In addition, Svebak and Murgatroyd's (32) Felt Arousal Scale (FAS) measured participants' perceived activation (1 = low arousal, 6 = high arousal), whereby "1" indicates feeling bored, relaxed or calm, and "6" indicates feeling excited, angry or frustrated. Using the FS alongside the FAS enhances construct validity by measuring activation in addition to affective valence [pleasure-displeasure; (33)]. Although the FS and FAS have not been validated in the SCI population, previous research has demonstrated the utility of these questionnaires for measuring affect in individuals with SCI participating in acute exercise (9, 10).

Protocol

Peak Power Output Graded Aerobic Exercise Test

Upon arriving at the lab, participants were given opportunity to use the toilet. After 5 min of seated rest, resting heart rate and blood pressure were measured. KRT explained how to use the Borg Rating of Perceived Exertion [RPE; (34)] (6–20) scale, adapted for persons with SCI (35). Participants then began the peak power output exercise test using a wall-mounted arm ergometer (Lode Angio, Groningen, Netherlands), with the height adjusted to align with the acromioclavicular joint. If required, participants' hands were bound to the handles of the arm ergometer with elastic tensor bandages. The test began with a 5-min warm-up at a self- selected cadence. Using a continuous graded exercise protocol (36), the resistance on the ergometer was adjusted such that power output was increased by 2 W/min for tetraplegics (37) and 10 W/min for paraplegics (38). Participants were asked to maintain a cadence of 55–65 revolutions per min (RPM) until volitional exhaustion. Within the last 10-s of each min of exercise, participants reported their RPE. The maximum wattage achieved during this exercise test was used to calculate the intensity (60% of maximum) of each participant's subsequent sub-maximal intensity aerobic exercise bout (operationalized as 60% peak power output).

Sub-maximal Aerobic Exercise Bout

Prior to arriving at the lab to complete the sub-maximal exercise bout, participants fasted for 12-h. Upon arrival, participants rested quietly for 10 min. Next, participants completed their baseline (T_0) measurements which consisted of verbally reporting their NPS, FS, and FAS ratings. Blood samples (TNF- α, IL-6, IL-1ra, IL-10) were then collected by the trained phlebotomist. Prior to exercising, KRT reminded participants

TABLE 1 | Demographic information of the sample ($n = 8$).

Participant	Sex	Age	Level of injury	AIS classification	Cause of SCI
1	M	56	T7	AIS A	Traumatic
2	M	32	T4-T5	AIS B	Traumatic
3	M	36	C6/C7	AIS B	Traumatic
4	F	56	T12-L1	AIS C	Traumatic
5	M	42	C5-C6	AIS C	Traumatic
6	M	35	C6/C7	AIS A	Traumatic
7	M	29	C6/C7	AIS C	Traumatic
8	M	25	T12/L1	AIS B	Traumatic

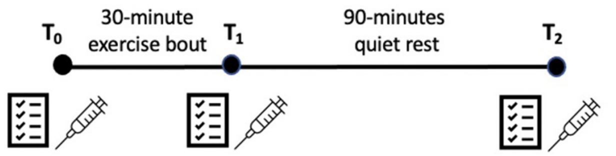

FIGURE 1 | Graphical representation of sub-maximal exercise protocol. Questionnaire symbol = neuropathic pain scale, felt arousal scale and feeling scale; syringe symbol = blood draws.

how to use the "6–20" RPE scale (34, 35). For the exercise bout, participants performed a 5-min warm-up at a self-selected pace and then performed 30-min of arm crank exercise at 60% of their maximum wattage, while maintaining a cadence of 55–65 RPM. Participants were prompted to report their RPE at the end of each minute during this exercise bout. If participants reported their RPE to be above "sub-maximal" (>16), the wattage was electronically lowered to 50% of participants maximum wattage, until their RPE recovered to "12–14," Immediately following exercise (T_1), participants verbally reported their NPS, FS, and FAS ratings, and provided another blood sample. Participants were then asked to quietly rest in a room separate from the testing room, and watch a video ["Planet Earth"; (BBC America)] for 90 min. After 90-min (T_2), participants repeated the NPS, FS, and FAS measures, and their blood was drawn one final time. The order of administration of the NPS, FS, and FAS was systematically randomized at each measurement timepoint to control for presentation biases. See **Figure 1** for graphical representation of study protocol.

Statistical Analyses

One-way repeated measures analysis of variance (ANOVA) with planned contrasts were conducted to assess change in neuropathic pain, cytokines and affect from pre- to post-exercise. Planned contrasts were the appropriate statistical method given that they allow for comparisons of just two means (of a set of means >2). The hypotheses of this study were based on changes in each dependent variable from pre- to post-exercise (i.e., not T_1T_2). Therefore, computing planned contrasts allowed for scientifically sensible comparisons and minimized the risk of a Type 1 error. Significance was set at $p < 0.05$. Effect sizes were calculated as Cohen's $d_{\text{av(average)}}$ with Hedge's g_{av} correction applied (39) and interpreted according to Cohen's conventions [small = 0.20, medium = 0.50, large = 0.80; (40)].

Simple change scores (Δ) were calculated for change in cytokine levels between T_0T_1 and T_0T_2. To control for the correlation between baseline and subsequent measures of the self-reported variables, residualized change scores were computed to measure change in NPS, FS, and FA scores between T_0T_1 and T_0T_2. Pearson's correlation coefficients were then computed to determine: (a) the relationships between change in neuropathic pain, change in inflammatory cytokines, and change in affect and arousal at timepoints $T_0T_{1,}$ and T_0T_2, and (b) whether baseline levels of inflammatory cytokines were related to change in neuropathic pain at timepoints T_0T_1 and T_0T_2. Two-tailed tests were used. Consistent with Widerstrom-Noga's recommendations (41), analyses were conducted separately for persons with tetraplegia and paraplegia. Cohen's conventions (40) were used for interpreting the magnitude of the correlations (small = 0.1, medium = 0.3, large = 0.5). SPSS version 22.0 was used for all analyses.

RESULTS

One male participant with tetraplegia withdrew from the study due to a pressure sore (unrelated to the study protocol). Additionally, the phlebotomist was unable to obtain blood from one male participant; his data are not included in the analyses. Therefore, the following results include data from 8 participants [4 tetraplegics (all male), and 4 paraplegics (3 male, 1 female)]. The average age of participants was 37.9 ± 10.9 years, and ranged between 25 and 56 years. Participants' average years post-SCI was 17.8 ± 8.9, and ranged between 3 and 36 years. Three participants were consuming cannabis at the time of study participation, however, no participants were taking pharmaceuticals at the time of study participation. Participant demographics for the 8 people who completed the study are presented in **Table 1**.

Exercise Intensity Manipulation Check

Manipulation checks were used to verify that participants were exercising at the intended training intensity. Mean_{RPE} during the exercise bout did not significantly differ between tetraplegic and paraplegic participants (M_{RPE} tetra = 13.07 ± 1.10, M_{RPE} para = 13.73 ± 0.69; $t = -1.02$, $p = 0.35$). Similarly, percent change in RPE between T_0T_1 did not significantly differ between tetraplegic and paraplegic participants [$\%_{\text{change}}$ tetra = 47.6 ± 24.49, $\%_{\text{change}}$ para = 47.48 ± 35.40; $F_{(1,6)} = 0.702$, $p = 0.99$]. Max_{RPE} (i.e., highest RPE reported during the exercise bout) for tetraplegic participants ranged from 12 to 15, indicating they were all exercising at a "somewhat hard to hard intensity" (32). For the paraplegic participants, Max_{RPE} ranged from 16 to 20, indicating that at certain points, they were exercising at "very hard intensity to exhaustion" (34). Results from an independent samples t-test indicated that average Max_{RPE} values for these two groups were significantly different (Tetra: $M = 14.00 \pm 1.4$; Para: $M = 17.25 \pm 1.89$; $t = -2.71$, $p = 0.03$). These data indicate that, consistent with the goal of the exercise manipulation, tetraplegic participants continuously exercised at a sub-maximal exercise intensity. However, paraplegic participants exceeded a sub-maximal intensity during their exercise bout (see 2-min average RPEs in **Figure 2**), and at points, may have exercised closer to maximal intensity.

Direct Effects of Exercise on NPS Scores, Affect, Arousal, and Inflammatory Cytokines

Given the small number of participants, results are reported and interpreted based on the magnitude of the effect sizes. Data are shown in **Tables 2–5**.

For persons with tetraplegia, between T_0T_1 there was some evidence of clinically meaningful changes in neuropathic pain [i.e., reductions > 30%; (42)] and improvements in affect and arousal. Effect sizes for changes in NPS scores, affect, and arousal between T_0T_1 were medium-to large ($g_{av} = -0.47$, 0.34, and 0.89, respectively). Changes in inflammatory cytokines between T_0T_1 were very small (g_{av} range: 0.02–0.16). Between T_0T_2, effect sizes for changes in NPS scores and affect were very-small to small (0.07 and 0.17, respectively). Effect sizes for arousal did not change between T_0T_2. Effect sizes for changes in inflammatory cytokines between T_0T_2 were also very-small (g_{av} range: 0.02–0.09).

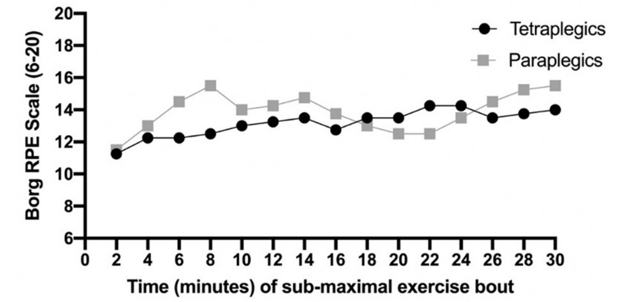

FIGURE 2 | Average of participants' RPE at 2-min intervals throughout the sub-maximal exercise bout.

For persons with paraplegia, calculation of the effect sizes provided support for some notable changes in pain, affect, and cytokine measures at both time points. Between $T_0T_{1,}$ the effect size for change in NPS scores was small (−0.15), and medium for affect and arousal (−0.51, 0.43, respectively). Effect sizes for change in inflammatory cytokines between T_0T_1 were very-small (0.08–0.17), except for IL-6 which was medium-large (0.66). Between $T_0T_{2,}$ change in NPS scores were small- to medium (−0.32), and change in arousal was medium (−0.51). Effect sizes for affect did not change between $T_0T_{2,}$. Effect sizes for change in T_0T_2 cytokines were very small (0.03–0.18), except for IL-6 which was medium-large (0.66).

Correlations Between Change in Inflammatory Cytokines and Change in Neuropathic Pain

For persons with tetraplegia, there were medium-to large, negative correlations between change in cytokine levels and change in NPS scores between T_0T_1 (*rs* ranged from −0.82 to 0.49), and T_0T_2 (*rs* > −0.67), except for IL-1ra ($r = -0.18$; **Table 3**).

For persons with paraplegia, correlations between change in T_0T_1 levels of inflammatory cytokines and change in NPS scores were medium to large (*rs* ranged from −0.60 to 0.79; **Table 5**) but were not in a consistent direction immediately post-exercise. However, correlations between change in NPS scores and change in levels of cytokines between T_0T_2 were small to medium (*rs* ranged from 0.17 to 0.42), except for IL-6 which was large ($r = 0.83$).

TABLE 2 | Changes in the study outcome measures among persons with tetraplegia ($n = 4$).

	T_0T_1					T_0T_2				
	Pre-exercise	Post-exercise	*p*-value	Effect sizes$^\sigma$	95% CI	Pre-exercise	90 min post-exercise	*p*-value	Effect sizes$^\sigma$	95% CI
Total NPS	2.60 (1.57)	1.75 (1.01)	0.10	−0.47	(−0.32, 2.02)	2.60 (1.57)	2.75 (1.70)	0.58	0.07	(−0.61, 0.91)
Feeling Scale	2.00 (1.41)	2.5 (0.58)	0.50	0.34	(−1.55, 2.55)	2.00 (1.41)	2.25 (0.50)	0.81	0.17	(−2.75, 3.25)
Felt Arousal Scale	2.50 (0.58)	3.00 (0)	0.18	0.89	(−0.42, 1.42)	2.50 (0.58)	2.50 (0.58)	1.00	——	—————
IL_6	1.72 (2.83)	1.63 (2.75)	0.26	−0.02	(−0.15, 0.33)	1.72 (2.83)	1.45 (1.59)	0.76	−0.09	(−2.35, 2.88)
IL_{10}	0.40 (0.34)	0.33 (0.31)	0.06	−0.16	(−0.00, 0.14)	0.40 (0.34)	0.36 (0.33)	0.11	−0.09	(−0.02, 0.10)
IL_{1RA}	137.4 (124.1)	143.1 (125.9)	0.40	0.03	(−14.50, 25.69)	137.4 (124.1)	143.1 (127.40)	0.33	0.03	(0.45, 10.95)
TNF-a	1.62 (1.31)	1.70 (1.45)	0.54	0.04	(−0.30, 0.46)	1.62 (1.31)	1.59 (1.35)	0.57	−0.02	(−0.12, 0.18)

Data are presented as Mean (SD); $^\sigma$ Hedge's g_{av}; Neuropathic Pain Scale (NPS) values are a composite value of 10 items rated on a numerical rating scale ranging from 0 to 10 (0 = no pain, 10 = worst pain imaginable). Feeling Scale (FS) is a single item rated on a bipolar scale (−5 = very bad, +5 = very good). Felt Arousal Scale is a 6-item scale rated on a 1–6 numerical rating scale (1 = low arousal, 6 = high arousal).

Cytokines are measured in pg/ml.

Correlations Between Change in Neuropathic Pain and Change in Affect

For persons with tetraplegia, as hypothesized, there were medium- to large, negative correlations between change in NPS scores, and change in FS and FA scores between T_0T_1, ($rs > -0.67$), and T_0T_2 ($rs > -0.67$).

For persons with paraplegia, between $T_0T_{1,}$ small- to-medium positive correlations were observed between change in NPS scores and change in FS scores ($r = 0.22$), and FA scores ($r = 0.33$). In contrast, between $T_0T_{2,}$ small-to medium, negative correlations were observed between change in NPS scores and FS scores ($r = -0.18$) and FA scores ($r = -0.53$).

Post-hoc Correlations Between Maximum Ratings of Perceived Exertion and Change in Neuropathic Pain

The hypotheses of the current study were based on acute responses to exercise at a sub-maximal intensity. However, the manipulation check of Max_{RPE} data revealed tetraplegic participants were indeed exercising at a sub-maximal intensity, but paraplegic participants may have surpassed this sub-maximal level at certain points in the exercise bout, and approached an exercise intensity near maximal. As previous research among chronic pain populations indicates that *extremely* high exercise intensities may acutely increase experimentally induced pain (43, 44), a *post-hoc* decision was made to compute Pearson's correlations between Max_{RPE} and NPS scores to determine if the relationship between Max_{RPE} is different for tetraplegic vs. paraplegic participants.

For persons with tetraplegia, there were medium-to-large, negative correlations between Max_{RPE} and change in NPS scores between T_0T_1 ($r = -0.91$) and T_0T_2 ($r = -0.68$).

In contrast, for persons with paraplegia, a medium-to-large positive correlation was found between Max_{RPE} and change in NPS scores between T_0T_1 ($r = 0.63$). However, between $T_0T_{2,}$ a large, negative correlation was observed between Max_{RPE} and change in NPS scores ($r = -0.83$).

TABLE 3 | Correlations between changes in pain, changes in affect, changes in arousal, and changes in cytokines among persons with tetraplegia ($n = 4$).

	T_0T_1		T_0T_2	
Correlations	***r***	***p*-value**	***r***	***p*-value**
r Δ NPS/ΔFS	−0.67	0.33	−0.67	0.33
r Δ NPS/ΔFAS	−0.75	0.25	−0.94	0.06
r Δ NPS/ΔIL6	−0.82	0.18	−0.67	0.33
r Δ NPS/ΔIL10	−0.40	0.61	−0.97*	0.04
r Δ NPS/ΔIL1RA	−0.62	0.38	−0.18	0.83
r Δ NPS/ΔTNF-a	0.49	0.52	0.83	0.17
r Δ NPS/MaxRPE	−0.91**	0.04	−0.68	0.16

*$p < 0.05$ (2-tailed); **$p < 0.05$ (1-tailed). Bonferroni adjusted for multiple comparisons.*

Correlations Between Baseline Levels of Cytokines and Change in Neuropathic Pain

A large-sized significant, positive correlation was observed between baseline levels of IL-1ra and change in NPS scores between T_0T_1 ($r = 0.833$; **Table 6**; **Figure 3**). However, this relationship was not sustained 90-min post-exercise ($r = -0.17$). Correlations between baseline levels of all other cytokines and change in NPS scores were inconsistent in size and direction at timepoints T_0T_1 and T_0T_2 ($rs = -0.27$–0.55; **Table 6**).

DISCUSSION

The primary purpose of this study was to test the acute effects of sub-maximal aerobic exercise on inflammatory cytokines, neuropathic pain, affect and arousal among individuals with SCI. Additionally, this study aimed to explore if the baseline levels of cytokines may be related to exercise-related changes in neuropathic pain. Consistent with our hypotheses, exercise led to decreased levels of neuropathic pain, and improved affect, in both participants with tetraplegia and paraplegia. However, changes

TABLE 4 | Changes in the study outcome measures among persons with paraplegia ($n = 4$).

	T_0T_1					T_0T_2				
	Pre-exercise	**Post-exercise**	***p*-value**	**Effect sizes$^\sigma$**	**95% CI**	**Pre-exercise**	**90 min post-exercise**	***p*-value**	**Effect sizes$^\sigma$**	**95% CI**
Total NPS	1.83 (1.45)	1.53 (1.39)	0.21	−0.15	(−0.30, 0.90)	1.83 (1.45)	1.28 (1.05)	0.09	−0.32	(−0.16, 1.26)
Feeling Scale	2.75 (1.26)	2.0 (0.82)	0.44	−0.51	(−1.97, 3.47)	2.75 (1.26)	2.75 (1.26)	1.00	−−	−−−−−
Felt Arousal Scale	2.75 (1.26)	3.50 (1.29)	0.32	0.43	(0.28, 1.22)	2.75 (1.26)	2.00 (0.82)	0.06	−0.51	(−0.04, 1.54)
IL$_6$	1.40 (0.13)	1.58 (0.35)	0.39	0.66	(-0.39, 0.75)	1.40 (0.13)	1.30 (0.17)	0.42	−0.66	(−0.26, 0.46)
IL$_{10}$	0.25 (0.19)	0.21 (0.16)	0.063	−0.17	(−0.01, 0.09)	0.25 (0.19)	0.23 (0.20)	0.29	−0.07	(−0.04, 0.08)
IL$_{1RA}$	179.7 (90.8)	198.0 (123.9)	0.79	0.12	(−178.6, 215.2)	179.7 (90.8)	161.9 (45.24)	0.53	−0.18	(−61.0, 96.6)
TNF-a	1.15 (0.84)	1.08 (0.81)	0.63	−0.08	(−0.37, 0.51)	1.15 (0.84)	1.11 (0.85)	0.69	−0.03	(−0.28, 0.36)

Data are presented as Mean (SD); $^\sigma$ Hedge's g_{av}; Neuropathic Pain Scale (NPS) values are a composite value of 10 items rated on a numerical rating scale ranging from 0 to 10 (0 = no pain, 10 = worst pain imaginable). Feeling Scale (FS) is a single item rated on a bipolar scale (−5 = very bad, +5 = very good). Felt Arousal Scale is a 6-item scale rated on a 1–6 numerical rating scale (1 = low arousal, 6 = high arousal).

Cytokines are measured in pg/ml.

in inflammatory cytokines following exercise participation were inconsistent in direction. For example, a small-sized acute decrease in IL-6 was observed for persons with tetraplegia following exercise participation, whereas a medium-large acute increase in IL-6 was observed for persons with paraplegia. Interestingly, lower baseline levels of IL-1Ra were significantly related to greater acute decreases in neuropathic pain. To the best of our knowledge, this is the first SCI study to evaluate the effects of exercise on neuropathic pain, inflammatory cytokines and affect.

TABLE 5 | Correlations between changes in pain, changes in affect, changes in arousal, and changes in cytokines among persons with paraplegia ($n = 4$).

	T_0T_1		T_0T_2	
Correlations	***r***	***p*-value**	***r***	***p*-value**
r Δ NPS/ΔFS	0.22	0.78	−0.18	0.82
r Δ NPS/ΔFAS	0.33	0.67	−0.53	0.47
r Δ NPS/ΔIL6	−0.60	0.40	0.83	0.17
r Δ NPS/ΔIL10	0.79	0.21	0.17	0.83
r Δ NPS/ΔIL1RA	−0.40	0.60	0.32	0.68
r Δ NPS/ΔTNF-a	0.70	0.30	0.42	0.58
*r*Δ NPS/ MaxRPE	0.63	0.19	−0.83	0.08

Bonferroni adjusted for multiple comparisons.

TABLE 6 | Correlations between baseline levels of cytokines and changes in pain for all study participants ($n = 8$).

	T_0T_1		T_0T_2	
Correlations	***r***	***p*-value**	***r***	***p*-value**
r baseline IL-6/Δ*NP*	0.55	0.155	0.24	0.576
r baseline IL-10/Δ*NP*	−0.27	0.521	0.11	0.793
r baseline IL-1ra/Δ*NP*	0.83**	0.010	−0.17	0.687
r baseline TNF-a/Δ*NP*	0.004	0.993	0.27	0.52

***$p \leq 0.01$ (2-tailed). Bonferroni adjusted for multiple comparisons.*

The observed positive effects of exercise on neuropathic pain align with previous SCI research (6, 7, 9–11). However, previous research suggests that exercise reduces neuropathic pain to a similar extent for paraplegics and tetraplegics. In our study, these effects were large for tetraplegics and small for paraplegics. These differences may be at least partly attributable to the significant differences in intensity at certain points of the exercise bout, as reported by tetraplegic vs. paraplegic participants.

Indeed, research in able-bodied individuals suggests that the analgesic effects of exercise are elicited once exercise exceeds a threshold of intensity. Moderate- to high-intensity exercise [but not maximal; (14, 45)], has been shown to lead to greater reductions in pain compared to lower intensity exercise in other chronic health populations. However, performing exercise that is perceived to be "very hard to exhaustive" may increase pain, at least in the short-term. Paraplegics reported exercising at a "very hard" subjective intensity at certain points during this exercise bout, and their neuropathic pain and Max_{RPE} were positively correlated immediately post-exercise. The correlation became negative at 90-min post-exercise, paralleling greater reductions in neuropathic pain at 90-min post-exercise for this group. Research suggests that exercise-induced hypoalgesia remains for ≤30 min after exercise, but may be impaired in individuals with chronic pain (14). Therefore, paraplegic participants may have highly exerted themselves during this bout of exercise, which inhibited exercise-induced hypoalgesia immediately after exercise completion. Exercising "near maximal" intensity, may have conflated paraplegic participants immediate post-exercise neuropathic pain reports, with muscle soreness. Ninety minutes of recovery may have allowed for sufficient rest, and for paraplegic participants to experience the benefits of high-intensity exercise, such as reduced neuropathic pain. Future research must continue to examine the impact of exercise intensity on neuropathic pain in persons with SCI, given the

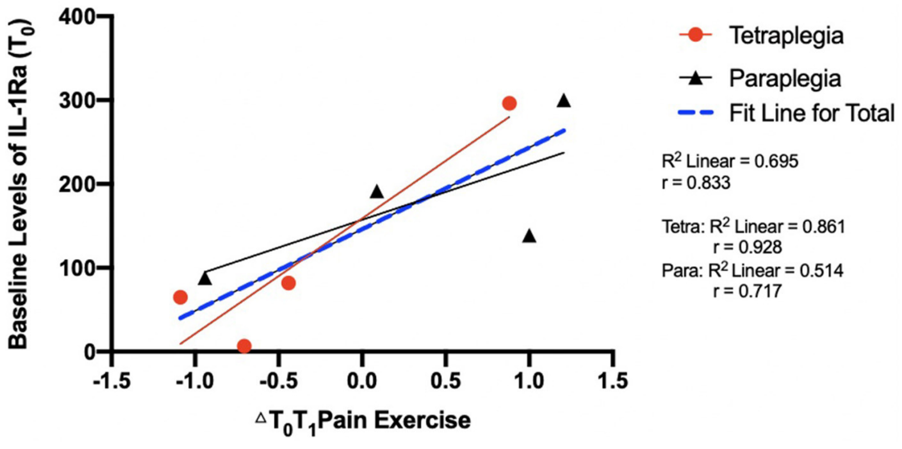

FIGURE 3 | Scatterplot of baseline levels of IL-1Ra and changes in neuropathic pain between T_0T_1.

distinctly different relationships with neuropathic pain when participants met, vs. exceeded a "hard" (34) exercise intensity in this study.

Among the able-bodied population, research evidence indicates that exercise-related changes in IL-6 is intensity dependent (46), and IL-6 can be sizeably increased compared to baseline levels following high-intensity exercise (47). Results from this study support previous findings, given the large increase in IL-6 that occurred from pre- to post-exercise for persons with paraplegia (i.e., "very hard") exercisers. IL-6 is an inflammation-controlling cytokine and stimulates the exercise-related anti-inflammatory cascade, which suggests that tetraplegic participants may not have been exercising at a high enough intensity to initiate increased levels of anti-inflammatory cytokines. Research evidence also indicates that exercise-related increases in IL-6, and the subsequent anti-inflammatory cascade is further responsible for eliciting analgesic effects (17, 18, 48). However, results from this study do not align with this evidence, given that a stronger correlation was observed between IL-6 and neuropathic pain for persons with tetraplegia. Therefore, conflicting correlations between neuropathic pain and inflammatory cytokines, coupled with the significant difference in Max_{RPE} reported between levels of injury, suggests the inflammatory etiology of exercise-related changes in neuropathic pain among persons with SCI may also be impacted by exercise intensity. Future research should test the mediating effects of inflammation, by identifying whether specific exercise intensities lead to decreases in neuropathic pain, through effects on the inflammatory cascade.

Although impaired autonomic nervous systems may contribute to the conflicting pattern of correlations observed for tetraplegic vs. paraplegic participants between neuropathic pain, affect and arousal, differences in exercise intensity reported between these groups may help further explain these correlations. The intensity of exercise required to stimulate optimal affective responses remains highly debated (43, 44, 49). For individuals with SCI, previous literature demonstrates that acute exercise-related decreases in pain are correlated with improvements in feeling states (9, 10). However, the intensity and type of exercise prescribed within these previous studies were inconsistent. For persons with tetraplegia, the large, negative correlations between neuropathic pain, affect and arousal align with previous research (9, 10). In contrast, for persons with paraplegia, exercise-related changes in neuropathic pain were positively correlated with changes in feeling states immediately post-exercise, and negatively correlated 90 min post-exercise. Indeed, moderate vs. high-intensity exercise has been shown to differentially impact affective responses and pain sensations in the general population (43). Future research should be directed toward understanding the impact of exercise intensity on these constructs among persons with SCI.

Further, the timepoint of assessments of affective responses may impact the interpretation of results. For participants with paraplegia, conflicting affective responses immediately post-exercise vs. 90-min post-exercise may be explained by the rebound model (49). High-intensity exercise often stimulates negative affective responses immediately post-exercise, whereas these responses become positive after a period of recovery. Paraplegic participants exercised near maximal intensity and may have experienced a disruption in physiological homeostasis immediately post-exercise. However, 90-min of rest may have been sufficient for paraplegics to recover and experience an affective "rebound." While the intensity of exercise and timing of affective assessments are not presumed to fully explain participants' affective response to exercise nor its correlation with neuropathic pain, they are presented here as potential exercise protocol characteristics that may partially explain these relationships. Future psychophysiological SCI-exercise research should also investigate the impact of additional social-cognitive and physiological predictors of affective responses to exercise (e.g., exercise self-efficacy, thermoregulation, and ventilatory threshold) and their relationship with neuropathic pain.

In addition to its primary purpose, this study provided the opportunity to assess whether inflammatory cytokines may be used as predictive biomarkers to determine individuals likely to benefit from exercise. Lower levels of IL-1ra at baseline were associated with larger exercise- related reductions in neuropathic pain. IL-1ra is an anti-inflammatory cytokine which is stimulated by post-exercise increases in IL-6. Therefore, individuals who have lower baseline levels of IL-1ra may have a greater capacity for upregulation of IL-1ra in response to exercise, thereby inducing anti-inflammatory and subsequent analgesic effects. Results from this study support previous research among persons with pain (e.g., knee osteoarthritis), whereby levels of plasma IL-1ra have been shown to predict response to treatment (50). Future research, including the RCT associated with this pilot study (51), should continue to investigate IL-1ra as a potential predictive biomarker for exercise-related changes in neuropathic pain among persons with SCI. Better understanding predictive biomarkers will enhance our knowledge of who may experience improvements in neuropathic pain following exercise, and therefore inform patient care decisions (52).

Study Strengths and Limitations

This study has several strengths. First, collecting multiple measurements and observing possible pathways responsible for exercise-related changes in pain allowed for a deeper evaluation of the relationship between exercise and neuropathic pain among persons with SCI. Results of this study provide rationale to further examine potential mechanisms impacting exercise-related changes in neuropathic pain, such as whether the intensity of exercise leads to decreased neuropathic pain, through its effects on inflammation among individuals with SCI. Second, evaluating the effect of exercise on neuropathic pain among humans with SCI (rather than animals), allowed for use of clinically relevant measurement tools and the ability to measure spontaneous neuropathic pain. Evaluating animal models would have precluded evaluating psychosocial contributors to neuropathic pain, given the difficulty of assessing affective measures of neuropathic pain in pre-clinical research (53). Third, assessing the concomitant impact of exercise on neuropathic pain, inflammation and affect allowed for a greater understanding of the dynamic interaction among physiological (i.e., inflammation) and psychological (i.e., affect) contributors to neuropathic pain.

Despite these strengths, some limitations must be noted. First, participants' individual sub-maximal exercise intensity was based on their peak power output rather than their VO_{2peak}. Although we intended for all participants to exercise at 60% peak power output, there was a significant difference in Max_{RPE} reported between tetraplegic and paraplegic participants at certain points, and paraplegic participants likely exceeded a sub-maximal exercise intensity. Further, paraplegic participants approached an "exhaustive" RPE (34) halfway through the exercise bout, which prompted the research team to decrease their wattage from 60 to 50% peak power output for 5 min to allow for a brief recovery period. Despite this limitation, the significantly different Max_{RPE} reported between tetraplegic vs. paraplegic participants provided valuable insight into the potential impact of exercise intensity on neuropathic pain, inflammatory cytokines, affect and arousal among individuals with SCI. Second, although International SCI Pain Data Sets have been introduced to measure neuropathic pain in adults with SCI (54), we employed the NPS due to its brevity, ease of comprehension, and ability to assess responses to treatment (30). Future research should incorporate the International SCI Pain Data Set in order to ensure proper identification of participants' pain, and to allow for comparison of results across studies (51). Third, the FS and FAS have been used extensively in exercise research to investigate individuals' affective responses to exercise. Additional research is needed to assess the validity of the FAS when used in exercise contexts with persons with SCI. Fourth, this study included a small number of participants, and only one female, which is limiting due to the research evidence supporting sex differences in post-exercise immune and pain responses (55, 56). While the ratio of male to female participants in this study is representative of the global SCI population (57), future SCI research must strengthen recruitment strategies to include more female participants and enhance the generalizability of findings. The small sample size may have influenced our results (in terms of both effect size and lack of statistical significance), and these results should be interpreted with caution as they were not all statistically significant. Fifth, this study evaluated only inflammation as a possible pathway responsible for exercise-related changes in neuropathic pain. It is understood that many additional mechanisms (e.g., microglial activation, cortisol levels, pain catastrophizing) may be responsible for exercise-related changes in neuropathic pain among adults with SCI. To progress toward mechanism-based treatment, future SCI research should investigate further mechanisms that may impact the relationship between exercise and neuropathic pain. And finally, we did not employ a control condition against which to evaluate the effects of the exercise bout. Designing a true control for tests of pain-reducing interventions is challenging, but should be considered in future studies to control for the effects of attention, distraction and other psychological variables on neuropathic pain and affect.

Conclusion

Taken together, the results of this study suggest that exercise may reduce neuropathic pain and improve affect in adults with SCI, and changes in inflammation may be related to these effects. Additionally, exercise intensity may play an important role in the exercise related changes in neuropathic pain, inflammatory profiles, and affect for adults with SCI. Last, levels of IL-1ra may help determine who experiences exercise-related reductions in neuropathic pain sensations. Future research should be directed toward understanding the ideal exercise intensity for decreasing neuropathic pain among adults with SCI, and the potential role of inflammatory cytokines and other possible mediators.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, upon reasonable request.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of British Columbia Clinical Research Ethics Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

KT was responsible for designing the study protocol, writing the protocol and report, participant recruitment, data collection, analyzing data, interpreting results, writing manuscript, updating reference lists, and creating tables and figures. JV, JL, and JK were responsible for designing the study protocol, interpreting results, and manuscript revision. JW was responsible for designing the study protocol, assisting with data collection, interpreting results, and manuscript revision. GJ was responsible for designing the study protocol, analyzing data, and manuscript revision. GD was responsible for designing the study protocol, assisting with data collection and analysis, and revising the manuscript. KMG was responsible for designing the protocol, assisting with writing the protocol and report, analyzing data, interpreting results, manuscript revision, and designing tables and figures. All authors contributed to the conception and design of this study, in addition to drafting, or critically revising the article.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.700780/full#supplementary-material

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The Experience of Locomotor Training From the Perspectives of Therapists and Parents of Children With Cerebral Palsy

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Objective: The objective of this study was to explore the experiences of intensive locomotor training from the perspective of therapists and parents of children with cerebral palsy.

Design: A qualitative study using semi-structured interviews was employed to capture perspectives following an intensive locomotor training intervention. Data were analyzed thematically, systematically coding and interpreted by grouping information into themes and sub-theme categories.

Participants: Five therapists and seven parents of children with high daily physical assistance and equipment needs participated in the study.

Setting: A pediatric tertiary hospital.

Results: Experiences of locomotor training were described with relation to the suitability of locomotor training with sub-themes of intervention length and time, engagement within sessions, the importance of support, and the utility of locomotor training beyond a research context. Motivation for participating in locomotor training was described in relation to the enjoyment of movement and for increasing activity level. The barriers and facilitators who participated in locomotor training provided environmental and personal factor subthemes. Finally, the outcomes from the intervention were related to improvements in physical health, sleep, affect and emotion, and ambulation in daily activities.

Conclusion: The experience of intensive locomotor training from the perspectives of parents of children who have high physical assistance and equipment needs and the therapists providing the intervention was described. Future studies should consider outcome measures beyond motor capacity to quantify the perceived outcomes of interventions that are meaningful to families.

Keywords: qualitative, locomotor training, cerebral palsy, physical activity, sedentary

INTRODUCTION

Cerebral palsy (CP) is a complex life-long neurological condition primarily affecting movement and posture. It is the most common cause of physical disability in childhood (1). For children and youth who are dependent on physical assistance and equipment for mobility throughout the day, there are limited evidence-based interventions available that aim to increase physical activity and improve gross motor function (2). This is despite secondary complications of inactivity and physical deterioration that most notably occurs between the ages of 7 to 9 years (3). These children and youth are described under the Gross Motor Function Measure Classification System (GMFCS) as being levels III (ambulators with assistance), IV (non-ambulant but able to sit unassisted), and V (non-ambulant and unable to sit) (4). For this group of children and youth, the combination of limited evidence-based interventions and physical deterioration significantly impacts quality of life and overall health and well-being (5).

Locomotor training is an activity-based approach that aims to support the development of stepping skills in individuals with significant gross motor limitations (6, 7). Locomotor training is usually delivered in two parts, first through partial body weight supported treadmill training (PBWSTT) to enable individuals to be supported in the development of stepping whilst managing less of their own body weight (8). A harness is used to support the safe attainment of a more upright position with reported benefits in walking speed, endurance, and potential efficacy in children with more severe physical limitations (9–11). Second, overground walking practice is also incorporated into locomotor training providing task specific whole-task practice (8). For children classified within GMFCS levels III, IV, and V, PBWSTT and overground walking practice have strong recommendations for the outcomes of improving walking distance, providing the experience of walking for well-being, and inclusion and for improving transfer abilities (12). With the growth of new technologies, locomotor training has also expanded to include robotic assistive gait training (RAGT). The use of RAGT is usually adopted to increase engagement and provide a higher dosage of training with less therapist involvement (13). Both PBWSTT and RAGT present as viable options to facilitate engagement in physical activity particularly for children with CP who have more physical limitations. Typically, engaging in locomotor training for individuals with neurological conditions requires a higher dosage of treatment where attendance occurs over several sessions a week for a number of weeks (14). This is mainly based on current recommendations for exercise and physical activity prescription for children with CP (15, 16).

Between June 2015 and January 2017, a clinical trial co-designed by consumers known as iStride was conducted in a pediatric tertiary hospital in Perth, Australia. The aim of the trial was to determine if the addition of RAGT (utilizing the RT600, Restorative Therapies, Baltimore, MD, USA) to PBWSTT improved motor outcomes compared to PBWSTT alone in a randomized controlled trial. This trial recruited 40 participants aged between 5 and 12 years and classified them as functioning at GMFCS levels III, IV, and V. The intervention involved a high dosage of treatment with participants attending three 1 h sessions a week for 6 weeks (17). As such, evaluating the experience of this high dosed intervention from the perspectives of parents, children, and therapists involved in the intervention was considered vital.

Although service providers endeavor to deliver holistic, strength-based interventions (18), there is a paucity of qualitative reports on the outcomes of intensive therapy models and, in particular, locomotor training in children with CP functioning within GMFCS levels III, IV, and V. Qualitative approaches can provide evidence related to the experience of an intervention (16), insight into the value that different stakeholders attach to different intervention outcomes, and uncover considerations relevant to implementation and practice (19). Understanding the experiences of locomotor training through a strengths-based lens, which is considered essential in childhood disability (18), ensures that the evidence-base represents outcomes that are meaningful to therapists, children, and their families.

Therefore, this qualitative study aimed to explore the experience of intensive locomotor training from the perspectives of parents of children and youth with CP (GMFCS levels III-IV-V) and therapists. Specifically, we aimed the following:

1. Describe the outcomes of locomotor training from the perspective of parents, children, and therapists providing the intervention and;
2. Inform future best practice care and research for children functioning with GMFCS levels III, IV, and V.

METHODS

Design

A qualitative description approach was employed to evaluate the iStride randomized controlled trial (https://www.anzctr.org.au/ Trial number: ACTRN12615001149550). Contrasted with other qualitative approaches that interpret the meaning or develop theory, the goal of qualitative description was to provide a rich and clear description of an experience or process to inform and improve healthcare (20, 21). Human ethics approval was obtained from the Human Research Ethics Committees of Perth Children's Hospital and Curtin University, Perth Australia. Written informed consent and assent for participant and publication was obtained from children and their parents.

Participants

Purposive sampling was used to select participants for this study. This was used to ensure that only children, parents, and therapists involved in the program were sampled. It was considered vital that all invited participants were within 2 weeks of completing the program (**Figure 1**) (22). This sampling strategy aimed to optimize the trustworthiness of the data so that it did not rely on parents, children, and therapists to recall experiences, rather so that they could reflect on a very recent experience of the intervention. Participants were invited to participate in this study based on their involvement in a 6-week locomotor training intervention (18 h in total). Participants were recruited from the following groups:

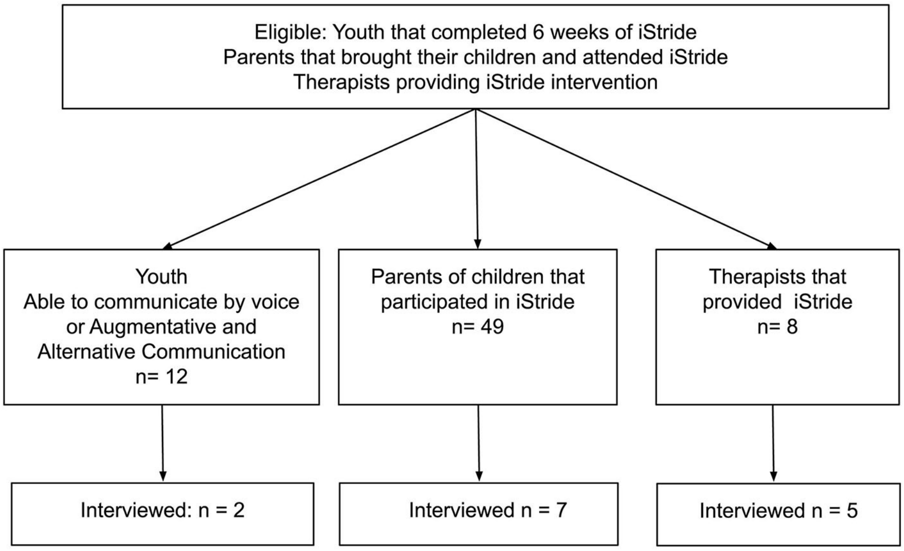

FIGURE 1 | Qualitative study flow chart.

TABLE 1 | Key topics and prompts in semi-structured interview guides.

Topic	**Prompts**		
	Therapists	**Parents**	**Children**
Experience	Explain your role in the intervention	Explain the child and parent experience in the intervention	e.g., Tell me about participating in iStride
Body structure and function-related outcomes	Strength, tone, postural control etc.; unintended outcomes.	Strength, tone, postural control etc.; unexpected outcomes.	e.g., Is anything about your body different?
Activity-related outcomes	Mobility, transfers, self-care etc.	Mobility, transfers, self-care etc.	e.g., Can you do new things?
Participation-related outcomes	Attendance and involvement in therapy sessions.	For child and family; Attendance and involvement at home, school, community.	e.g., What is it like when you are at school now?
Contextual factors	Use of robotic devices for therapy; beliefs and attitudes toward intensive intervention; training/skills required	Hospital-based; role of staff; interaction with other families; role demands; intervention equipment	e.g., What was it like using a robot to help you walk?
Impact	Professional practice; recommendations for practice	Goals for child; impact on parent and family; maintaining outcomes.	e.g., How would you explain this to other children?

Group 1: Parents of children that participated in either the RAGT or PBWSTT group of the locomotor training intervention.

Group 2: Therapists or research assistants who provided the locomotor training intervention throughout the course of the iStride study.

Group 3: Children and youth who participated in the locomotor training intervention. Children were eligible to participate in this study if they (i) were a participant in either the RAGT or PBWSTT group and (ii) had completed the 6-week intervention (18 sessions in total). Children were able to communicate by voice or by augmentative and alternative communication (AAC).

Data Collection Methods

Semi-structured interviews were carried out over October 2017 and March 2018. Interview guides were specific to each group to reflect appropriate language and jargon containing questions and prompts that were designed to guide the interview in a focused, yet flexible manner. The interview guide was developed and piloted with parents who participated in other mobility interventions in a pediatric rehabilitation outpatient department to obtain feedback of utility prior to use in data collection (**Table 1**).

An exercise physiologist (CW) with expertise in consumer engagement, interviews, and qualitative research in the area of participation in youth with physical disabilities conducted all of the interviews. The interviewer was not involved in any aspect of the intervention provided as part of iStride. The interviews were conducted with parents and children at the conclusion of their 6-week locomotor training intervention (dosage of three, 1 h sessions a week for 6 weeks). The interviews were conducted in the homes of children and parents, or in a private clinic room in the rehabilitation department at a pediatric tertiary hospital (Perth, Australia). Depending on participant preference and availability, interviews with parents also included interviews with youth. Interviews with therapists occurred at the conclusion of the iStride intervention in a private clinic room at the pediatric tertiary hospital. Interviews were approximately 60 min in duration. Audio recordings were used to capture the content of all interviews and field notes were taken.

Data Analysis

Discussions were transcribed verbatim and compared with field notes taken during interview sessions. Constant comparative coding was used, whereby new codes were compared with those that had emerged from previous interviews through a process of moving back and forth between transcripts. Open coding was applied to each of the interview groups (parents, children, and therapists) whereby three investigators (DP, LW, and AT) generated the initial codes, labeling the meaning units in a spreadsheet on Microsoft Excel. After applying the open coding framework, meaning units were reviewed to merge similar codes and generate themes. Two authors (DP and AT) then completed the thematic analysis over the course of multiple discussion meetings. Following these meetings, the critical friends approach (23), where the data could be challenged and interpreted by an investigator not involved in the generation of meaning units and themes occurred. The coding was combined for the three groups (parents, children, and therapists) with themes grouped into higher-order categories according to discussion areas and reflected broadly the (a) experiences of the intervention and (b) perceived outcomes of the intervention.

Trustworthiness

Ethical considerations relating to the qualitative component of this trial are largely addressed by having an independent researcher with no previous relationship to trial participants or involvement in the intervention. Several methods to ensure trustworthiness were also undertaken. Credibility was determined by member checking whereby participants reviewed transcripts to confirm the accuracy of their interview (23). In addition, the purposive sampling strategy enabled recent experiences to be explored from participants who were directly involved in the intervention. Comparable conclusions between the three investigators who reviewed the meaning units occurred, providing feedback on and enabling detailed discussions to resolve discrepancies whilst considering any alternative meaning units. Transferability was determined by purposive sampling where research participants were chosen purposively to include children with CP (GMFCS levels III, IV, and V) and their parents and therapists who provided the intervention (22). Dependability was determined by overlap methods where triangulation of data was also undertaken with the quantitative data that was obtained from the randomized controlled trial (24).

RESULTS

Twelve participants (including seven parents: six females, one male, and five therapists: all female) were interviewed. Combined between the seven parents included six children classified within each of the GMFCS levels III, IV, and V. Pseudonyms are used for each child when they are referred to by therapists, parents, or the child themselves. This includes Adam and Nathan (GMFCS level III), Jackson and Jordan (GMFCS level IV), and Dennis and Stephanie (GMFCS level V). In addition to this, only two of these children were available at the time to be interviewed (**Table 2**). This included Jackson (GMFCS IV, male, 8 years old, verbal, living at home with both biological parents) and Dennis (GMFCS V, male, 8 years old, using AAC, living at home with both biological parents). The discussions generated 58 pages of 12-point, single spaced text. Themes were grouped into higher-order categories according to discussion areas and reflected broadly the (a) experiences of the intervention and (b) perceived outcomes of the intervention. In the material that follows, we elaborate on these categories, and the supporting tables provide additional meaning unit examples to supplement those presented in the main text.

TABLE 2 | Participant characteristics (*n* = 14).

Characteristic	Frequency (%)
Parents	
Mothers	6 (42.9)
Fathers	1 (7.1)
Children	
GMFCS level IV	1 (7.1)
GMFCS level V	1 (7.1)
Therapists	
Physiotherapist	2 (14.3)
Therapy Assistant	2 (14.3)
Occupational Therapist	1 (7.1)

GMFCS, Gross Motor Function Classification System.

Experiences of the Intervention

Meaning units in this category were grouped according to whether therapists and families were discussing the suitability of the locomotor training (during and beyond the research project), their motivations for participating in locomotor training, and the barriers and facilitators to their participation in the locomotor training. A description of each theme, and sub-themes, are presented in **Table 3**.

Suitability of Locomotor Training

Parents and therapists described several structural and content elements of locomotor training intervention. Most notably, they emphasized (a) the intervention length and timing (b), engagement within the sessions, (c) the importance of support, both peer and program, and (d) the utility of locomotor training beyond research.

Intervention Length and Timing

Therapists felt that the 6 weeks of intervention offered to families was optimal. One therapist commented,

TABLE 3 | Feasibility and theme and sub-theme description.

Theme	Theme description	Sub-theme
Acceptability and suitability of locomotor training	The extent to which intensive locomotor training is judged to be suitable to therapists providing the intervention and for participants and their families receiving it, and their perceptions of its utility beyond a research project.	• Intervention length and time • Engagement within sessions • Importance of support • Utility of locomotor training beyond research
Motivations for participating	The extent to which intensive locomotor training is of interest to participants and their families. This includes participant accessibility to activity-based therapy programs that may already be available to them.	• Enjoyment of movement • Increasing activity level
Practicalities and implementation of an intensive therapy program	The personal and environmental barriers and facilitators that affect the implementation and provision of locomotor training and participation in intensive locomotor training.	• Environmental factors • Personal factors
Efficacy/Outcomes	Physical health benefits achieved through intensive locomotor training, including fitness, strength, gross motor, tone and well-being.	• Physical health
	Locomotor training contributes to improved sleep quality and duration.	• Sleep
	Locomotor training induced improvements in mood, confidence, motivation and enjoyment.	• Affect and emotion
	The ability to be more active and independent throughout the day.	• Participation in daily activities

> "A lot of these families are busy, so, so busy so I think 6 weeks is good. Any longer, and I think you probably wouldn't get the attendance."

Another spoke about the benefits they started to see for the participants toward the end of the 6 weeks,

> "Like we quite often don't see change or much change I should say until like week five. We call it magic week five, when all of a sudden it clicks and big things start happening but in saying that by 6 weeks, a lot of the kids are ready for it to be finished, they are tired and they still have school and all their other commitments."

Parents indicated that for them, a 6-week intervention was achievable, and they could be flexible to meet the needs of their child. However, parents believed that any more than 3 days per week dedicated to an intervention like locomotor training would be too big a commitment. As one parent said,

> "Three days a week is nice. I think anything more might be hard to come into the hospital that many times a week. It wouldn't be a problem if the sessions were longer but I think more frequently might be a bit difficult."

Engagement Within the Sessions

Parents also enjoyed how their children engaged with the sessions. When reflecting on the participation of their child during the sessions, Dennis' mother commented,

> "Well, they [the sessions] were fun–whether it's [fun] somewhere else I don't know…I think it needs to be fun because it's such hard work so it's good to take the focus off the hard work." (During this comment, Dennis also nods in agreeance).

Another described the enjoyment of their child in the sessions,

> "it was excellent, it was great....yeah just to see the difference in him, and he enjoyed it he really did enjoy it. He had a couple of "I don't want to do this" but more often than not he was quite up for it."

Children also enjoyed the chance to utilize the technology involved in the locomotor training. Jackson described that the feedback he received from the treadmill as an element of the program that facilitated engagement and enjoyment,

> "It [the RAGT device] would give you a little boost wouldn't it because you know that if you were at 80 percent, you needed to work harder to get to 90 percent."

The suitability of the structure of the interventions, and the positive engagement families had with the intervention was reinforced by the attendance rates that were observed by therapists, as one described,

> "…they are obviously getting what they want out of the program and they attended every single session which for some families has been a bit shock to us so it's us adjusting our expectation as well."

The Importance of Support, Both Peer and Program

A common theme across parents and therapists involved in the locomotor training intervention was one of support. Particularly important was both peer support and "top-down" support from the program. Support from the program was a crucial element for the families participating in and adhering to the intervention. One parent commented,

> "Sometimes just having that external support and making it more structured, you know you are going to do a bit more."

Parents also described the benefits they received from spending time with families who had similar experiences to them. One parent commented,

> "Being here with other families, it's good for networking with parents, even if it is just for a few minutes."

This was reinforced by therapists, who noticed the benefits families were getting from being involved in the intervention together. One therapist observed,

> "To have a group that is there for 6 weeks interacting with each other...just that social inclusion stuff is really helpful. I feel like it's good for parents to be able to connect with other parents."

Therapists also described the benefits of peer support for themselves, noting that team dynamics were important to the success of the intervention. Additionally, support from the principal investigator in the form of ongoing education and training was key to ensuring they felt confident and comfortable to deliver the intervention as intended. As one therapist described,

> "Just that support I suppose from a senior person like [the principal investigator] or another physiotherapist, to be able to discuss things and programs with them. They will always be there to update things and talk you through it but just having someone there helps."

The Utility of Locomotor Training Beyond Research

Parents and therapists were all able to see the utility of locomotor training beyond the scope of the research project they were involved in. One parent commented,

> "If another block is offered again maybe not even the 6 weeks maybe 2 or 3 weeks or something, I'm sure there would be more parents like me who just want those things happening in the future."

Therapists described the positive benefits they perceived for the families involved in locomotor training. One therapist observed,

> "Parents were so excited I think just seeing their children walk. I never understood what it might be like and now that I have a child and watching her walk, walking was such a milestone. For these parents with children with disability who maybe miss so many milestones compared to siblings and peers to see them walking you would see such joy on their faces, both the children and the parents."

When asked about the continuity of locomotor training beyond the research project, one therapist commented,

> "Yes 100 percent, even just going alone from what parents say and want, let alone what we can see happening… All people want is that maintenance and that its ongoing in the future and they are desperate for it."

Motivations for Participating in Locomotor Training

In this theme, parents, predominantly, along with children, described the motivations for their participation in locomotor training. Meaning units spanned (a) the enjoyment of the child of movement-based activities and (b) wanting to increase the activity levels of their children.

Enjoyment of Movement

Parents described that despite having movement limitations, their children loved having the opportunity to move their bodies. One parent commented,

> "Jordan does love movement, which is a big thing for him. It is probably one of his biggest drivers - is movement. So obviously he can't walk, he has a standing frame and a walking frame and so a walking clinic was just ideal for him."

Parents also described increasing the activity level of their child as a primary motivation for participating in locomotor training, in order to maintain or improve their mobility and/or fitness. On the issue of maintaining or improving mobility, one parent commented,

> "We just want to keep Nathan mobile, get his confidence [up] and build up his endurance and that is our goal...If there are things there to help us do that, we will always try to do those things."

Increasing Activity Level

The motivation to improve or maintain fitness levels was important for parents, as they perceived that this was of benefit to the overall health and well-being of their child. As one parent described,

> "One of the goals here was fitness you know it wasn't therapy. My goal was fitness because I knew it would get her fitter and I knew that was declining and if anything is going to get it will be pneumonia or some nasty bug. So I never saw it as free therapy I saw it as a chance for exercise, a chance for Stephanie to develop her lungs and get a bit fitter."

Jackson cited improved fitness as a motivation for participating in locomotor training. A notable comment by Jackson was,

> "Because when my body is tired I have to carry him in my wheelchair and he is very heavy."

Barriers and Facilitators to Participation in Locomotor Training

Parents and therapists described the (a) environmental and (b) personal factors that affected the implementation and provision of locomotor training.

Environmental Factors

For parents, organizing the attendance of their child at locomotor training was sometimes considered a logistical challenge;

> "I don't know about other mums, but it's stressful."

However, parents also reflected that the provision of parking by the study team, for families involved in the study sessions, helped ease aspects of the logistical burden. One parent commented,

> "I mean it's [hospital where the study took place] far, but you know…having a parking space was really good. Yes that was a godsend. It takes a lot of the stress out of it....you can just get in the car and leave and you don't have to allow and extra 15 min to find parking."

Parents also commented that as their children were most attentive during schooling hours, they would often miss parts of the school day to attend locomotor training. Whilst this was considered a challenge for some families, in most instances, there was a mutual understanding between school and family that locomotor training would be of benefit to the child,

> "So yeah we did miss [school] but I mean the school is quite good about it they understand that it [locomotor training] is important to us."

Personal Factors

From the perspectives of both parents and therapists, relationships between program staff and families were seen as positive influencers on locomotor training adherence. Consistency in service delivery helped to build rapport, as described by one therapist,

> "There isn't stopping and starting, we get to know the clients and how to read them as to when they need a break and what works best for them."

Parents also commented that therapists made them feel comfortable and confident that the needs of their child were met,

> "They were just really encouraging and they were all really understanding and they just know their stuff…I felt she was safe in their hands which is a big deal with a childlike Stephanie."

Relationships with program staff were highly influential on engagement during sessions and enjoyment of the locomotor training. As one parent commented,

> "They made it fun for him so they had his music and they would always give him goals like let's get to 100m then 200m. It suits Jackson's personality that if you give him something, he's got to get there."

Several therapists described the physicality of delivering the locomotor training intervention as a challenge, particularly with regards to the manual handling and positioning of participants. One therapist commented,

> "Yeah it is very energy intensive and physical. We are quite good at managing that, especially with the treadmill. That is the real physical part, we are very good at rotating to work out both sides of your arm and back and give your wrists a break when you are supporting from behind and [PI] is really good at splitting up who is doing too much on one day. I think at the moment we are sometimes doing four a day and that is probably the limit in terms of stress on your body and being bent over in that position the whole time."

However, therapists also explained that the physicality of the intervention ultimately, in their opinions, facilitated positive outcomes for participants. As one therapist reflected,

> "All their walking seems to be a lot easier, they are stronger and our jobs are a lot easier as they progress because they have gotten stronger and they can initiate so much more themselves."

Perceived Outcomes of the Intervention

Four themes were categorized as an outcome of participation in locomotor training. Specifically, as a result of participation in the locomotor training intervention, parents and therapists reported participant outcomes related to (a) physical health, (b) sleep, (c) affect and emotion, and (d) ambulation in daily activities. A description of each theme, and additional meaning units, are presented in the **Supplementary Material**.

Physical Health

Within this theme, parents and therapists highlighted improvements in strength, gross motor function, and overall physical well-being because of the involvement of participants in locomotor training. At the conclusion of the intervention, most of the parents described improvements in the strength of their child. As one parent commented,

> "He got the strength, you know more strength, and he got the confidence, which I think is what he is lacking."

The benefits seen from locomotor training also extended to gross motor activities such as walking. As one parent described,

> "My primary reason for participating was he was going to walk longer, he was going to walk better he was going to be stronger. And all three of those things happened."

Therapists also described further improvements in other gross motor activities, one observed

> "I think the big thing was that for the GMFCS V kids it was having an impact on their rolling and functional mobility which was interesting."

Another commented,

> "And just sitting, their ability to sit upright has improved with a lot of them who initially couldn't sit unsupported and by the end of it some of them have been able to which is really cool."

Parents and therapists noticed that, for participants who were more reliant on wheelchairs throughout the day, the impact of locomotor training was different, relating more acute changes in tone and movement patterns. One parent said,

> "So she started the day with a lot of uncontrolled movements but after her [locomotor training] session her uncontrolled movements reduced significantly. So that's on a hard day, it made it better."

The notion of acute changes in tone were also noted by therapists, as one described,

> "We see changes in tone as well because obviously we are very hands on especially on the treadmill when we are facilitating their stepping you can definitely feel over time the tone changing."

Pre-existing respiratory and digestion issues, commonplace among this population, seemed to be positively impacted by locomotor training. One parent described,

> "My thoughts were previous to the study [she] was getting sick every 2–3 weeks and they [were] always an upper respiratory sickness, she often has antibiotics to get over a sickness and fever. Her breathing at night was becoming more problematic with audible strider-like breathe and mum and dad have to sit up until she goes to bed with her… So all that got better. There has been no sickness so far and no antibiotic use. Previous to this Stephanie has been sick every 3 weeks and has missed school. Her overall temperature seems stable- previous to this it wasn't. She has had no [paracetamol] over this period...it's the walking".

Sleep

Parents reported that participation in the locomotor training contributed to improved sleep quality and duration for their children and, in some instances, themselves. For example, one parent commented,

> "Sleeping through is great for everybody.....So I don't have to get up and I don't have broken sleep, she doesn't have broken sleep it's heaps better."

Parents also reported that the easing of respiratory symptoms also contributed to improved sleep for their children. As one parent described,

> "Better breathing especially at night, constant sleeping through the night with no waking in the middle of the night. No sickness which is hugely significant, and better ability to cough."

Therapist observations mirrored these changes, with regards to the easing of respiratory symptoms and the impact on sleep. For instance, one therapist said,

> "They've been sleeping better, easier to change, the girl that became easier to change didn't really sleep through the night but then after the training was able to sleep through the night... and Mum was over the moon."

Affect and Emotion

Parents and therapists described participants having improvements in mood, confidence, and motivation following their involvement in locomotor training. Regarding mood, parents perceived their children to be generally happier and calmer following their involvement in locomotor training. As one parent said,

> "…he's waking up happy and excited for what the day is going to bring and I think he is a little bit hopeful now that every appointment is going to be a [locomotor training] appointment."

Improvements in mood also extended to parents, as they were able to see the benefits the locomotor training had for their family. As one parent explained,

> "Oh it's made us happy, well we have always celebrated Adam's little achievements we just seem to celebrate them a little more...And the fact that he is faster.… Tensions are down a little more too. You know, first thing in the morning when we're trying to get somewhere, it's just a bit easier."

Participation in locomotor training led to participants feeling more confident in themselves and their abilities, as described by parents and therapists. One therapist commented,

> "A lot [of participants] start out with "I can't do this" or "this is hard I don't want to do this. "By the end of the program it's "I can do this it's amazing look what I did this week" and "I did this at school today." One boy who does cross country, he was like "I did 1.2 km in 9 min today"... Even then he would fall, but he would get back up himself. Mum was in tears and the teachers were in tears.... their perceptions of themselves and what they are able to do is really cool."

This confidence, in turn, seemed to contribute to improvements in the motivation of participants to be involved in activities both within and outside of school. As one parent described,

> "he was faster and stronger and he wanted to get in his walker more so that was cool" and another, "We've seen better performance at school, more alert, and better attention."

This notion was reinforced by therapists, as one therapist commented,

> "What we have heard from parents and kids is that they've been more involved in sports. One of the girls was not involved in any kind of community activity outside of school and now she comes in and she is like "I'm going to sign up for wheelchair basketball and I'm going horse-riding" which is awesome and that's the whole point isn't it."

Ambulation in Daily Activities

In this theme, parents and therapists highlighted the impact that locomotor training had on the ability of participants to be more active and independent throughout the day. Endurance and speed of walking improved the ability of participants to ambulate in the community and at school. As one parent described,

> "Of course, the biggest thing from school is the distance between the bus stop and nappy changing area and his classroom is huge so before they would have to set aside 20 min just to get his nappy changed and back again. Whereas now they probably get it all

> done in about 8 min because he's walking and thinking "I'm doing it." The teacher is happy because she actually gets to spend more time with him. That transition from the bus stop to the classroom, every day he does it he gets faster".

Parents also reported an impact on activities within the home, as one parent said,

> "what I would do is I would get Jordan to walk at home you know little journeys from his bedroom to the lounge room I would get him to walk down the hall and prior to [the locomotor training program] he would occasionally put down one foot and then lifting both feet and basically I would carry him and he would put down one foot but now he is doing two steps most of the time again."

DISCUSSION

This qualitative study describes the experience of intensive locomotor training from the perspective of therapists and parents of children with CP. It was important to describe these experiences because intensive or more highly dosed interventions do require more time commitment from children and their families. By understanding both the experience of and perceived outcomes of locomotor training, the implementation of locomotor training beyond the research context can be shaped to facilitate the translation of research into the "real-world" in children with CP functioning within GMFCS levels III, IV, and V.

The optimum treatment dosage for locomotor training for children with CP is yet to be established. The current literature has reported positive mobility outcomes with treatment dosages that range from 2 to 5 days a week over a period of 2 to 25 weeks (25, 26). The wide range of treatment dosages reported in the literature is challenging for both therapists and parents who need to plan for a more highly dosed intervention. Long treatment durations may potentially be effective but can be expensive, place undue stress on families, and impacts schooling. Therefore, an important question is raised: what is the minimum duration that will facilitate meaningful changes? In this study, therapists described observable mobility changes from week 5. For parents, they reported that they felt ready to finish the program by week 6. Given that the current physical activity dosage recommends a dosage of three times a week for 8 weeks (27), it is likely that short intervention durations will result in minimal meaningful physical changes. Yet, when considering the reported impact of an intensive intervention on families, longer intervention durations (beyond 6 weeks) may be burdensome for children and their parents (28). Although no specific recommendations can be made from this qualitative study about optimum treatment dosage, the reported experiences from this study do highlight the importance of having conversations with families about the balance between optimal treatment dosages, meaningful physical outcomes, and the impact of lengthy appointment schedules on the family unit and education. This qualitative study provides a useful starting point to initiate discussions on balancing all of these factors. An important future research direction would be to determine optimal treatment dosages and to evaluate the cost-effectiveness of more highly dosed interventions in relation to outcomes.

Parents and therapists reported outcomes important to their family and child beyond what is traditionally measured in motor-based interventions. In children and youth with CP, controlled trials have largely focused on activity capacity motor outputs, such as improvements in walking speed (9, 25) and endurance (29). However, reports in this study are consistent with previously reported uncontrolled trials and case series which provided early evidence of outcomes beyond walking speed and endurance which include improved confidence and mood (7), improved weight acceptance and transfers (30), reduced caregiver support, improved skills in gait trainers, and improved bowel function (31). In addition to this, parents provided the context of the perceived improvements that were meaningful to them. For example, improvements in walking endurance were contextualized within the school environment, where improved walking speed and endurance enabled more time in class. Within the home, improvements in walking meant that morning routines were faster, with children being "happier" and "calmer," having a positive impact on family dynamics. Collaborative goal setting is highlighted here, and these parent and therapist experiences provides a much-needed prompt to consider goals that are beyond motor outputs alone.

The value of engaging in physical activity, particularly for children that tend to spend more time sedentary, is reinforced by parents and therapists as they reported outcomes that directly reflect the known effects of physical activity (32, 33). This included improvements in respiratory function which was reported to impact their sleep quantity and quality both for themselves and their child. Improved confidence was also reported, and parents related this to a more positive outlook and greater motivation to try new things to engage in community activities. Furthermore, parents noted that the increased activity levels during the locomotor training period were linked to improvements in mood. The link between activity and mental health is well established with low levels of physical activity being associated with poorer psychological well-being (34), and higher levels of physical activity being associated with improvements in the self-esteem and psychological health of children (34–37). For children with CP classified within GMFCS levels III, IV, and V, locomotor training provides an opportunity to improve and preserve both physical and mental health.

Based on the outcomes of this qualitative study, we recommend that future physical activity or motor-based studies consider the inclusion of children with greater equipment needs (GMFCS levels III, IV, and V) along with the inclusion of outcome measures on sleep quality and quantity, quality of life, respiratory outcomes, participation measures, mental health outcomes, and caregiver support. The breadth of outcomes reported by parents and therapists consolidates the importance of taking a 24-h "whole day matters" approach where the distribution of physical activity, sedentary behavior, and sleep impacts the health and well-being of a child (38). Further to this, quantifying the impact of treatments requires tailoring and the adoption of individualized measures is necessary to objectively and reliably determine meaningful activity and participation outcomes. Measures such as the Canadian

Occupational Performance Measure and the Goal Attainment Scale are particularly relevant and are likely to reflect specific and meaningful changes for the child and their family within a "whole day matters" approach (39).

There are some practical considerations that can be considered for future research and intervention provision. Parents and therapists both agreed accessibility (such as parking accessibility and scheduled session times) to the intervention are important considerations. Enjoyment or fun should be considered for both children during the sessions and for parents who may benefit from the company of other parents of children with CP. Parent-to-parent peer support provides opportunities for parents to share and explore their feelings and experiences with others who have similar journeys (40). Finally, for therapists providing the intervention, support and training are integral to optimize treatment fidelity and ensure co-ordinated care for children with higher physical needs.

The current study has several strengths which include a qualitative design with robust methods to enhance trustworthiness. The interview guide was developed with family engagement through a pilot study that was conducted prior to this qualitative study. The inclusion of therapists who were involved in providing the intervention, along with one child classified within GMFCS IV who was able to participate verbally, provided a different perspective. This research also provides insights from those involved in the provision of high dosed interventions for children with high physical assistance and equipment needs.

The main limitation is that only two children were interviewed, one of which was a child that used AAC and tended to nod in agreeance when his mother spoke about her experiences rather than answer questions directly about his own experiences. Gaining more perspectives of children and youth on their experiences, particularly for a more highly dosed intervention, would be an important future direction because it has the potential to influence knowledge translation of interventions in the community. Future studies should also consider designing specific questions for children using AAC and, where possible, conducting the interview without their parents in the room. However, we acknowledge that this may be challenging for children particularly if they are classified within GMFCS levels IV and V. Another limitation is that the timing of the interview was captured following the intervention which means any outcomes of retention were not obtained. Generalisability is therefore impacted as a result as is the fact that this study was only limited to one site. Finally, another limitation is that the effect of the intervention on schooling was not specifically explored. Given both the intensity of such a program and the uncertainty around recommended intervention dosages, exploring the impact of such a program on educational experiences is an important future direction.

CONCLUSION

From the perspectives of parents and therapists, locomotor training provided with a high dosage over 6 weeks provided children with CP classified as functioning at GMFCS levels III, IV, and V an opportunity to engage in physical activity. Engaging in physical activity was a consistent motivator, and improvements in physical health, sleep, affect, and emotion and ambulation in daily activities were meaningful outcomes for parents. Future studies are needed to determine the optimal dosage required for optimal outcomes and the inclusion of a broader range of health and well-being outcomes alongside individualized measures on activity and participation are needed to better understand and quantify meaningful changes for the child beyond motor-based outcomes.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Curtin University Human Ethics Research Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

DP and CE planned the iStride trial, wrote the protocol, obtained funding, and oversaw the study. CW conducted the interviews with all participants. DP and AT carried out the initial analysis of the manuscript with CW being the critical friend after the initial analysis. DP and AT drafted the manuscript and revised it in response to revisions from all authors. All authors have read and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2021.740426/full#supplementary-material

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Cognitive and Linguistic Benefits of Aerobic Exercise: A State-of-the-Art Systematic Review of the Stroke Literature

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This systematic review aimed to determine how aerobic exercise affects cognition after stroke, with particular focus on aphasia and language improvement. Methodological quality was assessed with the PEDro+ scale with half of the 27 included studies rated as high quality. Data extraction focused on cognitive effects of aerobic exercise post-stroke, intervention characteristics, outcome measures, and participant characteristics. Whereas attention, memory, and executive functioning measures were common across the included studies, no study included a language-specific, performance-based measure. Seventeen studies reported positive cognitive effects, most frequently in the domains of attention, memory and executive functioning. Variability in outcome measures, intervention characteristics, and participant characteristics made it difficult to identify similarities among studies reporting positive cognitive effects of exercise or among those studies reporting null outcomes. Only three studies provided specific information about the number of individuals with aphasia included or excluded, who comprise approximately one-third of the stroke population. The review identified patent gaps in our understanding of how aerobic exercise may affect not only the cognitive domain of language post-stroke but also the broader cognitive functioning of individuals with post-stroke aphasia. Methodological limitations of the reviewed studies also warrant further examination of the direct impact of aerobic exercise on cognition post-stroke with careful attention to the selection and reporting of population, intervention, and outcomes.

Keywords: stroke, aphasia, aerobic exercise, cognition, language, rehabilitation

INTRODUCTION

Language impairments are a common cognitive consequence of stroke and contribute to shrinking social networks and decreased quality of life (1, 2). Cognitive impairments, including language impairments, may also impact the ability to participate in and benefit from rehabilitation (3, 4). While behavioral interventions are the conventional, evidence-based approach to treating stroke-related cognitive deficits [e.g., (5)], they require considerable time and practice, and patients are often discharged prior to resolution of these difficulties (6). Accordingly, there has been growing empirical interest in adjuvant therapies, including exercise[1], that can maximize behavioral intervention outcomes (10–15). The focus of the current systematic review is whether aerobic exercise alone affects language and other cognitive outcomes following stroke, which will help ascertain its potential effectiveness as an adjuvant to behavioral therapy in this population.

Several lines of evidence suggest that physical activity holds merit as a means to ameliorate post-stroke language and cognitive sequelae. First, the physical and mental health benefits (e.g., increased strength, reduced falls, mood enhancement, better cardiovascular health) associated with physical activity are well-established in typical and atypical aging populations (16–20). Though there are challenges in introducing aerobic exercise to stroke survivors (21), positive effects of physical activity on the physical sequelae of stroke (e.g., hemiparesis) have strong empirical support, and accordingly, exercise (e.g., range of motion exercises, strength training) is a recommended approach in several stroke management guidelines [e.g., (22, 23)]. Second, although results have been mixed depending on study design and choice of outcome measures (24, 25), it is generally recognized that physical activity is beneficial for cognitive performance in older adults who are healthy or experiencing cognitive decline, with recent data demonstrating an inverse relationship between physical activity and risk of cognitive decline (16, 26, 27). Further, increased cardiovascular fitness associated with physical activity, specifically aerobic exercise, has been shown to positively affect cognitive function in studies of healthy and cognitively-impaired individuals [e.g., (26, 28)]. Third, neural changes associated with repeated or long-term aerobic exercise include increased concentration of neurotrophic and growth factors (e.g., BDNF), which can induce cellular changes such as creation of glial cells, neurons, synapses, and blood vessels (16, 29, 30). These cellular changes allow for structural enhancements such as increased perfusion and gray/white matter volume, and in turn, result in increased brain activation and functional connectivity. Collectively, these molecular, cellular, structural, and functional changes support improved cognitive and motor function. Finally, in animal models, aerobic exercise has been shown to promote functional recovery following neurologic injury [see (30) for a review].

It follows that aerobic exercise would benefit cognitive functioning following stroke; accordingly, recent systematic reviews have concluded that aerobic exercise may enhance cognition in the stroke population [e.g., (30–36)]. However, most advocate for further examination of the effects of exercise on post-stroke cognitive abilities, identifying methodological issues and inconsistent outcomes across studies. Importantly, the focus and methods of previous systematic reviews have varied, with no particular attention given to the cognitive domain of language. Considering that language difficulties are a common consequence of stroke, addressing this gap in knowledge has immediate clinical applicability and thus language outcomes are examined in the current review.

Related to the limited examination of language abilities as a cognitive domain that may be responsive to exercise, there is a concern as to whether stroke survivors with aphasia have been represented in the extant literature. For example, the word "aphasia" was absent in previous systematic reviews of exercise and cognition post-stroke, [e.g., (35–37)]. This is surprising, given that approximately one-third of stroke survivors are living with aphasia (38, 39) and experience changes with both language and other cognitive abilities (40). Understanding the impact of aerobic exercise on individuals both with and without aphasia post-stroke is vital to informing rehabilitation for stroke survivors, their caregivers, and rehabilitation professionals, particularly speech-language pathologists who address language and other cognitive abilities.

The overall objective of the Aphasia Writing Group, a subset of the Evidence-Based Clinical Research Committee of the Academy of Neurological Communication Disorders and Sciences, was to conduct a systematic review to examine what is currently known about the utility of aerobic exercise for improving cognitive abilities, including language, in individuals affected by stroke and stroke-related aphasia. The specific aims were to:

- Characterize how aerobic exercise affects different areas of cognition after stroke, paying particular attention to language outcomes.
- Characterize commonalities and differences across studies with positive vs. null cognitive outcomes following aerobic intervention, including outcome measures, participant characteristics, and intervention characteristics.
- Ascertain the representation of persons with aphasia (PWA) in this domain of the stroke literature.

METHODS

Search Strategy

A comprehensive and systematic literature search was conducted from 2008 through September 2020 to gather state-of-the-art information. Following consultation of librarians from the home universities of the authors of this manuscript, the following databases were searched: Web of Science, CINAHL, PubMed, Medline, ProQuest, PsycInfo, COMDisDome, SpeechBite, ASHAWire, and Scopus. Reference lists of included studies were reviewed to identify any studies that did not emerge from the databases search. The search targeted peer-reviewed, clinical trials (i.e., a study aimed at evaluating an intervention) reporting use of an aerobic exercise intervention post-stroke to improve cognitive outcomes. Specifically, inclusion criteria

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[1] For the purposes of this paper, we will use the following definitions for physical activity, exercise, aerobic exercise, and strength/resistance training. We attempt to preserve terms used by individual studies, but use a more general term when different terms are used across studies.

Physical activity: movement to increase energy expenditure above resting level (7).

Exercise: prearranged, regulated and repetitive physical activity (7).

Aerobic exercise: "exercises [that] demand oxygen without producing an intolerable oxygen debt, so they can be continued for long periods" [(8), p. 39].

Strength/resistance training: "a form of periodic exercise whereby external weights provide progressive overload to skeletal muscles in order to make them stronger" [(9), p. 208].

included the following: full text peer-reviewed journal article in English; participants must have had a stroke and be 18 years or older; the intervention must be aerobic exercise without language or cognitive training; and outcomes must include language and/or other cognitive domains. Studies that included language or cognitive training along with aerobic exercise were excluded in order to evaluate the direct impact of aerobic exercise on cognition. Gray literature and non-experimental publications (e.g., reviews) were excluded. The Preferred Reporting Items for Systematic reviews and Meta-Analysis Guidelines [PRISMA; (41)] were followed. Search terms reflected categories of population (e.g., stroke, aphasia), intervention (e.g., aerobic exercise), and outcome (e.g., cognition, language). Terms within or across a category were combined via "OR" or "AND," respectively (see **Supplementary Table 1**). This search yielded 7,771 articles after 4,444 duplicates were removed (**Figure 1**). Deduplication and screening were performed *via* Covidence, which uses PRISMA guidelines by default (https://www.covidence.org/).

The authors conducted a title and abstract review to manually scan for the inclusionary criteria of: original data from adults with stroke/CVA and no other neurological diagnoses, inclusion of an intervention group receiving aerobic exercise only, and measuring an outcome related to language and/or other cognitive domains. To examine reliability for this screening process, a randomly selected sample of 300 consecutive titles and abstracts was independently screened by one of the authors (LM). Inter-rater agreement was 98.7%, with disagreements resolved *via* discussion among all authors. A full-text review of each of the remaining 145 articles was then completed independently by two authors, resulting in 27 articles for inclusion. Inter-rater agreement for full-text review was 90.4% with disagreements resolved via discussion among all authors. **Table 1** lists the 27 studies and the type of design of each study, which consisted of randomized and non-randomized clinical trials.

Methodological Quality Review

Two reviewers independently performed a quality assessment of each included article. Inter-rater agreement prior to consensus was 87%. The quality assessment involved a "risk of bias comparison" based on the Physiotherapy Evidence Database Plus (PEDro+) Scale (69), which includes 13 criterion items with a maximum score of 12 (see **Supplementary Table 2** for a full description of the PEDro+). The PEDro+ scale features two additional criteria that are critical elements of behavioral treatment (Treatment Fidelity and Treatment Replicability), which are not present on the original PEDro scale. The PEDro and PEDro+ scales are commonly used in the stroke rehabilitation literature to evaluate the methodological quality of randomized controlled trials; however, these scales are also

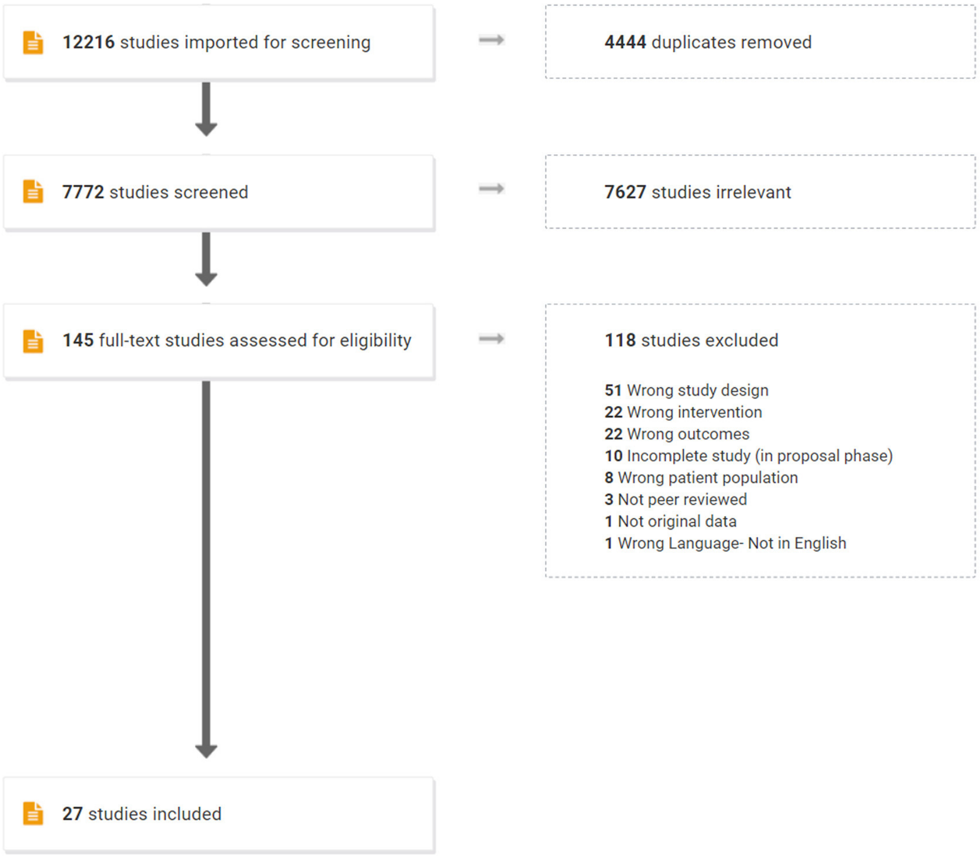

FIGURE 1 | Prisma diagram generated from Covidence.

TABLE 1 | Design and PEDro+ Score of included studies.

Study#	References	Design	PEDro+ Score (max = 12)
1	*Bo et al. (42)	RCT	8
2	Chan and Tsang (43)	RCT	8
3	*Colledge et al. (44)	non-randomized controlled trial	4
4	* El-Tamawy et al. (45)	case-control study	6
5	*Ezeugwu and Manns (46)	prospective cohort	5
6	*Ihle-Hansen et al. (47)	RCT	7
7	*Khattab et al. (48)	RCT	9
8	*Kim and Yim (49)	RCT	7
9	*Kluding et al. (50)	prospective cohort	3
10	Krawcyk et al. (51)	RCT	8
11	*Lee et al. (52)	prospective cohort	4
12	Macko et al. (53)	prospective cohort	4
13	*Marzolini et al. (54)	prospective cohort	4
14	*McDonnell et al., (34, 55)	RCT	5
15	Meester et al. (56)	RCT	6
16	*Moore et al. (57)	RCT	9
17	*Moriya et al. (58)	prospective cohort	3
18	Pallesen et al. (59)	RCT	8
19	Ploughman et al. (60)	RCT crossover	6
20	Ploughman et al. (61)	RCT	10
21	Quaney et al. (62)	RCT	7
22	*Rosenfeldt et al. (63)	RCT	8
23	Stuart et al. (64)	non-randomized controlled trial	8
24	*Swatridge et al. (65)	interrupted time series	5
25	Tang et al. (66)	RCT	8
26	* Unibaso-Markaida et al. (67)	non-randomized controlled trial	5
27	*Yoo and Yoo (68)	RCT	9

Studies that showed positive cognitive effects of aerobic exercise. RCT, randomized controlled trial. The study number in this table is used to identify these articles in the remaining tables.

often used to evaluate other types of clinical trials, including non-randomized controlled trials and case series research designs (69–74). For these reasons, the PEDro+ scale was deemed a valid and appropriate appraisal tool to evaluate the 27 clinical trials reviewed in this study.

Data Extraction

Data related to the specific aims were extracted from each article by two reviewers. Inter-rater agreement prior to consensus was 96%. These data included specifics on study design, participant characteristics (e.g., age, time, post-stroke onset), exercise intervention (e.g., type, frequency), cognitive outcome measures (e.g. self-reported, performance-based), and results pertaining to those outcome measures.

Identification of Outcome Measures and Cognitive Domains Tested

The language and other cognitive outcome measures used in each study were collated based on whether they were self-reported or quantitative/performance-based and what domain(s) of cognition they measured. **Supplementary Table 4** lists each outcome measure, its cognitive domain(s), and each study that included the measure. The self-report measures—SIS (75) and Stroke Specific Quality of Life Scale [SS-QOL; (76)]—both include domains related to communication and thinking and were thus included in the counts as measures of language and executive function.

RESULTS AND DISCUSSION

Methodological Quality

None of the 27 included studies received the maximum score of 12 on the PEDro+ scale (69). Across studies (**Table 1**), total scores ranged from 3 [high bias for nine items, e.g., (50)] to 10 [high bias for two items, e.g., (61)] with an average score of 6.39. It should be noted that if a PEDro+ item was not explicitly addressed, then it received a high bias rating and "0" score. Therefore, scores might reflect the report available to the reader rather than the procedures implemented by the authors. See **Supplementary Table 3** for PEDro+ scale item-level detail per study.

Fourteen of the 27 studies received a low bias judgment on more than half of PEDro+ items, suggesting they were of high quality (69). However, as shown in **Figure 2**, there was variation in terms of which PEDro+ items received low bias ratings across studies. High bias judgments were common for Allocation Concealment, Treatment Fidelity, and Blinding of Assessors, Subjects, and Therapists, with all studies lacking Therapist Blinding. De Morton (71) reported that Therapist and Subject Blinding criteria had the least adherence in her Rasch analysis of 200 clinical trials of various intervention types that were randomly selected from the PEDro database (www.pedro.org.au). De Morton explained blinding is often difficult or not possible in many clinical intervention trials. This logistical dilemma likely applied to some of the studies included in this review and should be considered when evaluating the quality scores.

Notably, studies with only one exercise group cannot satisfy the Therapist Blinding, Subject Blinding, nor Allocation Concealment criteria due to the nature of their design. Therefore, the seven single group studies lacking a comparison or control group received lower PEDro+ scores, highlighting these types of clinical trials are not as methodologically rigorous compared to clinical trials with random assignment of multiple groups. However, it is important to note that the PEDro+ score does not indicate study meaningfulness, treatment effect size, or generalizability (69), and studies with lower PEDro+ scores might perform well in those areas.

The Assessor Blinding and Treatment Fidelity PEDro+ items were applicable to all study designs included in this review, and the low reporting incidence of these items suggests these criteria warrant more careful consideration when designing and publishing future investigations. Assessor blinding is critical to obtain unbiased performance on outcome measures pre- and post-treatment, and reporting of treatment fidelity is essential for confirming valid, reliable treatment implementation.

Similar to previous systematic reviews focused on exercise and cognition after stroke [e.g., (34, 37, 77)], the quality of papers

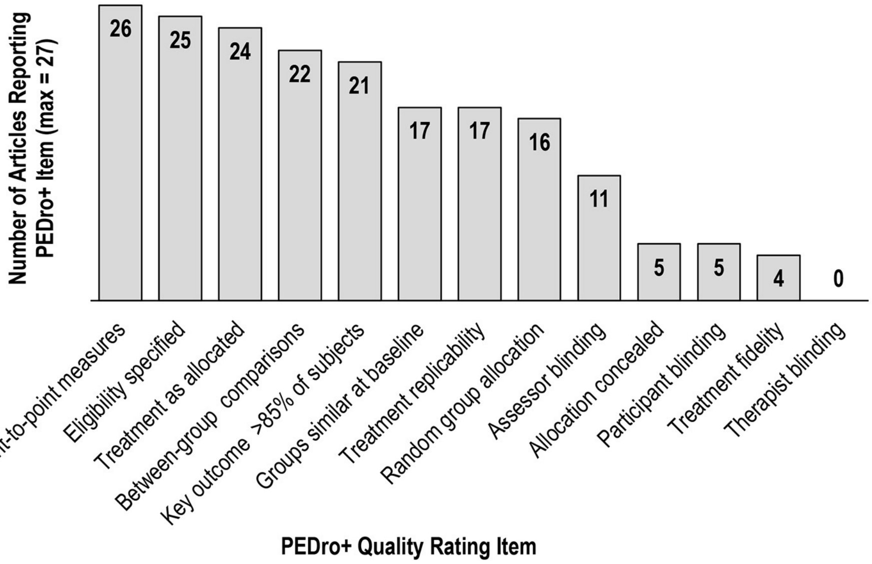

FIGURE 2 | PEDro+ Scale item reporting.

included in the current review was wide ranging, indicating more rigorous study design and reporting are needed to further strengthen this area of the stroke literature. Given the variation in methodological rigor and consistent under-reporting on several PEDro+ items across studies, results pertaining to the cognitive benefits of exercise subsequent to stroke should be interpreted with caution.

Outcome Measures and Cognitive Domains

Although cognition was not the primary focus of nearly half of the 27 included studies [which instead prioritized a physical outcome; e.g., (46, 51, 52, 57)], each reviewed study included some measure of cognition and was thus deemed appropriate for data extraction. A description of the cognitive outcome measures found in the studies reviewed is listed in **Supplementary Table 4**. An illustration of the inclusion of cognitive measures across studies and whether or not an effect was noted is included in **Figure 3**.

Overall, seven studies included a self-report measure and 23 included at least one performance-based cognitive measure. Fifteen studies included at least one measure of language, 20 included at least one measure of attention, 19 included at least one measure of memory, 20 included at least one measure of executive function, and 12 included at least one measure of visuospatial skills. Four studies used only a self-report measure of cognition [SIS, SS-QOL: (53, 63, 64, 68)]. Nine studies utilized cognitive screeners (MMSE, MoCA, or ACE-R), which offer a cursory assessment of a broad range of cognitive functions; two of these studies reported on specific domains tested by the cognitive screener (51, 56). Two studies that used a cognitive screener also included a self-report measure [SIS: (46, 57)], and two included additional tests of more specific cognitive abilities [TMT A and B: (47, 49) and Stroop Color and Word Test (SCWT): (49)]. Sixteen studies included performance-based tests of specific cognitive abilities, five of which focused specifically on one cognitive domain (43, 44, 58, 61, 65). Six studies specifically reported the results from language subtests of screeners or self-report measures (50, 51, 53, 56, 59, 64). No study used a language-specific, performance-based measure. Seven studies quantitatively measured all of the following cognitive domains: attention, memory, executive functioning, visuospatial skills, and language. Of these, three reported the effect of exercise on each domain separately (51, 56, 59).

Interestingly, no study used a full battery of standardized cognitive testing, even in studies focused specifically on cognitive outcomes of aerobic exercise after stroke. Relatedly, the rationale for the selection of cognitive measure(s) was infrequently provided. Relevant to our particular interest in language outcomes, no study used a performance-based measure that focused solely on evaluating language abilities, with language assessment being primarily limited to brief spoken naming tasks. Therefore, whether aerobic exercise can positively influence the broad range of language abilities that may be compromised by stroke, particularly those abilities enmeshed in daily communication activities (e.g., discourse comprehension; grammatical skills germane to spoken or written sentence formulation), has not yet been examined.

Aim 1: Effect of Aerobic Exercise on Cognition

As shown in **Tables 2** and **3**, of the 27 studies reviewed, 10 reported no statistically significant improvement in any cognitive domain. Of the 17 studies that showed improvement in at least one cognitive domain, only two reported a significant increase in language as measured *via* the communication

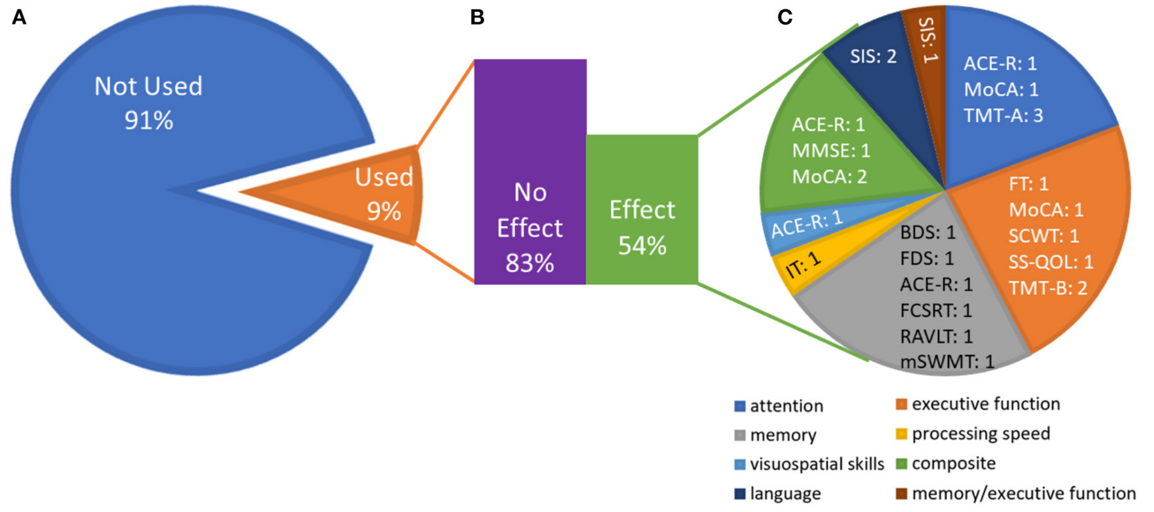

FIGURE 3 | Effects broken down by outcome measures and cognitive domains. **(A)** Proportion of studies that utilized the outcome measures identified in this review, averaged across outcome measures (e.g., the Stroop task was included in 6 of the 27 studies). This emphasizes the variability in the outcome measures chosen across studies. **(B)** Average (across outcome measures) of the proportion of studies that used the outcome measure and showed an effect vs. no effect (e.g., among the six studies using the Stroop task, five showed no effect). The numbers do not add up to 100% because many measures were only reported in one study. **(C)** Domains of outcome measures that showed an effect. Each domain contains measures that contribute to that domain and the number of studies that showed an effect on that measure. The "composite" domain represents effects for which only the composite score on cognitive screeners was reported. See **Supplementary Table 4** for test abbreviations.

domain of the SIS (46, 63). Six studies reported a significant increase in memory (44, 45, 50, 58, 59, 67), five reported a significant increase in executive function (42, 49, 54, 65, 67), five reported a significant increase in attention (47, 49, 54, 67), and one reported a significant increase in visuospatial skills (45). Additionally, one study reported improvement on the memory/thinking portion of the SIS (50), and one study reported improvement on the thinking portion of the SS-QOL (68). Four studies only reported the overall score on a broad cognitive screener (46, 49, 52, 57), making it difficult to determine which specific cognitive functions improved in these studies. Notably, no study identified a significant decrease in specific or general cognitive abilities subsequent to aerobic exercise.

It is difficult to derive strong conclusions regarding the specific cognitive effects of aerobic exercise for stroke survivors for the following reasons: (a) the heterogeneity of domain-specific measures, (b) the use of cognitive screeners—which may not be suitable for identifying nor measuring change in single domain strengths and weaknesses (78, 79), (c) the use of self-report as the sole measure of cognitive outcome, and (d) the reliance on subtests of cognitive screeners and self-report measures to assess language abilities. This is not entirely unexpected given that cognitive outcomes were often not the focus of the studies. However, the modest cognitive effects in concert with methodological variability and study quality concerns as identified in the current review, were previously reported by Zheng et al. (37), indicating little progress in this aspect of the literature in the past 5 years. Thus, there is a clear opportunity for future studies to address these concerns.

Aim 2: Commonalities Across Studies

Substantial variability in outcome measures, and both variability and reporting issues regarding participant profiles and intervention characteristics across studies made it difficult to identify similarities among the studies showing positive vs. null cognitive effects of exercise. With respect to cognitive outcome measures, the TMT was most commonly used; yet, just 2 of the 10 studies that utilized this measure reported statistically significant improvement (42, 67). This either suggests that the TMT B is not sensitive to the effects of aerobic exercise, or that aerobic exercise does not improve executive function, though other measures of executive function did show improvement across studies (see section Aim 1: Effect of Aerobic Exercise on Cognition and **Figure 3**). The next most common measures were utilized in 5-6 studies and included the TMT A, forward and backward digit span, the Stroop task, Stroke Impact Scale, and the MoCA. Of these, the only measure that showed more positive than null effects among studies that used the same measures was the MoCA, with three studies (out of five) reporting improvements (46, 49, 54). Again, this either suggests that the TMT A, forward and backward digit span, the Stroop task, and the Stroke Impact Scale are not sensitive to the effects of aerobic exercise or that exercise does not improve attention, short term memory, working memory, inhibition, and self-reported cognition and communication, respectively. However, the MoCA, and other measures of attention, memory, and communication, did show improvement across studies (see section Aim 1: Effect of Aerobic Exercise on Cognition and **Figure 3**). Notably, only Marzolini et al. (54) specifically reported whether the MoCA improvement was meaningful

TABLE 2 | Participant characteristics in reviewed studies.

Study#	N	Age M	Sex M/F	TPO M (SD) in months	Stroke characteristics
Studies with positive cognitive effects					
1	42	65.12	23/19	>6	NR
3	15	57.3	4/11	44 (28.9)	SAH; 4 RMCA, 3 LMCA, 3 RpComm, 3 LaComm, 1 RaComm
4	15	48.4	NR	3-18	anterior ischemic stroke
5	34	64.6	19/15	3.5 (1.1)	19 RH, 15 LH; 26 ischemic, 8 hemorrhagic
6	177	71.4	99/78	NR	163 infarction; 14 hemorrhage
7	22	65.9	13/9	39.6 (48.0)	15 RH, 10 LH; 3 lacunar, 7 ischaemic, 9 hemorrhagic, 6 unknown
8	14	50.71	9/5	12.79 (7.34)	NR
9	9	63.7	5/4	50.4 (37.9)	3 RH, 6 LH
11	28	56.74	all M	26.04 (19.08)	8 RH, 20 LH; all ischemic
13	41	63.6	30/11	>10 weeks	65.9% ischemic
14	17	70.1	12/5	106.8 (CI = 67.2, 146.4)	52% LH; 82% ischemic
16	20	68	18/2	21 (37)	10 RH, 9 LH, 1 bilateral; 1 hemorrhagic; 5 cortical, 4 BG, 2 cerebellar, 7 subcortical other, 2 unknown
17	11	69.6	7/4	NR	5 ischemic, 6 hemorrhagic
22	32	55.5	22/10	14 (average median)	NR
24	9	57.8	6/3	37.6 (23)	7 RH, 2 LH; 6 ischemic, 1 ischemic and hemorrhagic (2 unreported)
26	15	58.43	10/5	1-12	11 RH, 4 LH; 10 ischemic, 5 hemorrhagic
27	28	60.86	17/11	66.6 (21.6)	17 RH, 11 LH
Studies with null cognitive effects					
2	17	62.7	10/7	93.6 (73.2)	7 RH, 10 LH; 11 ischemic, 5 hemorrhagic, 1 both
10	31	63.7	23/8	NR	19 RH, 12 LH; lacunar: 17 BG, 9 thalamus
12	20	70	9/11	56 (22)	NR
*15	21	62.25	11/10	25.71 (32.70)	13 RH, 6 LH, 5 midbrain; 13 ischemic, 10 hemorrhagic, 1 both
18	16	55	9/7	NR	6 RH, 8 LH, 2 (unknown); 13 CVA, 3 SAH
19	21	61.4	13/8	20.1 (14.6)	19 ischemic, 2 hemorrhagic; 6 cortical, 11 subcortical, 3 both, 1 brainstem
*20	13	58.4	9/4	36.0 (53.4)	6 RH, 6 LH, 1 bilateral; 12 ischemic, 1 hemorrhagic
21	19	64.1	10/9	55.44 (38.52)	all ischemic
23	40	66.6	25/15	50.4 (9.6)	26 RH, 14 LH
25	25	66	14/11	42 (range: 26.4-80.4)	15 RH, 10 LH; 3 lacunar, 7 ischemic, 9 hemorrhagic, 6 unknown; 4 cortical, 7 subcortical, 5 brainstem, 9 unknown

**Studies that included PWA; NR, not reported; RH, right hemisphere; LH, left hemisphere; CVA, cerebrovascular accident; PWA, persons with aphasia; SAH, subarachnoid hemorrhage; BG, basal ganglia; aComm, anterior communicating artery; pComm, posterior communicating artery.*

[i.e., performance changed from impaired to unimpaired; (80)], which is important, given that the purpose of the MoCA is solely to detect the presence or absence of cognitive impairment (81).

With respect to participant characteristics, range of average participant age for studies reporting positive effects of aerobic exercise on cognition was 48-70 years, whereas the range for studies reporting null effects was 58-70 years; the average age across participants appeared similar in the positive and null outcomes studies (61.6 vs. 63 years, respectively). Regarding time post-onset (TPO), studies with positive effects had a range of 1-66 months, while those with null effects all included individuals who were at least 2 years TPO (note two studies in each of the positive and null outcomes categories failed to include any TPO information). While spontaneous recovery could have contributed to the gains seen in the 17 studies with positive effects, 11 of these studies solely included individuals more than 2 years TPO, suggesting that aerobic exercise is associated with cognitive change in the chronic stage (i.e., more than 6 months

TABLE 3 | Intervention characteristics in studies reviewed.

Study#	Group vs. individual	Exercise type	Intensity	Intensity definition	Activity	Supervision	Session duration x frequency	Total min/week	Program duration
Studies with positive cognitive effects									
1	Group	Aerobic + Resistance	Moderate	BPES 13-15	jogging, cycling, strength, balance	Supervised	50 m × 3 d/w	150	12 w
3	Individual	Aerobic	Moderate to High	55-85% HRmax	walking	Both	30-45 m × 3-5 d/w	90-225	12 w
4	Group	Aerobic + Resistance	NR	NA	PT + cycling	Supervised	30 m PT + 45 m aerobic × 3 d/w	135	8 w
5	Individual	Aerobic	NR	6,000+ steps/day	STUFFS, walking	Unsupervised	NR	NR	8 w
6	Individual	Aerobic	Goal: High	Self-report	individualized: any physical activity 30 m/d + "vigorous" activity 2-3 d/w	Unsupervised	30 m/d active + 45-60 m vigorous × 2-3 d/w	90-180	72 w
7	Group	Aerobic	Moderate to High	40-80% HRmax	walking, cycling, marching, sit-to-stand, platform steppers	Supervised	60 m × 3 d/w	180	24 w
8	NR	Aerobic + Resistance	NR	NA	handgrip exercise + walking (treadmill)	Supervised	15 m strength + 20 m walking × 5 d/w	100	6 w
9	Group	Aerobic + Resistance	Light to Moderate	50% VO2max	TBRS + resistance training (bands) of lower extremities	Supervised	60 m × 3 d/w	180	12 w
11	Group	Aerobic + Resistance	Moderate	60% VO2reserve	cycling + strength	Supervised	55 m aerobic × 3 d/w + strength × 2 d/w	165	24 w
13	Both	Aerobic + Resistance	Moderate	40-70% HRreserve/ VO2max; BPES 11-16	walking, stationary recumbent, upright cycling	Both	20-60 m × 5 d/w	100-300	24 w
14	Group	Aerobic + Resistance	NR	NA	aerobic circuit class + resistance training	Supervised	60 m × 1-2 d/w	60-120	24 w
16	Group	Aerobic + Resistance	Light to Moderate	40-80% HRmax	FAME: strength, balance, walking/jogging, marching	Supervised	45-60 m × 3 d/w	135-180	19 w
17	NR	Aerobic	Moderate	40% VO2max	Ergometer	Supervised	15 m × 1 session	NA	NA
22	Group	Aerobic + Resistance	Moderate to High	60-80% HRreserve	recumbent cycling + UE-RTP	Supervised	45 m × 24 sessions	NR	NR
24	NR	Aerobic	Moderate	45-55% HRreserve	semi-recumbent stepper	Supervised	20 m × 1 session	NA	NA
26	Group	Aerobic	NR	NA	Nintendo Wii Sports Resort: archery, tennis, golf, bowling, cycling, air sports	Supervised	30 m × 3 d/w	90	8 w
27	Group	Aerobic + Resistance	NR	NA	walking + functional UE tasks (theraband, armcycle, lifting weighted box)	Both	90 m × 3 d/w	270*	24 w

(Continued)

TABLE 3 | Continued

Study#	Group vs. individual	Exercise type	Intensity	Intensity definition	Activity	Supervision	Session duration x frequency	Total min/week	Program duration
Studies with null cognitive effects									
2	Group	Aerobic + Resistance	NR	NA	walking + seated stretching and strengthening	Supervised	60 m × 2 d/w	120	12 w
10	Individual	Aerobic	High	77-93% HRmax; BPES 14-16	Home-based HIIT, self-chosen exercise modality; provided stationary bike if needed	Unsupervised	3 × 3 m + 2 m "active recovery," 5 d/w	55	12 w
12	Group	Aerobic	NR	NA	walking with handrail support, weight-shifting, ROM	Supervised	60 m × 2 d/w	120	8 w
15	Individual	Aerobic	Moderate to high	55-85% HRmax	treadmill walking	Supervised	45 m × 2 d/w	90	10 w
18	Group	Aerobic	Moderate to high	>70% HRmax	cycling, platform stepper, sit-to-stand	Supervised	50 m × 2 d/w	100	4 w
19	NR	Aerobic	Moderate	70% HRmax	treadmill walking	Supervised	20 m × 2 sessions	NR	NR
20	Group	Aerobic	Light to moderate	60% VO2max	treadmill walking	Supervised	60 m × 3 d/w	180	10 w
21	Group	Aerobic	Moderate	70% HRmax	stationary bike	Supervised	45 m × 3 d/w	135	8 w
23	Group	Aerobic + Resistance	NR	NA	APA: walking, strength, balance	Supervised	60 m × 3 d/w	180	24 w
25	Group	Aerobic	Moderate to high	40-80% HRreserve	individualized aerobic training (modality NR)	Supervised	60 m × 3 d/w	180	24 w

NR, not reported; NA, not applicable; BPES, Borg Perceived Exertion Scale; STUFFS, Stand up Frequently from Stroke; PT, physical therapy; TBRS, Total Body Recumbent Stepper; FAME, Fitness and Mobility Exercise Program; UE-RTP, upper extremity repetitive task practice; HIIT, High Intensity Interval Training; ROM, range of motion; APA, Adaptive Physical Activity stroke program; HRmax, maximum heart rate; HRreserve, heart rate reserve (difference between maximum and resting heart rate); VO2max, maximal oxygen uptake; VO2reserve, oxygen uptake reserve (difference between maximum and resting oxygen uptake); d/w, days per week; m, minutes.

post-stroke). Nonetheless, TPO may be an important factor to examine in future work.

Regarding intervention characteristics, across all studies, exercise intensity (i.e., level of effort) and modality (e.g., cycling, walking, stepping) were highly variable, with no discernable pattern differentiating studies reporting positive vs. null cognitive outcomes. Of note, five positive and three null outcome studies failed to report any information regarding exercise intensity, and the remaining studies defined and monitored intensity levels in a variety of different manners (e.g., relative to maximal heart rate, maximal oxygen consumption, or rating of perceived exertion), mirroring a general lack of consensus across the stroke rehabilitation literature (82). Likewise, no patterns emerged for studies reporting group vs. individual exercise programs, nor supervised vs. unsupervised exercise sessions, supporting the idea that the beneficial effects of exercise are dissociated from the social engagement inherent in group settings [e.g., (83)]. Visual inspection of the studies reporting positive cognitive outcomes revealed that a substantial proportion (60 vs. 20% of null outcome studies) utilized a combination of aerobic exercise and strength training, consistent with evidence in the healthy aging literature of a positive and possibly synergistic effect of aerobic exercise combined with strength training regimens (83–85). Additionally, those studies with positive cognitive outcomes tended to provide exercise programs more frequently (3-5 days/week) and for a longer overall duration (at least half lasting for longer than 19 weeks), compared to studies with null effects. Indeed, it has been suggested that fitness programs for older adults are more likely to engender positive cognitive effects when implemented for 6 or more months (66, 86), leading some to conclude that cognitive changes following exercise programs are time- rather than intensity-dependent, particularly for stroke survivors (85).

Taken together, our search for commonalities across studies to guide future incorporation of aerobic exercise into post-stroke and aphasia management yielded little definitive information due to a combination of factors, including a diversity of cognitive assessment measures and lack of consideration of clinically vs. statistically significant change across studies, and underreporting of participant and intervention characteristics. Although trends supporting possible best practices for evoking cognitive effects were found (e.g., pairing aerobic exercise with resistance training, providing intervention 3 or more days per week), these patterns were not robust nor detailed enough to guide clinical practice without further study. Enhanced reporting of exercise interventions, including greater detail regarding protocol and dosage, in future work will allow for better comparison across studies and replication to validate findings.

Aim 3: Representation of PWA

Across the 27 reviewed studies, nine had no mention of including or excluding PWA. In the remaining 18 studies, 15 reported inclusionary and/or exclusionary criteria that directly [e.g., exclusionary criterion of "severe aphasia" (53), p. 324] or indirectly [e.g., exclusionary criterion of "…inability to follow 1-2 step commands" (63), p. 924] pertained to aphasia, only two of which (59, 61) provided information regarding how many of their participants did or did not have aphasia. The other three studies referring to aphasia (48, 56, 66) indicated consideration of PWA in the absence of any aphasia-related inclusionary/exclusionary criteria. For example, Khattab et al. [(48), p. 3] reported excluding the data of four participants (two with "significant aphasia," two with "difficulty understanding test instructions") who completed the aerobic exercise intervention but had incomplete cognitive outcome data. Khattab et al. (48) did not, however, specify if any remaining participants had less "significant" aphasia symptoms. Similarly, Tang et al. [(66), p. 843] stated that cognitive test data were missing for four participants due to "significant aphasia" and one participant due to "difficulty understanding test instructions;" they also stated that "for those with missing pre-training data points, these participants were excluded from the analysis." Because (a) Tang et al. (66) did not explicitly state whether the participants with significant aphasia were in their aerobic exercise vs. control balance training group, (b) it is not clear in their cognitive outcome table which participants are linked to the missing data (e.g., participants with significant aphasia vs. comprehension issue), and (c) these researchers did not specify if any of their other participants had less "significant" aphasia, it is difficult to determine if there was any aphasia representation in their study. Furthermore, those studies directly or indirectly excluding those with severe PWA often failed to report whether any participants did present with mild-to-moderate aphasia or another cognitive-communication disorder. Across the 18 studies that made some mention of aphasia or acquired language difficulties, there was nominal information (e.g., no mention of what aphasia test was used or what healthcare professional made the aphasia diagnosis) about how the presence and severity of aphasia was determined.

In total, only three reviewed studies (56, 59, 61) detailed how many participants had or did not have aphasia. There was a total of 12 PWA across these studies who completed aerobic exercise intervention and whose cognitive test data were used in analyses. Meester et al. (56) specified including six participants with mild aphasia but did not specify how mild aphasia was identified. Ploughman et al. [(61), p. 204] also included six participants with expressive aphasia ("mild-severe") in their two participant groups (three in each), stating that the National Institutes of Health Stroke Scale (87) was used to identify expressive aphasia but failing to report aphasia severity from the Best Language item of this rating scale. McDonnell et al. (55) stated that individuals with severe expressive or receptive aphasia were excluded from their study on the basis of interviews, and that none of their participants had expressive aphasia, but neglected to specify who completed those interviews, how the absence of expressive aphasia was confirmed, nor whether any participants had mild or moderate receptive aphasia.

Accordingly, it is challenging to ascertain representation of PWA within the 27 included studies. A third of the studies offered no information about the aphasia status of participants, and the vast majority of studies that reported inclusionary/exclusionary criteria related to aphasia failed to specify if such criteria resulted in inclusion of PWA. Only three studies (56, 59, 61) provided sufficient information to determine how many of their participants had aphasia; however, for the only 12 PWA in these studies whose data were utilized, aphasia characteristics were not properly reported and the described aphasia assessment procedures were inadequate. Without knowing these participants' aphasia profiles, it cannot be determined whether their language symptoms confounded performance on cognitive measures. Additionally, none of these studies reported language outcomes. Such deficient reporting of aphasia has been identified in previous systematic reviews of the stroke literature [e.g., (88)] and more broadly in stroke rehabilitation studies (89), and is problematic given the prevalence of stroke-related aphasia (38, 39). Given our findings regarding aphasia representation within the included studies, it is not yet possible to determine if exercise can positively affect the cognitive symptoms of individuals with stroke-related aphasia.

There are methods to foster the participation of PWA in future exercise intervention research. Modifications to physical home exercise programs to reduce cognitive and language load such as simplifying syntax and vocabulary in instructional text, adding supportive images (personalized photographs of the participant performing each step may be particularly helpful), and using supportive communication techniques to maximize auditory comprehension and verbal expression have been used successfully to ensure program adherence in the context of aphasia (90, 91). Indeed, Marsden et al. (92) indicated that they had not used aphasia as an exclusionary criterion to assure that a broad spectrum of stroke survivors could participate and that they used "aphasia-friendly" [(92), p. 341] written material and presentation when providing education about physical activity and other aspects (e.g., goal-setting, fatigue management) of their community-based group intervention for stroke survivors and their caregivers. Nonetheless, Marsden et al. did not specify how many of their participants had aphasia, did not describe the "aphasia-friendly" modifications within the education component of their intervention, nor indicate if during the physical activity training component of their intervention, staff members who were supervising the activity sessions utilized supportive communication techniques. In studies including PWA and/or individuals needing cognitive or language supports, modifications to protocol instructions and materials to reduce cognitive and language load should be clearly detailed (91).

Study Limitations

It is important to acknowledge limitations of the current review that may temper confidence in its findings. Only peer-reviewed articles published in English were considered. Because of the publication bias toward studies yielding positive findings, there may be studies yielding null findings in the gray literature that were missed. Because this review was intended to provide state-of-the-art information, only articles from 2008 and later were included; thus, the time-bound nature of this type of review may not capture the whole picture of the development of this subject area.

CONCLUSION AND FUTURE DIRECTIONS

Although results were mixed, this systematic review found some evidence for positive, direct effects of aerobic exercise on attention, memory, executive function, visuospatial skills, processing speed, and language after stroke (see **Figure 3**); however, methodological variability across studies limited our ability to identify which factors drove positive or null effects. Importantly, our review highlighted four primary areas to address in future research. First, individuals with aphasia were often excluded. As this represents approximately one-third of the stroke population (38), the inclusion of individuals with aphasia will help make the findings more generalizable. Second, a number of studies relied on cognitive screening measures (e.g., MMSE) that are not suitable for identifying nor measuring change in single domain strengths and weaknesses (78, 79). Thus, future work should include cognitive measures that are most appropriate for measuring change to improve sensitivity and reproducibility [see (55)]. Third, there was an absence of performance-based measures of language. Including performance-based language measures that can capture deficits often encountered in both right and left hemisphere strokes will help with a more complete understanding of the potential benefits of exercise in this population. Finally, much of the extant literature included cognitive measures that rely heavily on intact language for accurate measurement of the cognitive domain being tested (40, 93). For example, it is impossible to selectively measure executive function using a semantic fluency task with an individual who has anomia. Additionally, auditory comprehension difficulties may negatively affect performance on cognitive tests with complex instructions. Including non-linguistic cognitive measures that are accessible to stroke survivors who have difficulty with language will improve the interpretability of the results of exercise studies in this population.

Given the preliminary evidence that exercise improves cognition across multiple healthy and neurogenic populations [e.g., (13)], future studies are highly warranted for those with language and other cognitive sequalae following stroke. Future work should continue examining the direct impact of aerobic exercise on cognition post-stroke, as well as the combined effects of aerobic exercise and cognitive interventions for stroke survivors (12), particularly individuals with aphasia.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

All authors contributed equally to the manuscript, including review conception, database search, data extraction, and manuscript writing. All authors have read, revised, and approved the submitted version of the manuscript.

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SUPPLEMENTARY MATERIAL

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REVIEW
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A Scoping Review of Barriers and Facilitators Affecting the Lives of People With Disabilities During COVID-19

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Purpose: This scoping review aimed to identify the barriers and facilitators to everyday activities and social participation of people with a disability (PWD) during the first wave of the COVID-19 pandemic.

Methods: The search terms (disability and COVID-19) were used in four databases: CINAHL, Medline (Ovid), EMBASE and Web of Science. The search conducted from January 2019 to September 22, 2020, identified 465 peer reviewed articles and abstracts and were screened in Covidence software. Studies were included if they had the terms "COVID-19" and "disability," were published in English, and specifically examined how COVID-19 impacted the daily lives of PWD. Exclusion criteria included: disability as a symptom or result of COVID-19, the health outcomes when PWD acquired COVID-19, disability leave for someone who is sick and the risk of acquiring the disease for PWD. 74 articles met the inclusion criteria and were analyzed via data charting. Charting began with existing barriers and facilitators identified by the World Health Organization and new barriers and facilitators, that emerged from the texts were added during this process.

Results: The barriers that emerged included: access to information, ease of communication, financial impacts, mental health impacts, access to essential services, physical safety, educational challenges, and changes to care and rehabilitation. Significant facilitators included: changes to care and rehabilitation, new innovations, social and familial support and inclusive policy measures.

Conclusion: COVID-19 exacerbated existing challenges in the lives of PWD and raised new quality of life concerns. Findings also demonstrate that policy makers, health care professionals and others continually support PWD in times of crisis.

Keywords: disability, COVID-19, pandemic, inclusivity, barriers, facilitators, social participation, rehabilitation

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INTRODUCTION

In December 2019, doctors in Wuhan, China identified a cluster of pneumonia cases that were caused by the novel coronavirus SARS-Co-V2 (1). Despite efforts to contain the virus and its consequent disease COVID-19, the World Health Organization (WHO) declared that the outbreak had reached pandemic levels by March 2020 (2). Six months later, the WHO had reported nearly 33 million cases, alongside almost one million deaths worldwide, with both statistics still climbing rapidly. As with most health and humanitarian crises, certain vulnerable populations are more susceptible to adverse outcomes during this pandemic. The United Nations (UN) definition of disability inclusion is the meaningful participation of people with disabilities (PWD) in all their diversity, the promotion of their rights and the consideration of disability-related perspectives (3). The UN stated that PWD are particularly disadvantaged by the socio-economic and health consequences of the COVID-19 pandemic (4). This is not surprising, as PWD have regularly experienced a variety of access and inclusivity barriers (5). PWD across all socioeconomic circumstances struggle to receive enough financial compensation, equal job opportunity and inclusive care and rehabilitation (5). The UN has predicted that the COVID-19 pandemic has exacerbated aforementioned existing inequalities, in addition to proposing new challenges to PWD (4).

Nearly all countries have implemented stringent measures to mitigate the risk of COVID-19 spread, which commonly includes physical distancing policies, economic lockdowns and a rapid shift to virtual life, among other modifications (6). These changes have dramatic impacts on the lives of all individuals, but create unique challenges for PWD. For example, many PWD have health issues that require them to frequently attend in-person care appointments, many of which would have been cancelled or switched to virtual format at the beginning of the pandemic. These challenges will be explored at length in the results section. Even though the UN (4) and the WHO (7) have released documents outlining specific frameworks for governments to incorporate disability inclusion in their pandemic responses, a preliminary search through the literature conducted by Samantha Croft (SC, author) revealed that many inequities are not being addressed.

In contrast to the barriers and increasing exclusion of PWD in certain activity and participation situations (8), there is some evidence that individuals and corporations have found innovative ways address inclusivity barriers in the time of COVID-19. For example, a popular media piece at the beginning of the pandemic outlined how certain grocery stores were offering unique hours for "vulnerable" populations to shop, and soon after, many supermarkets worldwide implemented the same change (9). This suggests the pandemic may be highlighting the inequalities that PWD have been facing for decades, and may provide an opportunity for governments to address them and offer solutions (10). Overall, new findings are emerging that examine both the barriers and facilitators for the daily lives of PWD during this pandemic.

While it is important to address the health outcomes for COVID-19 cases, as well as the risk of acquiring the disease for PWD, the focus of this review is to examine how pandemic responses have affected the daily lives of PWD. While some studies have reported findings on how the daily lives of PWD have been affected by the COVID-19 pandemic (11, 12), to our knowledge, no review has been published that has compiled the aforementioned research to identify common experiences of activity restrictions and social participation limitations of PWD during the COVID-19 pandemic. This scoping review intends to fill this gap in the literature by answering the following research question: How have the pandemic responses affected the daily lives of PWD worldwide? The objective of this paper is to identify common inclusivity successes and failures of the COVID-19 pandemic response seen globally. Emphasizing these common themes has the potential to shape the future of inclusive policy-making, inform research on this topic, and maximize positive, inclusive approaches for PWD and their families and caregivers.

MATERIALS AND METHODS

Researchers Arksey and O'Malley define a scoping review as, a review technique that involves 'mapping' the relevant literature in a field of interest (13). A scoping review is particularly suited for the exploration of broader and newer research topics (13) and therefore is appropriate to examine the COVID-19 pandemic and its impacts on PWD as this is a novel research area. Scoping reviews do not assess the quality of the studies included but are often used to: decide whether to proceed to a systematic review, summarize the and disseminate the research findings in a specific area and identify any potential gaps in the literature (13). The authors were guided by the methodological framework set out by Arksey and O'Malley (13), and the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) (14).

SC developed the search strategy alongside an experienced librarian in the Health Sciences department at the University of Ottawa, and the final search terms were confirmed by SC and Sarah Fraser (SF, author). The key search terms were "COVID-19" and "disability," but they also encompassed the range of alternate vocabulary used to identify these topics to ensure all relevant research was included (see **Figure 1** for full details).

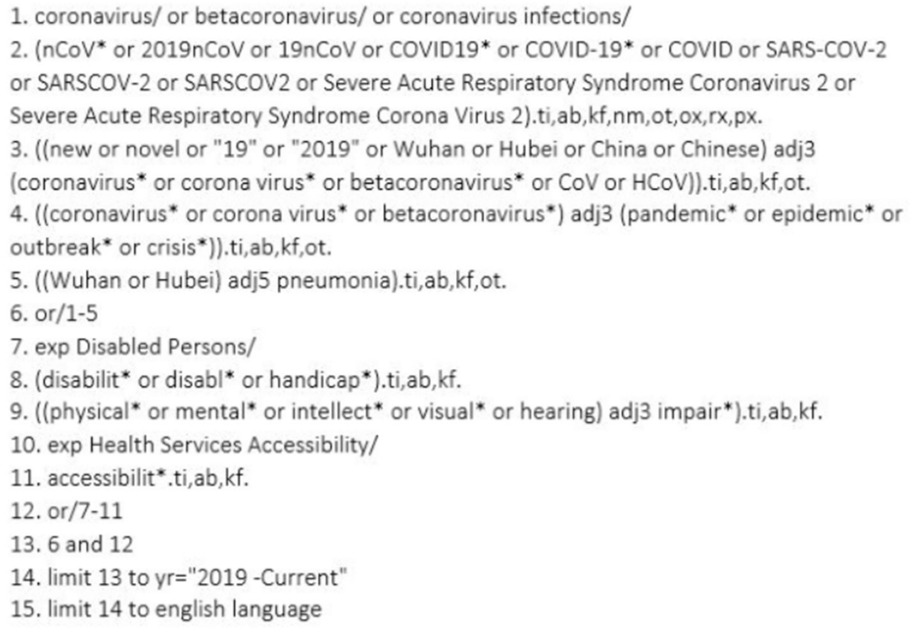
Inclusion Criteria

It is important to note that the authors of this paper, SC and SF, included intellectual/developmental, sensory, physical and psychological disabilities, as types of disability that are impacted by the current pandemic. Additionally, articles discussing PWD of any age, children, or adult, were included for analysis. Furthermore, for feasibility reasons, this study focused on contexts that would encompass the daily lives of PWD, such as community, home, rehabilitation, and educational settings. Articles published from January 2019- September 22 2020 were targeted, in order to ensure that only articles examining the novel SARS-Co-V2 virus were included. Furthermore, articles written in English were eligible for review as it is the only language fluently shared by both reviewers. Except for existing reviews on this topic, all types of published journal articles were eligible for review (i.e., opinion articles, commentaries, etc.).

Exclusion Criteria

Articles were excluded if they discussed disability as a symptom or result of COVID-19, the health outcomes when PWD acquired COVID-19, disability leave for someone who is sick or the risk of acquiring the disease for PWD. We also omitted other review articles in the screening process.

The search was conducted on September 22, 2020 and the final search terms were used in the following databases: CINAHL,



1. coronavirus/ or betacoronavirus/ or coronavirus infections/
2. (nCoV* or 2019nCoV or 19nCoV or COVID19* or COVID-19* or COVID or SARS-COV-2 or SARSCOV-2 or SARSCOV2 or Severe Acute Respiratory Syndrome Coronavirus 2 or Severe Acute Respiratory Syndrome Corona Virus 2).ti,ab,kf,nm,ot,ox,rx,px.
3. ((new or novel or "19" or "2019" or Wuhan or Hubei or China or Chinese) adj3 (coronavirus* or corona virus* or betacoronavirus* or CoV or HCoV)).ti,ab,kf,ot.
4. ((coronavirus* or corona virus* or betacoronavirus*) adj3 (pandemic* or epidemic* or outbreak* or crisis*)).ti,ab,kf,ot.
5. ((Wuhan or Hubei) adj5 pneumonia).ti,ab,kf,ot.
6. or/1-5
7. exp Disabled Persons/
8. (disabilit* or disabl* or handicap*).ti,ab,kf.
9. ((physical* or mental* or intellect* or visual* or hearing) adj3 impair*).ti,ab,kf.
10. exp Health Services Accessibility/
11. accessibilit*.ti,ab,kf.
12. or/7-11
13. 6 and 12
14. limit 13 to yr="2019 -Current"
15. limit 14 to english language

FIGURE 1 | Full search strategy used on database Medline (Ovid). #1-5 are referring to articles containing coronavirus and all varying modifications to the term. #7-11 denote articles containing disability, accessibility and all varying modifications to the terms. Finally, #12 and #13 are a combination of the above concepts, while #14 and 15 are limiting the search by year and English language.

Medline (Ovid), EMBASE and Web of Science. After duplicates were removed in Covidence review software (15), 465 articles remained, which were then screened by title and abstract. Articles were retained if they supported the research question and inclusion criteria. Both SC and SF screened the title and abstracts of the first 200 articles, and any necessary discussion and conflict resolution was conducted at regular bi-weekly intervals. As there was 81% agreement between the raters and a Kappa of 0.6 after half the abstracts were screened, the remaining abstracts were screened solely by SC. Of the 465 articles, 179 were retained for full text screening. Articles were dismissed if their titles and abstracts did not conform to this research question (i.e., discussing the impacts of pandemic responses on the daily lives of PWD). The full text screening was conducted by both authors, which also included regular discussion and conflict resolution via video call. After careful consideration of the exclusion and inclusion criteria, 74 articles remained for thematic analysis (see **Figure 2** for full details of the screening process). Specifically, 31 articles were excluded for discussing the wrong patient population (i.e., non-disabled individuals) and 26 articles were excluded for insufficient information regarding this study's research question. Further, the authors omitted 24 articles based on the exclusion criteria, (i.e., the risk of PWD acquiring COVID-19). Thirteen studies had wrong outcomes, for example measuring doctor errors when treating PWD. Seven articles were excluded for having the wrong study design (i.e., existing rapid scoping reviews). Finally, one study was set in hospitals, rather than in everyday settings for PWD, and the remaining three articles were excluded because they were either duplicates that were initially missed, or the full text could not be located.

The data-charting template was created by author SC and refined by SF. Barriers and facilitators were created a priori based on results from preliminary searches through the literature, specifically from the WHO report "Disability considerations during the COVID-19 outbreak" that was released early in the pandemic (7). However, the barriers and facilitators were also updated iteratively as new themes emerged from the included articles. The authors chose to extract general data such as main author name, article type and length, country of origin, population of study, and key topics covered. Additionally, SC extracted thematic and sub-thematic data, as well as significant notes and quotations from the articles. An example of the data chart used can be found in **Figure 3**. Please note only the data from one included article is shown as an example. Finally, SC conducted the summarization, collation and report of the results, using the information she extracted and charted, and this was deliberated and discussed with SF.

RESULTS

Article Characteristics

After the full text screening process, the authors were left with 74 articles for extraction and analysis. Of these, 55 articles (74.3%) were literature such as editorials, opinion and commentary articles. The remaining 19 articles (25.6%) were observational studies, including cross-sectional, case and framework analysis studies. The articles examined barriers and facilitators for PWD from a wide variety of countries, the most common being the USA, the United Kingdom and

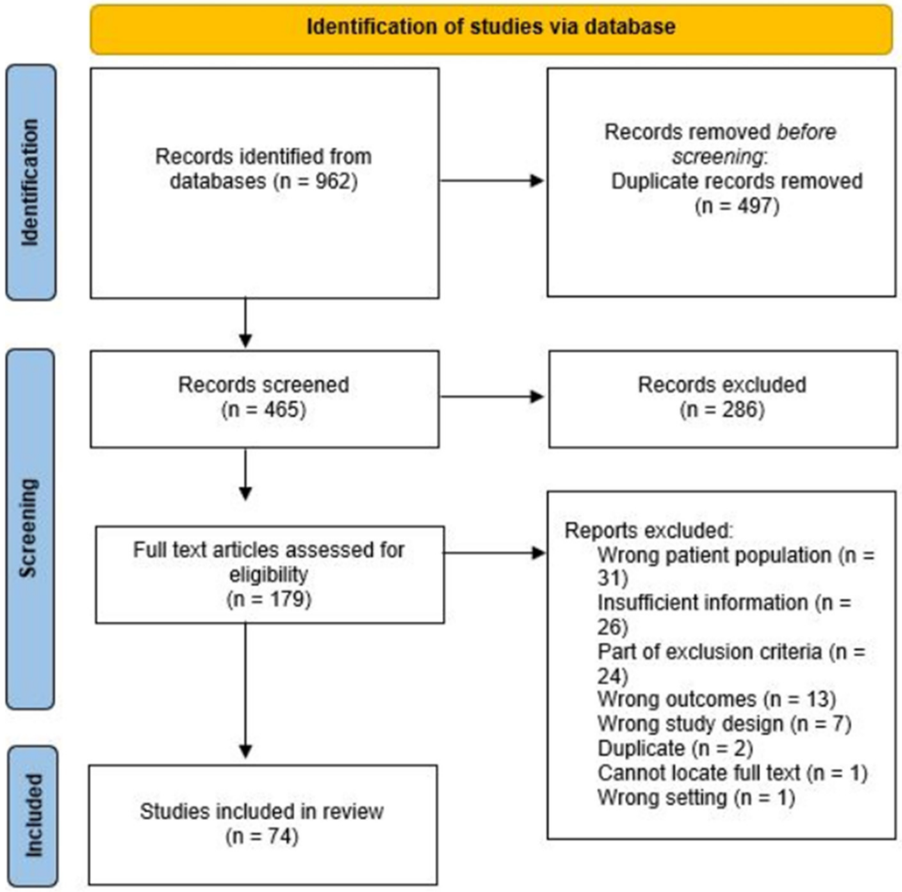


FIGURE 2 | Scoping review flowchart.

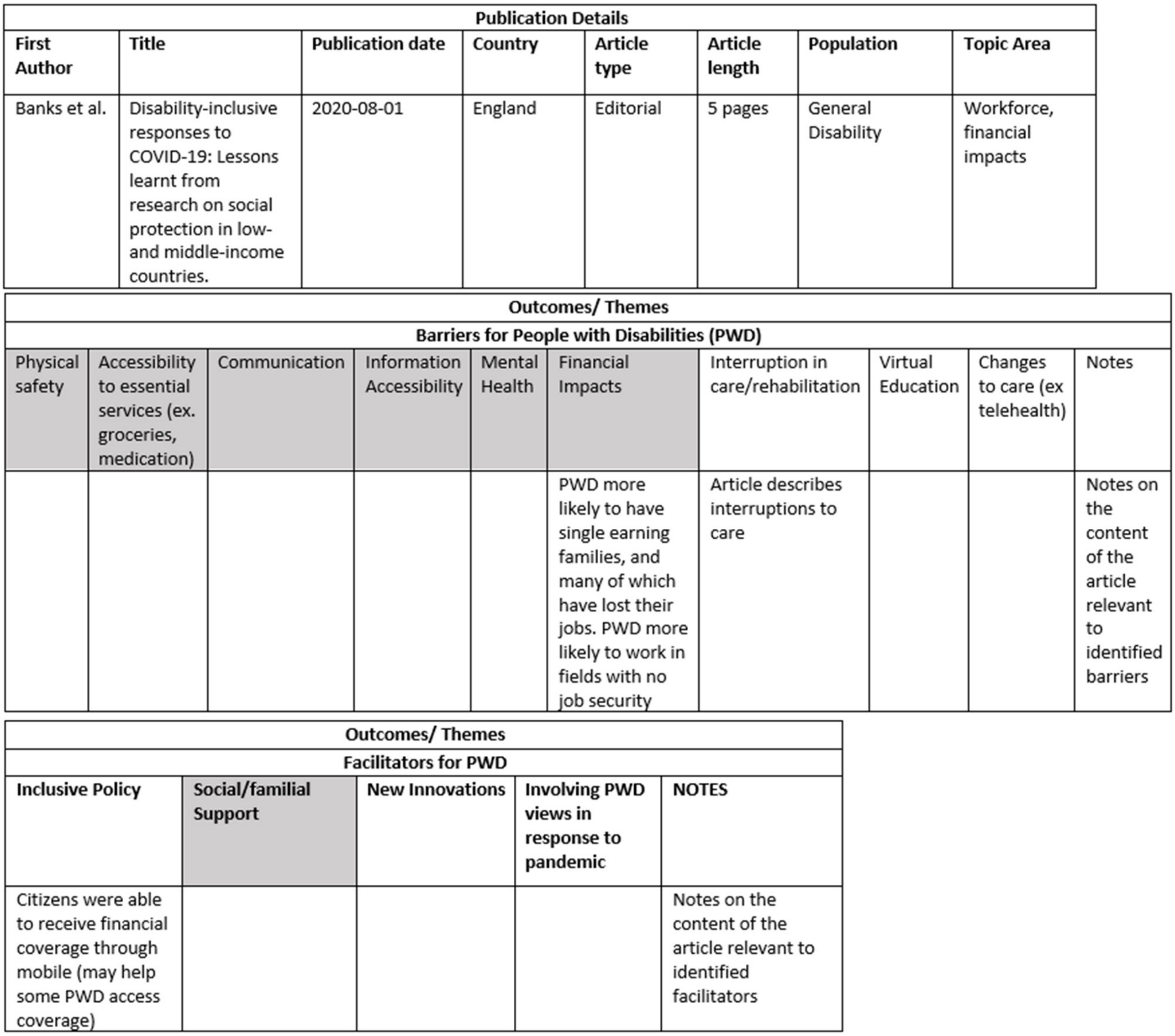
Brazil. However, many included commentaries and opinion articles did not specify a country of study, which is why there is no detailed data on country of origin for the included articles. When examining the type of disability, 34 articles (45.9%) did not specify a disability type, but rather discussed PWD in general. Furthermore, nine articles (12%) focused on individuals with sensory impairments, including populations who are deaf or hard of hearing (DHH), visually-impaired or blind. Additionally, 17 articles (22.9%) discussed how COVID-19 has impacted individuals with intellectual and developmental disabilities (IDD), such as autism spectrum disorder (ASD). Meanwhile, three articles (4%) detailed mental and psychological disabilities, such as obsessive-compulsive disorder (OCD). There were six articles (8%) included that discussed physical disabilities, including spinal cord injuries (SCIs), chronic pain conditions and osteoarthritis. Finally, there were five articles (6%) pertaining to a variety of classifications (i.e., diseases of the nervous system, congenital malformations of the nervous system, etc.), such as Parkinson's disease (PD), multiple sclerosis (MS), and spina bifida (17). This category will be referred to as "Other" (see **Figure 4** for a visual representation of this data).

Important Themes That Emerged From the Data

The findings were first categorized into two main themes: barriers and facilitators for the lives of PWD during the COVID-19 pandemic. These were then subcategorized into several subthemes that were commonly presented in the articles. The barriers noted included: (a) access to information, (b) ease of communication, (c) financial impacts, (d) mental health impacts, (e) access to essential services, (f) physical safety, (g) educational challenges, and (h) changes to care and rehabilitation. Significant facilitators included: (a) changes to care and rehabilitation, (b) new innovations, (c) social and familial support and (d) inclusive policy measures. It is noteworthy that one subtheme, changes to care and rehabilitation, is described as both a barrier and facilitator. This will be explored at length in the discussion (see **Figures 5**, **6** to visualize how many articles discussed each theme. Please note that many articles discussed multiple themes).

Barriers for PWD During the Pandemic

(a) Access to Information: In a crisis such as the COVID-19 pandemic, messages from the government and international organizations such as the WHO must reach the public quickly



Publication Details							
First Author	Title	Publication date	Country	Article type	Article length	Population	Topic Area
Banks et al.	Disability-inclusive responses to COVID-19: Lessons learnt from research on social protection in low- and middle-income countries.	2020-08-01	England	Editorial	5 pages	General Disability	Workforce, financial impacts

Outcomes/ Themes									
Barriers for People with Disabilities (PWD)									
Physical safety	Accessibility to essential services (ex. groceries, medication)	Communication	Information Accessibility	Mental Health	Financial Impacts	Interruption in care/rehabilitation	Virtual Education	Changes to care (ex telehealth)	Notes
					PWD more likely to have single earning families, and many of which have lost their jobs. PWD more likely to work in fields with no job security	Article describes interruptions to care			Notes on the content of the article relevant to identified barriers

Outcomes/ Themes				
Facilitators for PWD				
Inclusive Policy	Social/familial Support	New Innovations	Involving PWD views in response to pandemic	NOTES
Citizens were able to receive financial coverage through mobile (may help some PWD access coverage)				Notes on the content of the article relevant to identified facilitators

FIGURE 3 | Data-charting template used by the authors. Data charting included three sections: Publication details, Barriers for people with a disability (PWD) and Facilitators for PWD. Barriers and Facilitators in grey in the table represent existing barriers identified by the WHO and additional barriers/facilitators that emerged from the data in are white. This figure includes the data charting for one article by Aishworiya et al. (16), and notes are not included in this example of the data charting as they are lengthy.

to keep the citizens informed (18). This often leads to a lack of attention paid to those who may have alternate communication needs, despite the UN's Convention on the Rights of Persons with Disabilities specifying that access to information is vital during humanitarian crises (19). Therefore, it is not entirely surprising that the findings revealed that many PWD had difficulties accessing public health messaging regarding COVID-19 due to a lack of proper accommodation, such as subtitles and sign language interpreters (19–25).

The results indicate that information regarding COVID-19 has often been inaccessible to individuals with IDDs specifically (26, 27). For example, Fernandez-Diaz et al. (28) found that the WHO website rated poorly on an operability scale, meaning that it was difficult to navigate and find relevant information within the website for people with IDD.

Individuals with visual disabilities or blindness have also faced challenges retrieving accessible COVID-19 information (27–29). A study by Fernandez-Diaz et al. (28) revealed that while the WHO's website did have some alternative text available, many pages had errors, or did not have any alternative text available. Furthermore, the website rated poorly on color contrast, which can aide visually-impaired individuals with website navigation (28). Sabatello et al. (29) noted that COVID-19 information is frequently portrayed in infographics that are very difficult for visually-impaired individuals to read. Finally, Guidry-Grimes et al. (27) identified the need for audio descriptions to increase inclusion during press conferences.

This situation is similar for people who are DHH, who experience difficulties understanding information from government press conferences unless subtitles or sign language interpreters are utilized (29). Unfortunately, Yap et al. (19) discovered that only 65% of publicly-available pandemic-related briefings that occurred in low- and middle-income countries, and 0% of that of international organizations such as the WHO offered a sign language interpreter during the first months of the pandemic. Additional articles note similar findings in other nations, such as China (27, 30, 31).

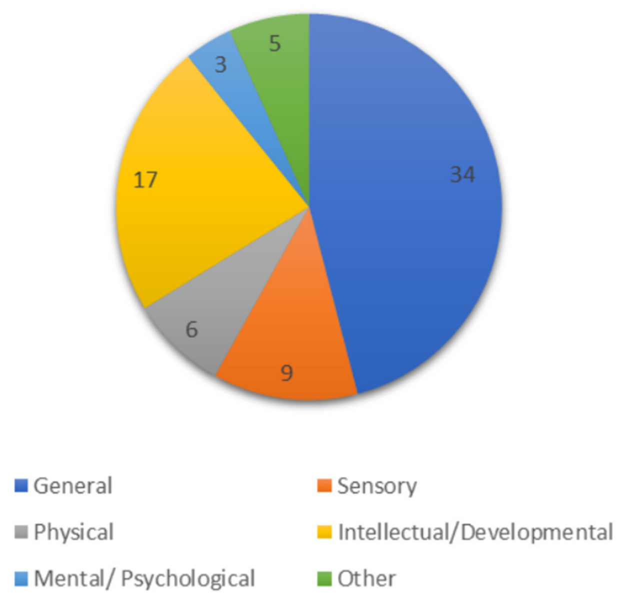


FIGURE 4 | Publications per disability type.

(b) Ease of Communication: New COVID-19 policies, such as virtual health care and mask mandates, have implemented barriers for certain PWD to communicate. Several included articles note that it is extremely challenging for many DHH individuals to communicate in public while wearing masks, as it directly impedes one's ability to lipread or hear what is being said (32–35). Medical student Isabelle Williams (36), who is DHH, states that her ability to communicate on the job is severely limited by masks, and she is troubled by the lack of guidance regarding how to wear both full personal protective equipment (PPE) and an auditory aide at work. Furthermore, for PWD who are attending school or care appointments virtually due to the current social distancing protocols, effective communication has been reported as nonexistent where no accommodation is provided (37–39).

(c) Financial Impacts: In general, PWD are more likely to have financial disadvantages, due to underlying factors including the increased cost of living for PWD, the inaccessibility of certain jobs, and expensive health concerns (6, 40). The financial disparity between disabled and able-bodied individuals is exacerbated by the COVID-19 pandemic (26). PWD are often a part of single-income families, have jobs with lower wage and/or security, and are often the first to lose their jobs during budget cuts such as those seen during COVID-19 (41–43) and face limited financial supports during the pandemic (44).

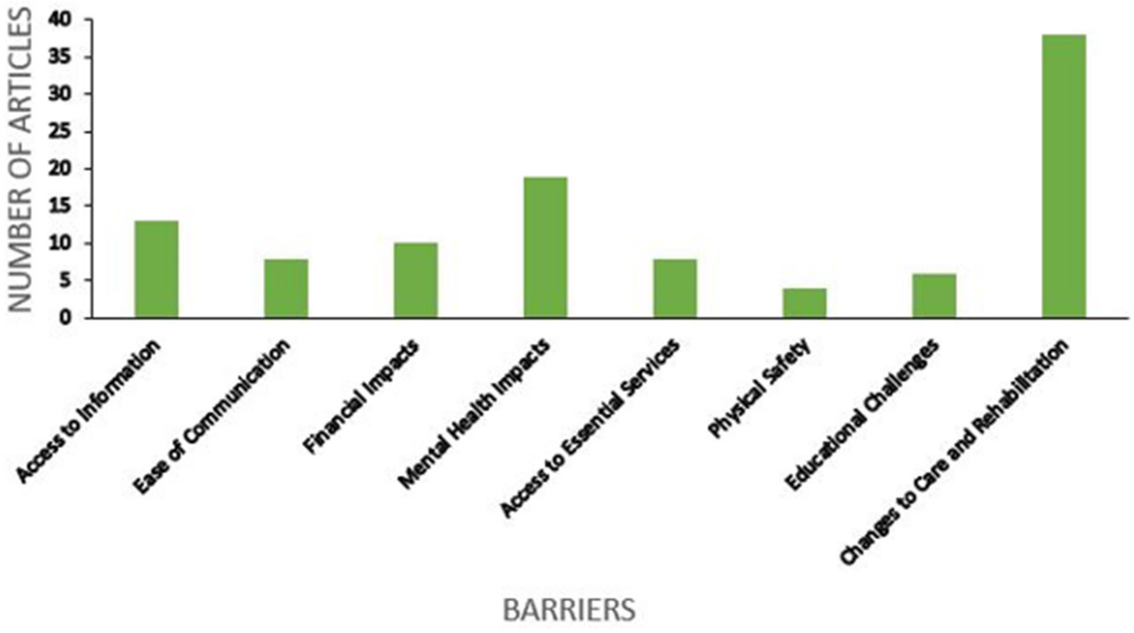


FIGURE 5 | Number of articles per barrier.

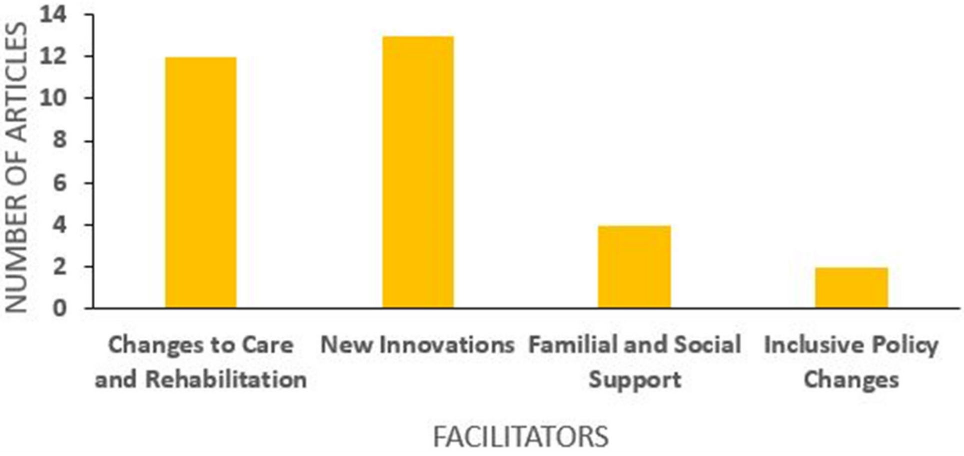


FIGURE 6 | Number of articles per facilitator.

While some countries have offered financial support to PWD, these packages were often insufficient (45–47) and/or not provided to those who already received a disability pension (47).

There are also other inequities regarding financial wellbeing during the pandemic. A commentary published in the USA states that among people with acquired brain injuries (ABIs), women and members of the lesbian, gay, bisexual, transsexual, queer plus (LGBTQ+) community are more likely to have negative financial impacts due to the pandemic (48).

(d) Mental Health Impacts: The COVID-19 restrictions have created negative mental health impacts on nearly all societal groups; however, the literature suggests that PWD may be facing mental health issues at a greater rate (16, 35, 44, 49–52). Boldrini et al. (44) note that Italian citizens with disabilities have experienced an amplification of isolation, loneliness and a perceived lack of support since the pandemic began. Children with IDD, such as ASD, are particularly vulnerable to adverse mental health outcomes of the lockdown (16). As many children with IDD rely on structured programs, routines and clear expectations set out for them for mental health benefits, the disruption of everyday life is difficult for them to accept and comprehend (16, 35, 49, 51, 52). This disruption of routine and consequential confusion has led to an increase in anxiety, distress and even a deterioration in development and intellectual progress for children with IDD (21, 52–54).

Where programs have continued, the increase of preventative measures, such as staff wearing full personal protective equipment (PPE) and the ban of visitors to care centers, have also created decreased mental health outcomes for individuals with IDD (50). It is also more difficult for people with IDD to utilize social media for social connection, which exacerbates their feelings of isolation (45). Furthermore, the United Kingdom initially implemented a COVID-19 policy that only allows individuals to go outside once per day (55). Children with IDD faced mental health impacts from this sudden containment and restriction, and the government has since allowed increased time outside if it benefits individuals' mental health (55). Overall, in the United Kingdom, mental health decline has resulted in increased requests for psychotropic medication by caregivers of people with IDD in attempt to handle their behavioral challenges that have been exacerbated by the COVID-19 lockdowns (21).

The physical distancing and program closures related to COVID-19 have also affected people with physical disabilities. A French study conducted by Cacioppo et al. (11) on the mental health of children with physical disabilities revealed that more than half of the participants had increases in behavioral issues and sleeping difficulties since the lockdowns were implemented. Furthermore, individuals with chronic conditions are more at risk of acquiring mental health conditions such as anxiety and depression, even during normal circumstances (56). Finally, a survey implemented by Azzam et al. (57) also revealed that individuals with irritable bowel disease experienced significantly decreased body image scores over the course of the pandemic.

Additionally, the COVID-19 lockdowns have intensified the symptoms of people who live with mental health disabilities, and in some cases, increased the risk of suicide and severe psychiatric morbidities (23). For individuals with obsessive compulsive disorder (OCD), the government recommendations to adapt to increased safety and hygienic precautions may trigger an exacerbation of one's obsessions and compulsions, specifically hand-washing and cleaning, and may contrast what their doctor is instructing them to do to treat their OCD (58). Similarly, many Japanese citizens with anorexia have experienced a worsening of their condition due to anxieties surrounding the pandemic (42).

A cross-sectional study by Umucu and Lee (59) examined coping strategies of PWD during the pandemic and the consequent impacts on mental health status. One of the most common coping strategies used by the participants during the pandemic was self-distraction, which is correlated with increased stress levels, along with denial, substance use, behavioral disengagement, venting, planning, religion, and self-blame.

(e) Access to Essential Services: During the COVID-19 crisis, many PWD worldwide have experienced complications when accessing essential needs such as food and medication (14,

23, 31, 60, 61). A British study conducted by the Chronic Illness Inclusion Project revealed that 86% of respondents with chronic illnesses reported that the pandemic has had a negative effect on their ability to access food and essential goods (13). Commentaries by researchers Jumreornvong et al. (61) and Kuper et al. (23) have noted similar observations in the USA and United Kingdom, respectively. A worldwide study by Cheong et al. (60) found that nearly half of all participants with PD experienced barriers to accessing their regular medications, usually due to transportation interruptions and financial issues. These occurrences were much more frequent in low- and middle-income nations (60). Similarly, Qi and Hu (31) reported that individuals with MS in China were undergoing difficulties when purchasing medication and essential supplies due to public transportation closures.

When examining access to food and medication, some PWD experienced challenges when utilizing virtual grocery ordering services (33). While certain vulnerable populations were offered financial support for essential supply delivery in the United Kingdom, some PWD were not eligible (33). In other locations within the United Kingdom, essential supply delivery was canceled altogether to reduce the risk of disease spread, which posed challenges for PWD who usually rely on the service (20).

(f) Physical Safety: The COVID-19 pandemic has threatened the physical safety of PWD in several ways (33, 40, 48, 61). Firstly, the stress surrounding stringent public health measures and financial pressures due to COVID-19 may trigger the commencement or increased intensity of domestic abuse toward a disabled family member (40). Furthermore, there may be an increase in domestic violence simply because a person with a disability is spending more time at home with their abuser (61). Lund (40) also notes that it is much more difficult for victims to report this violence since PWD no longer see trusted contacts they confide in, in-person organizations were closed or offering virtual care in which a perpetrator may be in earshot within the home. This produced an environment where it is nearly impossible for PWD to report abuse (40). Furthermore, among individuals with ABIs, women and members of the LGBTQ+ community are more likely to experience interpersonal and domestic violence (48).

Additionally, certain physical safety barriers arise when PWD cannot ask strangers for assistance due to physical distancing mandates (33) or are waiting for necessary equipment or assistive devices to be repaired (40). For example, debris and other hazards on the sidewalk become much more dangerous for visually-impaired individuals when they are unable to ask for a stranger's aid for fear of contracting COVID-19 (33).

(g) Educational Challenges: Many educational settings worldwide transitioned to a wholly or partially virtual method for a period of time during the COVID-19 pandemic, which generated issues for PWD (37, 39, 47, 52, 62, 63). Few schools accommodated students who are DHH, even though they face challenges with virtual communication in the absence of accommodations such as a sign language interpreter (37). Furthermore, virtual education platforms are often inaccessible for people with physical, sensory or intellectual disabilities who normally have structured and individualized educational plans involving educational assistants, interpreters and other services, have not had these accommodations applied during virtual education use (47, 52, 62). Online schooling removed many educational aspects that PWD benefit from. For example, without the recreational activities and exercise that children with ASD usually access at school, they can get irritated, confused, and can even demonstrate stunted development (52). Furthermore, one Brazilian university professor notes difficulties teaching students with IDD challenging anatomical details without the practical use of cadavers, 3D models, and other helpful applications (63).

(f) Changes to Care and Rehabilitation: Many of the included articles across several countries such as the United Kingdom, Australia, Italy, France and Turkey note that PWD have experienced interruptions in their regular rehabilitation (14, 16, 20, 22, 41, 45, 50, 64–67). This occurrence has been noted across many disability types, including psychological and intellectual and developmental (64, 67).

In particular, 65% of PWD reported disruption to care during the pandemic, and more than half reported worsening health outcomes (13). Among children with IDDs, 30% within the USA and 50% outside the USA lost *all* rehabilitative services (67). A French survey revealed that 77% of children with physical disabilities experienced cancelled or postponed medical consultations during the pandemic (12). A study by Negrini et al. (68) estimated that in Europe, more than one million people per day were being denied their rehabilitation services in acute, post-acute and outpatient settings.

Closures of pain management clinics have had adverse impacts on individuals who are disabled by chronic pain and rely on the clinics for appropriate treatment (69). This has led to the increase of harmful or inappropriate pain interventions such as the abuse of nonsteroidal anti-inflammatory drugs, opioids and illegal substances (69). Secondly, for children who are having the first signs and symptoms of a disability during the pandemic, proper assessment, diagnosis and initial care are being delayed due to perceived non-urgence of the situation (22, 70). Tied closely to the mental health subtheme, individuals with IDDs can suffer from extreme mental distress as a result of disruption in regular care routines (51). These individuals, as well as those who are DHH or visually-impaired, may experience an intensification of their disabilities when being denied from the care they rely on for progress (39, 51, 71). Moreover, despite the importance of dental care, there is evidence that vulnerable populations have been neglected during the pandemic (72, 73). Dental workers acknowledge the increased anxieties surrounding the pandemic and recognize that going to the dentist is another cause of anxiety (72).

Individuals with specialized rehabilitation needs also suffered from the closures, as it was hard to access proper professional expertise (35, 44, 74). For example, Italian citizens with SCIs or a rare condition called Charcot Marie Tooth Disease experienced difficulties acquiring a care plan, as the few experts in the field were not accepting patients (44). In Austria, many individuals needing osteoarthritic knee and hip replacements were forced to postpone their surgeries, which ultimately decreased joint agility, physical function and activity levels in the individuals (74). Finally, the lack of blood donation drives due to pandemic closures has had major implications for populations who rely on regular blood transfusions, such as individuals with thalassemia major (35).

Additionally, rehabilitation services may be difficult to access even if the services are open. For example, care may be abruptly halted if support workers get sick or must tend to a sick family member (75). When these situations have occurred in China, agencies and governments have had difficulties finding last-minute replacements, which left PWD without essential care (31). Unfortunately, some countries have witnessed cases where no back-up care was provided, and PWD were left alone without care for several days, with one person in China even dying due to neglect (30, 31, 76). Furthermore, in a country such as Iran, where nearly all rehabilitation services are offered in urban settings and public transportation was temporarily shut down due to COVID-19, PWD living in rural settings had no way of accessing necessary services for their care (46).

A large portion of health care and rehabilitation has transitioned to telehealth, using video and audio calls to conduct treatment. The included articles reported that a significant disadvantage of telehealth is the need for high-speed internet (38). Families without technological applications such as computers and a strong internet signal risk being excluded by new telehealth services (16). Telehealth also poses significant barriers for individuals with sensory disabilities if a sign language interpreter is not available (32, 34). This type of platform may not be useful or appeal to certain PWD (i.e., cognitive impairment, impulse control, psychosis) (60). It is also challenging for health care professionals to utilize telehealth to prescribe painkillers to individuals living with chronic pain, as this often requires in-person testing (54, 69). Furthermore, audio tests conducted to monitor and diagnose individuals with DHH require perfect audio quality, which is rare via telehealth, making the service inappropriate for use of hearing impairment diagnoses and testing (70). Kolakowsky- Hayner and Goldin (48) note that women and members of the LGBTQ+ community with ABIs are more likely to lack the technological access required to attend a telehealth appointment.

It is true that family members of PWD can ease many barriers of telehealth (77) such as setting up the person in front of the computer, and conducting small assessments for the physician that require touch. However, this also has its drawbacks. Firstly, many individuals do not have a reliable and engaged familial support system, and it is unfair for this to be a requirement to access rehabilitation (78). Kolakowsky- Hayner and Goldin (48) also state that women and members of the LGBTQ+ community with ABIs are less likely to have the familial support often required to attend a telehealth appointment. Longo et al. (78) state that even if parents are willing to take over therapy and rehabilitation at home, this can put an additional stress on the family. Family members may be asked to help conduct tests or make specific observations, which may be inappropriate and risky, as the caregiver may not be trained in medicine or not understand the scientific implications (45). For PWD who need technological help from others in order to use telehealth, the possibility of privacy and confidentiality during care is eliminated (29, 40).

Facilitators for PWD During the Pandemic

(a) Changes to Care and Rehabilitation: Despite the several aforementioned disadvantages of telehealth, it is important to note the benefits it provides, as well (77). The pandemic situation makes safe face-to-face care relatively impossible, and thus, it is important to note that a key benefit of telehealth is that it provides an opportunity for safe continuity of care (61). Another commonly-cited advantage of telehealth usage for PWD is the increased ease of access (38, 64). Whereas, traveling to and from a clinic can often be time-consuming and inconvenient, specifically for PWD, telehealth can be accessed from the comfort and convenience of home (46, 60). Telehealth also gives the opportunity for health care professionals to observe the client in their own home, which may lead to greater understanding of an issue and easier ability to propose a solution (45).

Telehealth is reported to be an appropriate method of care for several disability populations. For example, Dorsey et al. (79) state that PD possesses very characteristic symptoms that can usually be diagnosed visually, which makes telehealth perfectly suitable in this case. Furthermore, there are a variety of smartphone apps (i.e., Doctot & CloudUPDRS) that can make telehealth more efficient for individuals with PD (80). Miele et al. (80) also outlined several other useful virtual strategies for patients with PD, such as the NMS Scale to examine non-motor symptoms, and Lift Pulse (Lynx Design, National Institutes of Health, Bethesda, Maryland) to monitor resting tremor.

There are similar virtual tools available for patients with MS. Researchers Moccia et al. (81) specifically recommend using the Patient Determined Disease Steps program and the MS Impact Scale before a telehealth consultation to assess the patient's state. Furthermore, doctors and patients have had success using neurological tests and the oral version of the Symbol Digit Modalities Test during telehealth to analyze disease progression (81). Health care professionals have also noted effective use of telehealth for evaluation of muscle strength, balance, fatigue and stamina for individuals with neurodevelopmental disabilities (82). Telehealth has also been shown to be beneficial in treating and assessing pediatric feeding disorders, behavioral issues and language delay during ASD (83).

(b) New Innovations: The COVID-19 pandemic posed unprecedented issues to PWD that required novel solutions. As Bruder (84) states, the pandemic gave individuals "a chance to demonstrate the resilience of the human spirit when faced with adversity and uncertainty." Around the globe, a variety of virtual leisure programs have been offered throughout the pandemic in order to help PWD exercise, socialize and keep busy while at home. Exercise programs were developed that aimed to mitigate the adverse outcomes of increased isolation and sedentary routines (85). These virtual exercise programs were offered by personal trainers, yoga instructors, and a variety of other specialists and may even offer a more accessible, inclusive and convenient way for PWD to exercise compared to traditional in-person gyms and classes (86). A notable case study by Lai et al. (87) applied a virtual reality exercise technology to

children with spina bifida for use in their homes. The researchers reported that the participants experienced increased motivation, calorie expenditure and sense of accomplishment while using the technology (87).

There were also technologies that emerged during the pandemic that facilitated communication for individuals with sensory impairments. A simple yet efficient solution was the creation of clear masks so that individuals who are DHH could lipread (32, 34). Other innovations for DHH individuals include communication boards, automatic speech recognition platforms and virtual interpreters (34). Furthermore, Martinez et al. (88) developed a device for blind individuals that will give them a sound warning when they are within six feet from another person, in hopes of guiding them through physical distancing restrictions. Blindfolded and blind individuals using the prototype both reported successful results (88).

Included articles also reported a variety of other creative methods to increase inclusivity toward PWD, while remaining physically distanced and safe. Firstly, the National Autistic Society offered virtual movie and craft nights to mitigate the adverse effects of disrupted routines on individuals with ASD (55). Other inclusive innovations include special grocery store hours for vulnerable populations, and Zoom calls hosted by Delaware's Developmental Disabilities Services to provide relevant information pertaining to the COVID-19 pandemic and disability to families (26, 76). Innovations were also implemented in educational institutions. For example, certain schools offered specialist teachers and one-on-one virtual calls with the professor to solve any pertinent issues (39, 62). Finally, the Dutch program DigiContact was in place pre-pandemic to allow individuals with IDD who live independently to access virtual support 24/7 (89). As the use of this program was significantly increased at several points throughout the pandemic, it is clear that DigiContact facilitated access to any necessary online support for these individuals (89).

(c) Familial and Social Support: Since care, education, and work are occurring mostly from home, many PWD are able to spend more time with their families, which has increased quality of life for PWD in many circumstances (51, 53). One study by Neece et al. (51) revealed that many families with disability appreciated a slower pace of life, and enjoyed the increased opportunity for sleeping, relaxing and meditating. Finally, the use of telehealth sometimes offers the opportunity for parents to be more involved in the care of their child with a disability. In this way, some health care professionals are noticing an increase in enthusiasm and engagement in rehabilitation through this method compared to normal face-to-face care, during which the parents are usually less involved (90). PWD have also pursued extra social support, increasing their use of social media to connect with others and overcome negative aspects of isolation (64).

(d) Inclusive Policy Measures: A study by Sakellariou et al. (47) examined how disability-inclusive new COVID-19 policies have been in four South American countries: Peru, Brazil, Argentina, and Chile. Peru was the only country studied that passed COVID-19-specific legislation protecting PWD's rights to equal education, employment and health care during the pandemic, explicitly citing the United Nations Convention on the Rights of Persons with Disabilities (47). The Peruvian government implemented measures to guarantee the wellbeing of PWD, including monitoring their access to care and ensuring their protection from any potential violence. Peru, as well as Argentina, and Brazil began to offer remote registrations for financial support for PWD. These three nations, as well as Chile also increased the financial support that certain PWD could receive during the first months of the pandemic (47). Additionally, Argentinian citizens with a disability were automatically re-registered for financial support during the COVID-19 pandemic. Furthermore, the Chilean government ensured that PWD were able to receive care by granting them special privileges, including a permit to visit care providers, and the ability to be accompanied by a caregiver during appointments. Similarly, Argentina, Chile and Peru all introduced policies that allowed PWD and their caregivers to go for walks without special permission and/or ensured caregivers could attend work, even if the area was under strict quarantine measures (47). Finally, a study by Banks et al. (41) reported that Georgia, Mexico, Mongolia, Lesotho, Tunisia and São Tomé and Principe plan to implement new or expand existing financial assistance programs for PWD in their COVID-19 response. Further, Gambia, Morocco and Togo now offer cash transfer distribution through mobile applications, rather than in-person, which would increase accessibility for some PWD (41).

DISCUSSION

This scoping review is unique because it focused on how the daily lives of PWD have been affected by COVID-19 protection measures, rather than examining COVID-19 treatment and risk of PWD acquiring the disease (91, 92). Several important barriers and facilitators present in COVID-19-infected countries were revealed, which highlight certain areas of concern for the social participation of PWD and potential avenues for future research.

Many of the themes that emerged in the findings were common issues that PWD faced prior to the pandemic that were exacerbated by pandemic conditions (5). For example, PWD still experienced inaccessibility to adequate health care services and negative financial impacts during the first wave of the pandemic, and there was evidence that the occurrence of these situations was increased due to COVID-19 (12, 16, 20, 22, 26, 41–47, 50, 64–67). In addition, new issues arose for PWD during the pandemic, such as: access to accurate COVID-19 health information, virtual education, and health care challenges. However, certain topics that were anticipated to be abundant in the literature were not, including employment changes for PWD ($n = 3$) (41–43), as well as instances where PWD were included in pandemic decision-making ($n = 0$).

In the included articles, it is important to address that telehealth exists as both a barrier and a facilitator to PWD during this pandemic. This technology can add both significant advantages, such as eliminating the need for travel (38, 46, 60, 64), as well as important disadvantages, such as difficulty of use for some PWD (32, 34, 54, 58, 69, 70). Researchers note that telehealth is effective for treatment and diagnosis of PD, MS and pediatric feeding disorders (79–81, 83). However, many articles cited unavoidable difficulties when utilizing telehealth for diagnosis and treatment of chronic pain, hearing impairments and individuals with symptoms of psychosis (54, 58, 69, 70). Furthermore, telehealth may be difficult to use if the patient does not have high-speed internet or a helpful family member to assist with calls. Researchers Chang and Lipner (32), as well as McKee et al. (34) note the importance of sign-language interpreters wherever needed for PWD during virtual appointments. It is imperative for health care professionals to discuss and plan with each patient that has a disability about the aforementioned potential telehealth issues, to ensure appropriate accommodations. Researchers and health care professionals should examine the telehealth application reviews by Miele et al. (80) and Moccia et al. (81) for useful telehealth strategies and tools to implement this technology with different PWD.

In the New Innovation subtheme, it was promising to see that several individuals, businesses and governments identify inclusivity issues and provide inventive solutions (32, 34, 55, 85–87). Certain innovations, such as virtual exercise programs, may be even more convenient, affordable and successful for PWD than traditional in-person methods (86). The examples presented in this subtheme may provide inspiration for others to implement the same or similar innovations in their region and potentially reach out to lesser served rural areas. Several of the included articles present clear issues that still require novel solutions (28). For example, the global use of innovative tools such as alternative text, descriptive video and proper color contrasting could increase accessibility of COVID-19-related information for individuals with visual impairments or blindness (28). This innovation would be helpful not only during a pandemic but would likely facilitate accessibility for this population for any future public health communications.

When discussing PWD, who are often marginalized, it is important to address the potential for intersectionality (93). The theory of intersectionality proposes that social inequality, discrimination and social hierarchy can be influenced by several aspects of one's identity, such as race, gender and disability, thereby creating a situation in which marginalized populations experience increased inequalities when several aspects of their identities intersect (93). For example, researchers Kolakowsky-Hayner and Goldin (48) describe how disability and gender can interact to create adverse outcomes. Specifically, the authors explain how among individuals with acquired brain injuries (ABI), women are more likely to experience disadvantageous financial situations, and increased difficulties accessing essential services and regular care (48). Here, it is clear that being a person with a disability and being a woman, both of which are aspects of identity that commonly are associated with inequality intersect to create an exponentially detrimental situation for the individual (94). Similarly, Kolakowsky-Hayner and Goldin (48) also identify an increased risk for women and members of the LGBTQ+ community to experience intimate partner violence during the pandemic. Lund (40) also details how stay-at-home orders can make it very difficult to report this violence, even when using telehealth calls, as many PWD may require a family member to assist with the appointment. During virtual appointments, health care professionals should be aware of these risks and acknowledge the possibility that the patient's family member may be their perpetrator. Wherever possible, health care professionals should ask caregivers to leave the room to provide privacy and a consequent opportunity for abuse reporting.

Implications for Health Care Professionals and Government Officials

This review highlighted several implications for health care professionals. Firstly, accessible information is imperative for PWD to be informed regarding a crisis, and to be able to make the appropriate risk management decisions (28, 71). Governments have a responsibility to ensure accurate and accessible information is being presented at press conferences and on government websites. Researchers identify the need for organizations to portray important information in a simpler and easier-to-understand manner (26, 27). Specifically, Goggin and Ellis (26) emphasize the importance of using "easy English," which they describe as, "using common words, simple sentence structure and using meaningful images to support information." Guidry-Grimes et al. (27) make similar recommendations using what they term "simple English." This method can make it easier for individuals with IDDs to understand complex health information such as COVID-19 details. Furthermore, in many public and health care settings around the world, there are little to no accommodations provided for people whose communication abilities have been hindered. This, as noted by McKee et al. (34), is in direct violation of the Americans with Disabilities Act. Clear masks, speech recognition applications and educational assistants must be provided to individuals who are DHH in order to improve communication.

Limitations

This study was limited by the type of articles included. Many eligible articles did not contain evidence-based information but rather detailed experiences and opinions from the research experts in the form of editorials, opinion and commentary articles. While this literature did expose the authors to new and important information, evidence included from commentary and opinion articles are not as accurate and robust as scientific studies. Furthermore, our study was limited to articles written in English and many originated from Western, developed countries. The information from developing countries reveals that PWD in those locations are likely more at risk of experiencing negative impacts from the pandemic. Thus, it will be imperative for researchers to carefully examine developing countries' experience with COVID-19 and disability and how this may be different from developed countries. Finally, because the articles included were all published before September 22, 2020, the results of this study may be limited to the first wave of the pandemic and do not reflect additional barriers and facilitators that might emerge over the long-term (95).

Implications for Future Research

The number of themes ($n = 8$) that emerged in this review highlight the importance of this topic, the barriers PWD face and some areas that may need more research. For example, telehealth, access to information and mental health were commonly mentioned in the recent literature, while topics such as domestic violence and financial impacts on PWD seemed under-researched. These neglected topics should be emphasized during future research in this area, in order to better comprehend the overall impact of the pandemic response on PWD. Additionally, as more data regarding inclusive policy successes is conducted, a review on this area could lead to a wider uptake of inclusive policies globally. As previously mentioned, there is a gap to be filled by answering our research question in other languages and in a wider range of countries to better understand the impacts of COVID-19 for PWD on a global scale. Finally, the authors originally expected to identify instances of inclusion of PWD in the pandemic response decision-making process, but unfortunately, no articles mentioned such occurrences. Upcoming research should investigate whether this occurred in any circumstances and if so, compare instances in which PWD were consulted to instances in which PWD were excluded from decision-making.

CONCLUSION

This scoping review examined 74 articles and the analysis exposed several significant barriers and facilitators for the daily lives of PWD. Significant barriers in the daily lives of PWD during the pandemic included: access to information, ease of communication, financial impacts, mental health impacts, access to essential services, physical safety, educational challenges, and changes to care and rehabilitation. The noted facilitators for daily life of PWD included: changes to care and rehabilitation, new innovations, social and familial support and inclusive policy measures.

The results of this study reveal that pre-pandemic issues that were already barriers for PWD in their daily lives, such as access to inclusive care, financial barriers and communication issues, were exacerbated by the current pandemic. Furthermore, new challenges, such as access to COVID-19-related information and challenges regarding mandatory telehealth, were presented to PWD during the pandemic. Therefore, this review provides insight into the variety of inequalities still pervasive worldwide. Alternately, there were several examples of technological and policy innovation that attempted to solve inclusivity issues for PWD. These results have the potential to inspire further changes and solutions to issues that PWD are facing, in the context of COVID-19 and beyond. Furthermore, the information presented can inform policy decisions, and provide future researchers with the groundwork for more detailed investigations into the topics presented.

AUTHOR CONTRIBUTIONS

SC and SF database search, screening and selection of articles, analysis and interpretation, and final write-up. Both authors contributed to the article and approved the submitted version.

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Criterion Validity and Test-Retest Reliability of a Modified Version of the International Physical Activity Questionnaire–Short Form (IPAQ-SF) in Kidney Transplant Recipients

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Introduction: Accelerometry, the clinically valued standard of physical activity monitoring, has limited acceptance in transplantation rehabilitation; therefore, the International Physical Activity Questionnaire (IPAQ) self-report instrument is widely used. However, while the IPAQ's repeatability is good, its criterion validity is unsatisfactory. We hypothesized that adding a concise oral introduction would help overcome this shortfall.

Materials and Methods: This is a secondary analysis of a RCT in a sample of kidney transplant recipients that underwent observational follow-up. We assessed criterion validity of our modified version of the four-item IPAQ–Short Form (mIPAQ–SF) *via* Pearson, and test-retest reliability *via* intraclass correlation coefficients. The main difference in the new version is an oral pre-measurement introduction to the questionnaire's concepts. We compared our results with those of published studies.

Results: Post-kidney-transplantation data of 92 patients were analyzed. Across the four IPAQ-SF/mIPAQ–SF items, values of correlations between mIPAQ-SF responses and accelerometry records ranged from 0.07 (min in vigorous activity) to 0.35 (min in moderate activity) for criterion validity, and from 0.19 (days with moderate activity) to 0.58 (min in moderate activity) for test-retest reliability.

Discussion: Regarding moderate-to-vigorous physical activity, mIPAQ-SF self-reports' correlations to accelerometry records improved considerably on those of the IPAQ-SF ($r = 0.18$ vs. $r = 0.33$), i.e., improved criterion validity. We therefore conclude that a pre-measurement oral explanation of key IPAQ-SF/mIPAQ concepts enhances criterion validity regarding self-reported moderate-to-vigorous physical activity.

Keywords: psychometric properties, clinimetrics, kidney transplantation, physical activity, accelerometry

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INTRODUCTION

Physical activity (PA) monitoring has gained increasing consideration in recent years. Related research has achieved substantial findings, e.g., specifying the impact of regular PA vs. sedentary behavior in healthy and clinical populations (1, 2). Building on the available evidence, the World Health Organization (WHO) published an action plan with the goal of reducing the global prevalence of physical inactivity in adults first by 10%, then by 15%, respectively by 2025 and 2030 (3). Recognizing the benefits of sufficient PA regarding obesity, diabetes, hypertension and cardiovascular disease (3), the WHO now recommends age group-specific periods to spend in moderate-to-vigorous PA (MVPA) each week (4). These MVPA recommendations have been transferred to kidney transplant recipients (KTRs) with high cardiovascular risk (5, 6).

However, this group's adherence to PA recommendations tends to be poor (7). Both before and after transplantation, compared with healthy subjects, KTRs show low mean levels of PA (8). With age, their PA decreases to levels below those of other chronic disease groups (8). Therefore, ongoing support to help patients implement sufficient activity into their daily lives is now standard in post-transplantation clinical management.

Additionally, structured post-transplantation rehabilitation programmes have been developed to prevent, arrest or reverse pre-existing PA impairment. In many cases, such programmes can effectively halt the development of frailty and peripheral muscle dysfunction, both of which deteriorate health-related quality-of-life and increase mortality (6, 9).

Naturally, tailoring PA counseling to transplant populations requires adequate standardized monitoring. The gold standard of PA monitoring is accelerometry (7). However, for reasons thought to include poor acceptability to patients, device cost, difficulties with data extraction, the need for specially trained staff and the required seven-day period to collect reliable data (10), accelerometry has not yet become popular in clinical practice.

As an alternative, self-reporting questionnaires asking patients to rate their PA over a specified recall period may be used. Such questionnaires may be completed *via* a short interview format and are easily applicable across a broad range of clinical settings.

One widely-used example is the International Physical Activity Questionnaire (IPAQ). Also available as an easily applicable short-form questionnaire (IPAQ-SF), the IPAQ is designed to capture self-reported PA and sedentary behavior over either the last seven days or a hypothetical normal week (11). Translations into several languages allow broad international applicability (11). Furthermore, it can provide separate, detailed assessments of PA and sedentary time (i.e., covering 10-min segments and various intensities of PA) (11). This last feature makes the IPAQ appealing for transplantation rehabilitation settings, as it allows healthcare practitioners to target their feedback to specific intensity categories.

Problematically, despite the IPAQ's broad acceptance, its psychometric properties are limited. Specifically, while its test-retest reliability (repeatability) is good (11, 12), its criterion validity (correlation with known accurate measures, e.g., accelerometry) is poor (13). I.e., while repetitions of the test yield similar results, those results are not valid. This is particularly true regarding MVPA, which is widely overestimated (14). It is hypothesized that this results from a combination of social desirability and recall biases (15, 16). As older adults tend to perform their higher intensity PA in an unstructured manner their recall bias is generally more pronounced (17). As patients receive transplantation through a broad distribution of ages, they require a concise questionnaire that works similarly well for younger and older patients.

Rather than attempting to replace the IPAQ, we hypothesized that modifying the existing version would improve its accuracy without endangering its widespread acceptance. Accordingly, we made only minor changes to the questionnaire, but added a pre-measurement oral introduction to key concepts, plus descriptions clarifying the PA intensities asked for in the various items. We further hypothesized that this would reduce the respondents' recall bias, thereby enhancing their responses' accuracy (15, 17).

Our objectives were a) to assess the criterion validity and test-retest reliability of our modified version of the IPAQ-SF (mIPAQ-SF) in a sample of KTRs and b) to compare the results to those previously reported for the IPAQ-SF across healthy and other clinical populations. We hypothesized that, compared to the traditional IPAQ-SF, the mIPAQ-SF would show increased criterion validity, with test-retest reliability remaining similar.

MATERIALS AND METHODS

Design

This study includes a secondary analysis of data from an RCT investigating the effects of a self-management program on weight management immediately after kidney transplantation, including three study visits (i.e., 2–6 weeks, 8 months and 12 months after kidney transplantation) (18). Patients received no further intervention between months 8 and 12, were in a stable condition, and showed little variation between their PA levels (18).

Setting

The study was carried out at the University Hospital of Zurich. It was conducted in accordance with the declaration of Helsinki and all subjects provided written informed consent. The Ethics Committee of the Canton of Zurich approved the study (EK-ZH-NR: 2011-0411), which was registered on www.ClinicalTrials.gov (NCT02282124).

Population

The sample was drawn from the population of patients who received kidney transplantation at our center from May 2012 to February 2018 (94 months). Data were collected over the entire period. Patients screened for eligibility were adults (i.e., 18 years or older) who had received a new kidney transplantation (18). We included patients with a complete data set, namely, for whom at least 4 days of accelerometry data were available at month 8 and 12, as well as complete mIPAQ-SF data at month 8 and 12.

Sampling

The sample used for this analysis showed baseline characteristics similar to those of the Swiss KTR cohort study (19); therefore, it is considered generalizable to the Swiss KTR population.

Data Collection

Modified IPAQ-SF

Self-reported PA was assessed *via* the mIPAQ-SF at months 8 and 12 after renal transplantation. The mIPAQ-SF assessed the number of days on which patients performed vigorous activity, moderate activity and walking. Then, the usual time per day spent in each specific activity was assessed (as reported in 10 min bouts). Aiming to enhance the original IPAQ's test-retest reliability and criterion validity, we modified the questionnaire's introduction (see **Supplementary Material**).

This began by introducing the patients to the mIPAQ-SF's conceptual framework, i.e., orally explaining the four levels of intensity (vigorous, moderate, walking and sitting) in a fully structured manner, along with criteria for bodily indicators of these levels (e.g., changes in breathing, heart rate and sweating) and examples of typical activities characterizing each intensity. Whereas, the original IPAQ-SF provided only breathing criteria (e.g., moderate: breathing somewhat harder than normal; vigorous: breathing much harder than normal), we added criteria for heart rate (i.e., moderate: pulse slightly faster; vigorous: pulse substantially faster) and perspiration (i.e., moderate: moderate perspiration; vigorous: profuse perspiration) (see **Supplementary Material**). Following this oral introduction, patients were given time to reflect on their activity levels over the previous 7 days. Thereafter, the mIPAQ-SF items were assessed by a registered nurse in a structured interview format.

Accelerometry

Accelerometry data on step counts and intensity levels were recorded *via* the ankle-worn StepWatch 3 Activity Monitor (Orthocare Innovations, Seattle, WA, USA) at 8 and 12 months after RT. The StepWatch 3 is validated in rehabilitation settings and has shown acceptable accuracy in individuals with slow walking speed and short stride (20). Following published guidelines, PA was categorized into three intensity levels: low (1–30 steps/min), moderate (31–80 steps/min) and vigorous (80 or more steps/min) (21). Patients were instructed to wear the device on their left ankle for a period of 7 days. Wearing time was only interrupted while taking showers. As proposed in best practice guidelines, accelerometry was only considered valid for analysis if wearing time was at least 4 days with at least 22 h of wearing time per day (10, 22).

Data Analysis

In accordance with the Qualitative Attributes of PA (QAPA) checklist (23) and using accelerometry as the gold standard, Pearson correlation coefficients were used to investigate the mIPAQ-SF criterion validity regarding study visit data from month 12 (24). Intraclass correlation coefficients of type 3,1 ($ICC_{3,1}$) were used to investigate the mIPAQ-SF's test-retest reliability between the study visits at months 8 and 12 (24). Reporting of test-retest reliability was conducted in accordance with the Guidelines for Reporting Reliability and Agreement Studies (GRAAS) (25), and the QAPA checklist (23).

Criterion validity was reported in accordance with the QAPA checklist (23). Considering MVPA's importance in clinical practice and health prevention settings (4), as it is not directly assessed by the mIPAQ-SF, we gauged validity and reliability based on its calculated value (i.e., summing the minutes spent in moderate and vigorous activity). We did the same for total physical activity (TPA), including walking and MVPA, which were also calculated (12). Only positive correlations were present in this analysis. In accordance with published recommendations, they were divided into five classifications: negligible ($0 \leq r \leq 0.29$), low ($0.3 \leq r \leq 0.49$), moderate ($0.5 \leq r \leq 0.69$), high ($0.7 \leq r \leq 0.89$) and very high ($0.9 \leq r \leq 1$) correlation (26). In accordance with published recommendations, ICCs were divided into five classifications: poor ($0 \leq ICC \leq 0.20$), fair ($0.21 \leq ICC \leq 0.40$), moderate ($0.41 \leq ICC \leq 0.60$), strong ($0.61 \leq ICC \leq 0.80$) and near complete ($0.81 \leq ICC \leq 1$) agreement (27). To visualize agreement between the mIPAQ-SF domains regarding average minutes of PA per day and accelerometry measurements, as well as regarding test-retest reliability, we used Bland-Altman Plots. These depict agreement between two measurement methods by plotting the differences between individual data points and their means (28, 29). In addition, we reported Bland-Altman statistics, i.e., mean differences and 95% limits of agreement (i.e., 1.96 SD) to allow conclusions on the magnitude of bias in the measurements and agreement.

Unless otherwise stated, reported results are shown as means (with SDs) or medians (and interquartile ranges). Significance level was set at $p \leq 0.05$.

Statistical analyses were performed using R version 4.0.3 (R Core Team 2021, R Foundation for Statistical Computing, Vienna, Austria).

Procedure

Patients were asked to wear accelerometers for the seven days before their study visits in months 8 and 12 post-transplantation. At each study visit they were also asked to complete the mIPAQ-SF in a structured interview format by a trained study nurse.

RESULTS

Study Participants

Of the 123 patients participating in the study, 92 supplied complete accelerometry and mIPAQ-SF data for their follow-up visits. The sample consisted mainly of male (65%) patients with a median age of 55 (45–62) years. Baseline characteristics are presented in (**Table 1**); further sample information is published elsewhere (18). Detailed values of the data for each measurement time point is provided in (**Table 2**).

Criterion Validity of the Modified IPAQ-SF

For numbers of minutes spent in vigorous activity (per day) we found a negligible correlation between mIPAQ-SF reports and accelerometry records [r (95% CI) = 0.07 (−0.13, 0.26), $p = 0.52$]. We found low correlations between number of minutes spent in MPA [r (95% CI) = 0.35 (0.16, 0.5), $p < 0.001$], MVPA [r (95%

TABLE 1 | Patient characteristics ($N = 92$).

Variables	Value
Age, years (median, interquartile range)	52 (45–62)
Sex, male/female (numbers, %)	60/32 (65/35)
Body-Mass Index, kg/m^2 (median, interquartile range)	25.2 (22.4–27.7)
Steps per day, *n* (median, interquartile range)	5,196 (3,518–6,569)

kg/m^2, kilogram per squaremeter.

TABLE 2 | Values of the accelerometer and mIPAQ-SF measurements at 8 and 12 months.

Intensity	8 Months	12 Months
Accelerometer		
Minutes low	1,049 (979.9–1,095.3)	1,069.3 (983.4–1,135.6)
Minutes moderate	105.4 (75–131.3)	97.6 (70.5–126.1)
Minutes vigorous	23.1 (12.2–35.5)	20.7 (12.9–32.75)
Minutes moderate to vigorous (MVPA)	129 (89.2–164.5)	119 (79.3–157.3)
Days moderate	7 (7–7)	7 (7–7)
Days vigorous	5 (3–6)	5 (3–6)
mIPAQ-SF		
Minutes seated	270 (150–360)	270 (180–450)
Minutes moderate	60 (30–97.5)	60 (25–120)
Minutes vigorous	30 (0–60)	0 (0–60)
Minutes moderate to vigorous (MVPA)	105 (46.3–150)	75 (38.5–156)
Days moderate	5 (2–7)	5 (2–7)
Days vigorous	0 (0–2)	0 (0–2)

Values are median (interquartile range). mIPAQ-SF, modified International Physical Activity Questionnaire – Short Form.

TABLE 3 | Pearson correlation coefficient table for validity between accelerometry and the mIPAQ-SF.

Variables	r (95% CI)	*p*-value
Minutes vigorous	0.07 (−0.13, 0.26)	0.52
Minutes moderate	0.35 (0.16, 0.51)	< 0.001
Minutes moderate to vigorous (MVPA)	0.33 (0.14, 0.45)	0.001
Minutes low-moderate-vigorous (TPA)	0.34 (0.15, 0.51)	< 0.001
Days vigorous	0.15 (−0.05, 0.33)	0.15
Days moderate	0.11 (−0.10, 0.30)	0.31
Days walking	0.19 (−0.01, 0.37)	0.07

mIPAQ-SF, modified International Physical Activity Questionnaire – Short Form.

CI) = 0.33 (0.14, 0.45), $p = 0.001$], and TPA [r (95% CI) = 0.34 (0.15, 0.51), $p < 0.001$].

Regarding correlations between number of days with episodes of at least 10 min of different levels of activity, we found negligible correlation between mIPAQ-SF reported and accelerometry recorded number of days with bouts of at least 10 min of vigorous activity [r (95% CI) = 0.15 (−0.05, 0.33), $p = 0.15$], the number of days with bouts of at least 10 min of moderate activity [r (95% CI) = 0.11 (−0.10, 0.30), $p = 0.31$], and the number of days with bouts of at least 10 min spent walking [r (95% CI) = 0.19 (−0.01, 0.37), $p = 0.07$]. Correlations for validity are displayed in (**Table 3**).

The Bland-Altman analysis depicting criterion validity regarding average minutes of TPA per week shows a mean difference (95% CI) of 97.70 (56.73, 138.67) minutes, indicating significant underestimation of TPA per week in the mIPAQ-SF. Limits of agreement were from −296.50 to 491.90 min. Regarding average minutes per day, the mean differences (95% CI) were −4.20 (−13.23, 4.82) min for vigorous PA, 6.54 (−15.36, 28.44) min for moderate PA, and 5.88 (−17.94, 29.70) min for MVPA, indicating no significant over- or underestimation in the mIPAQ-SF. Limits of agreement were from −91.08 to 82.67 min for vigorous PA, from −206.41 to 219.49 min for moderate PA, and from −223.28 to 235.04 min for MVPA. Scattering of data gets wider with increasing number of minutes reported for all items. The results of the Bland-Altman analysis are displayed as Bland-Altman plots in (**Figure 1**).

Test-Retest Reliability of the Modified IPAQ-SF

Comparing the two follow-up tests (conducted in months 8 and 12), we found moderate agreement between reported numbers of minutes spent in vigorous activity [$ICC_{3,1}$ (95% CI) = 0.58 (0.47, 0.67), $p < 0.001$], walking [$ICC_{3,1}$ (95% CI) = 0.52 (0.40, 0.62), $p < 0.001$], TPA [$ICC_{3,1}$ (95% CI) = 0.48 (0.36, 0.59), $p < 0.001$], and sitting [$ICC_{3,1}$ (95% CI) = 0.55 (0.43, 0.64), $p < 0.001$]. We also found fair agreement between numbers of minutes spent in moderate activity [$ICC_{3,1}$ (95% CI) = 0.23 (0.09, 0.37), $p = 0.005$], and between the two reports' numbers of minutes spent in MVPA [$ICC_{3,1}$ (95% CI) = 0.28 (0.13, 0.41), $p < 0.001$]. Correlations for reliability are displayed in (**Table 4**).

The Bland-Altman analysis depicting test-retest reliability for average minutes per day of TPA shows a mean difference (95% CI) of 32.70 (−8.93, 74.32) min, indicating no significant over- or underestimation between the visits. Limits of agreement were from −390.94 to 456.33 min. However, for average minutes per day of vigorous PA, the mean difference (95% CI) falls to 14.99 (6.87, 23.10) min, indicating significant overestimation at visit 2. Limits of agreement were from −68.41 to 98.39 min. Regarding mean inter-test differences (95% CI) between average reported daily times were −3.10 (−27.80, 21.60) min for moderate PA, 11.29 (−15.32, 37.89) min for MVPA, and −29.84 (−63.79, 4.12) min for sitting, all indicating no significant over- or underestimation between the visits. Limits of agreement were from −259.31 to 253.11 min for moderate PA, from −260.76 to 283.32 min for MVPA, and from −383.64 to 323.97 min for sitting. The agreement does not diminish with increasing numbers in reporting. The results of the Bland-Altman analysis are displayed as Bland-Altman plots in (**Figure 2**).

DISCUSSION

This study investigated the criterion validity and test-retest reliability of a modified version of the IPAQ–SF, a physical

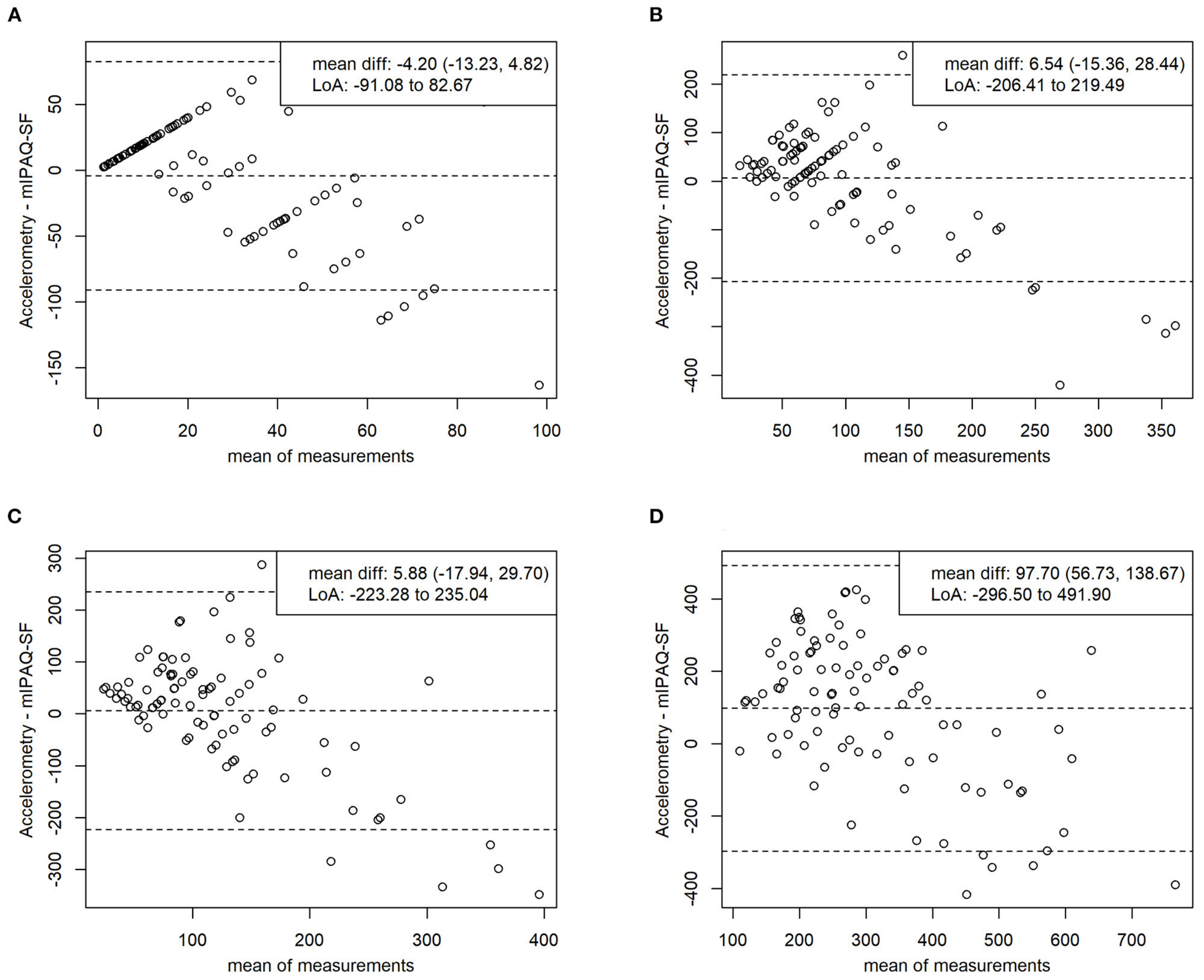

FIGURE 1 | Bland-altman plots validity. Comparing average minutes intensive per day **(A)**, average minutes moderate per day **(B)**, average minutes moderate to vigorous per day **(C)**, average minutes total physical activity **(D)** between mIPAQ-SF and accelerometry. Accelerometry – mIPAQ-SF: difference between methods; mIPAQ-SF: modified International Physical Activity Questionnaire – Short Form; mean diff: mean difference with 95% confidence interval; LoA, limits of agreement (1.96 SD).

activity (PA) self-report instrument, in a sample of (*n* = 92) KTR. The mIPAQ–SF version adds a structured oral introduction explaining the instrument's key concepts. This and other minor modifications were aimed at enhancing the earlier version's low criterion validity as well as its test-retest reliability.

According to WHO recommendations, time spent in MVPA is particularly important for health-related outcomes (4). In fact, combining data on these two activity levels into a single score—MVPA—is a technique commonly used in clinical practice. Regarding patient-reported data, it has been suggested that MVPA also offers greater recall accuracy than either of its separate domains. Comparing patient-reported mIPAQ-SF data on MVPA to accelerometry records, we found significant low correlation, i.e., criterion validity. Regarding test-retest reliability, the evidence indicated fair agreement in this domain.

In our analysis, comparing the mIPAQ-SF's criterion validity to that of the IPAQ-SF, MVPA was the domain whose validity improved most (r = 0.33 vs. r = 0.18) (13). In addition, no significant bias was shown in the Bland-Altman analysis, indicating improved validity as compared to the IPAQ-SF (14). However, agreement is highest in individuals with low level of MVPA. As physical activity acts as an independent predictor of weight gain and glucose tolerance in kidney transplant recipients (30, 31), this is an important finding with practical implications for the transplant setting. Regarding the modified instrument's increased validity regarding MVPA, the mIPAQ-SF's combination of low cost, convenience and especially validity make it a promising alternative to accelerometry in clinical practice (4). Considering test-retest reliability, our results indicate fair agreement regarding MVPA. This was lower than previously reported (12, 13).

For moderate PA, compared with accelerometry results, the mIPAQ–SF showed improved criterion validity ($r = 0.35$ vs. $r = 0.30$) (12, 13). However, compared to the traditional form, it showed lower validity regarding minutes spent in vigorous activity ($r = 0.07$ vs. $r = 0.31$). In addition, Bland-Altman analysis of test-retest reliability showed significant bias. We can only speculate on the underlying reasons for this. However, as intensity thresholds commonly implemented in accelerometry reflect healthy population data, we hypothesize that patients with chronic disease may experience, and accordingly classify, vigorous activity differently. Our increased use of bodily symptoms to determine activity levels might have increased this discrepancy.

TABLE 4 | ICC table for reliability of the mIPAQ-SF between 8- and 12-month follow-up.

Variables	$ICC_{3,1}$ (95% CI)	*p*-value
Minutes vigorous	0.58 (0.47, 0.67)	< 0.001
Minutes walking	0.52 (0.40, 0.62)	< 0.001
Minutes walking-moderate-vigorous (TPA)	0.48 (0.36, 0.59)	< 0.001
Minutes sitting	0.55 (0.43, 0.64)	< 0.001
Minutes moderate	0.23 (0.09, 0.37)	0.005
Minutes moderate to vigorous (MVPA)	0.28 (0.13, 0.41)	< 0.001
Days vigorous	0.34 (0.20, 0.46)	< 0.001
Days moderate	0.19 (0.04, 0.03)	0.017
Days walking	0.25 (0.10, 0.38)	0.003

mIPAQ-SF, modified International Physical Activity Questionnaire – Short Form; ICC, intraclass correlation coefficient.

The strength of this study is its investigation of the criterion validity of two identically scaled activity count-based measures (i.e., minutes spent at a defined intensity) in their respective questionnaires as compared to the gold standard. Furthermore, we were able to analyse all of the questionnaires' subdomains except the validity of items regarding time spent sitting, as the accelerometer used could not record this.

This study also has notable limitations. The mIPAQ-SF requests information on PA during a "usual week," aiming to obtain an individually generalizable conclusion on PA levels. In contrast, accelerometry recordings may have recorded unstructured and intermittent PA that is unusual for the individual participant, not covering one of the "usual weeks" and therefore not delivering generalizable information. Accordingly, our study might have underestimated the mIPAQ-SF's validity. Secondly, despite the participant's apparent lack of changes regarding either PA or demographics between their visits at months 8 and 12 post transplantation (18), other changes may have occurred, influencing factors related to PA and not covered by our assessments. Last, the accelerometer used lacks validation studies regarding the different intensity thresholds implemented. To the best of our knowledge, existing validation literature focuses exclusively on the validity in step count.

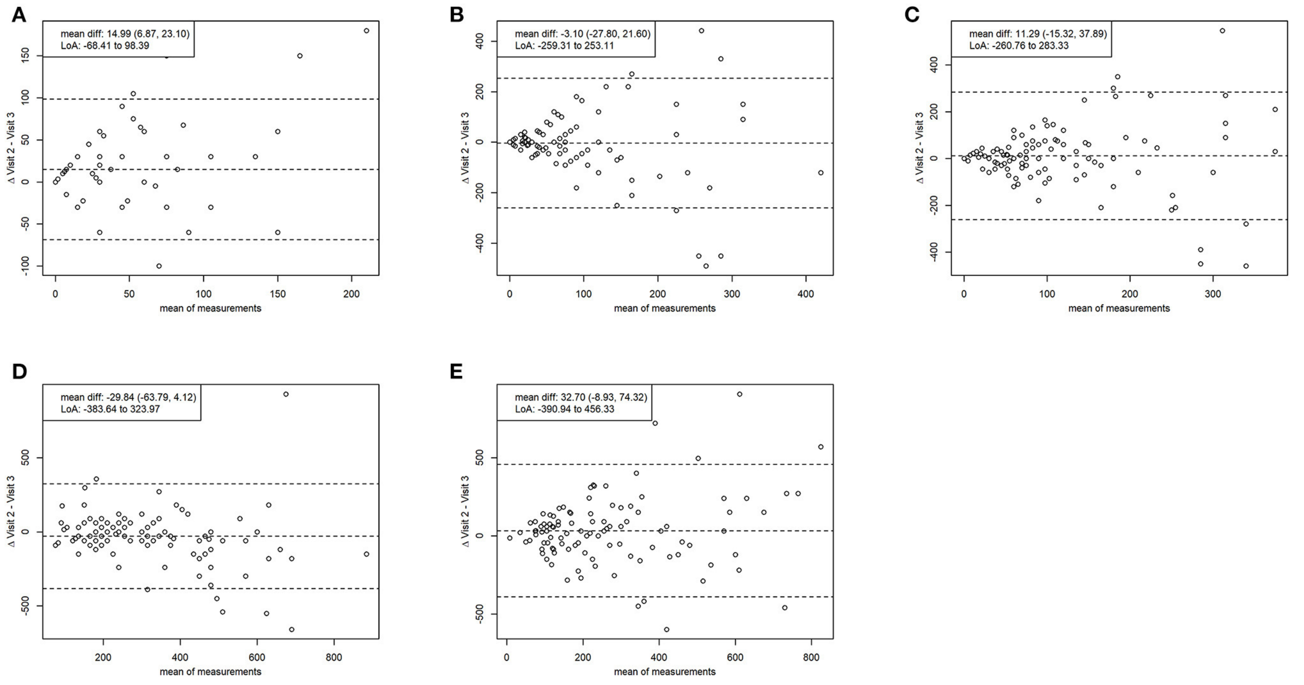

FIGURE 2 | Bland-altman plots reliability. Comparing average minutes intensive per day **(A)**, average minutes moderate per day **(B)**, average minutes moderate to vigorous per day **(C)**, average minutes sitting per day **(D)**, average minutes total physical activity **(E)** between the two measurement time points. Mean diff., mean difference with 95% confidence interval; LoA, limits of agreement (1.96 SD).

The development of well-tolerated customer-grade PA monitors is a very dynamic and rapidly developing field. There remains the possibility that accurate, robust, well-tolerable, and reasonably priced accelerometers become available in due time. However, we still believe that activity questionnaires remain a rapid and easily applicable tool in clinical rehabilitation. It remains to be studied if combining objective and subjective activity monitoring would add valuable information for clinicians.

In conclusion, compared to the original IPAQ–SF, the mIPAQ–SF may be more accurate and therefore more suitable for patients when MVPA is of special interest—as is the case in KTR. However, it remains to be considered that validity, although improved with the mIPAQ-SF, remains mostly in the low spectrum. The mIPAQ-SF is less suitable for populations in which vigorous PA is of interest; and further investigation in larger samples would be necessary before broad application. Accordingly, clinicians should always consider if the application of an accelerometer is feasible and appropriate in the first place.

Nevertheless, considering the IPAQ-SF's importance as a PA outcome measure, the modified version's improved validity regarding MVPA and sufficient test-retest reliability are promising developments. As a further implication for future research, we would recommend investigating the mIPAQ-SF in other populations.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of the Canton of Zurich, Switzerland. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

GS-M: design and data collection. DK: data analysis and manuscript writing. DK, GS-M, and SM: interpretation and manuscript revision. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2022.808476/full#supplementary-material

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Interventions for social and community participation for adults with intellectual disability, psychosocial disability or on the autism spectrum: An umbrella systematic review

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Objective: This umbrella systematic review examined the effectiveness, facilitators, and barriers of interventions for social, community and civic participation for adults on the autism spectrum, or with intellectual or psychosocial disability.

Data Sources: Eight databases were searched to identify eligible reviews defined by the: Sample (≥50% adults on the autism spectrum or with intellectual or psychosocial disability), Phenomena of Interest (interventions in community settings that aimed to improve social, community or civic participation, or capacity to participate), Design (any), Evaluation (any method that evaluated impacts on participation or capacity to participate), and Research type (reviews as journal articles, dissertations or in grey literature, in English, published 2010-2020).

Review Methods: Rapid review methods were used. One researcher screened 27,890 records and 788 potentially eligible full texts. A second reviewer independently screened 20% of records, and ambiguous full text publications. Study quality was extracted, and review quality was assessed with the Assessing Methodological Quality of Systematic Reviews (AMSTAR) checklist. Data from 522 studies in 57 eligible systematic reviews were extracted for narrative synthesis. The Corrected Covered Area (CCA) was calculated to indicate overlap between reviews.

Results: There was a pooled sample of 28,154 study participants, predominantly from studies in North America, the UK and Europe. There was very low overlap between reviews (CCA = 0.3%). Reviews were predominantly low quality: 77.2% of reviews met <50% of AMSTAR criteria. Most studies were low (45.4%) or moderate (38.3%) quality. Three broad intervention categories improved participation, inclusion and belonging outcomes: (1) interventions to help people identify and connect with participation opportunities (e.g., person centred planning); (2) participation opportunities or activities (e.g., joining a community group, sports or outdoor activities, or arts-based activities); and (3) supports to build skills and capacity to participate socially and in the community.

Conclusions: The evidence highlighted that improved social and community participation requires purposeful strategies that identify meaningful participation preferences (e.g., where, when, how, and with whom) and provide support to build capacity or enable ongoing participation. Community capacity building, peer support and advocacy may also be needed to make the community more accessible, and to enable people to exercise genuine choice.

KEYWORDS

disability, community, participation, inclusion, belonging

1. Introduction

Social, Community and Civic participation have important benefits for people with disabilities, as well as for their family and carers, including improved wellbeing (1) and increased study, volunteering or paid employment opportunities (2). There are also likely to be broader social benefits including improved social capital and accessibility for all members of the community when social settings are more accessible and welcoming to everyone in society, including people with disabilities (3).

Social, Community and Civic participation is considered one of the core domains of the International Classification of Functioning (ICF), Disability, and Health framework (4), which recognizes the important relationships between disability, function, the environment, and health. While most of the specified domains in the ICF framework focus on individual activities or functions rather than participation, Chapter 7 (Interpersonal Interactions and Relationships) and Chapter 8 (Major Life Areas) outline key aspects of participation (5). For this review, we considered participation in line with the Convention on the Rights of Persons with Disabilities (CRPD) (6), defined as the rights to: full inclusion and participation of people with disability in the community (**Article 28**); effective and full participation in political and public life (**Article 29**); participation in mainstream and disability-specific sporting and recreational activities at all levels to the fullest extent possible (**Article 30**); and access to sporting, recreational and tourism venues or services for organizing recreational, tourism, leisure and sporting activities (**Article 30**). Participation and related outcomes were conceptualized as activities that: (a) are ideally chosen or desired by the individual with a disability; (b) occur in a social, community or civic setting; and (c) enable people with disabilities to participate alongside and/or with people without disabilities, or to build the skills, self-efficacy, or social networks to enable participation alongside/with people without disabilities (7).

In Australia, the most prevalent barriers to social and community participation are experienced by people living with Autism Spectrum Disorder, Intellectual and Psychosocial Disabilities (8). *Autism Spectrum Disorder* is a developmental condition that includes persistent deficits in social communication and interaction across multiple contexts; restricted, repetitive patterns of behavior, interests, or activities; and disturbances that cause clinically significant impairments (9). *Intellectual Disability* is defined as a disability that originates before the age of 18 with significant limitations in intellectual functioning, with an IQ < 70, and impairments in adaptive behavior related to many everyday social and practical skills (10). *Psychosocial disability* is a term used to describe disabilities arising from mental health conditions that are “multi-axial” comprising psychological, social, and occupational impacts of psychiatric, psychological, or developmental disorders (11). Co-occurrence of two or more of these disabilities is common. For instance, adults on the autism spectrum have higher rates of psychiatric comorbidity (12), and 50%–60% of people on the autism spectrum also have an intellectual disability (13).

Consistent with the social model of disability (14), difficulties with participation often arise due to both societal and environmental factors including availability and access to transport in the community, accessibility of information and buildings, and community perceptions, actions and attitudes. Moreover, individual factors play a role, including health, mobility, poverty, communication, support from family/carers or friends, confidence, life experience, and interests (15). Finally, participation can be obstructed by the systemic exclusion of people with disabilities, availability of supports for disability needs, and lack of support for, or access to, education or employment (15).

To enhance social, community and civic participation for people on the autism spectrum, or with intellectual or psychosocial disability, we must identify and enable access to interventions that overcome the social, individual, and systemic barriers to participation. Such interventions may work by improving the fit between the person and their physical, social, or institutional environments (e.g., by making the environment more accessible to enable their participation), or by building the capacity of the individual with a disability to participate. Therefore, this review sought to systematically identify and synthesize the available evidence for the effectiveness of interventions or supports that aim to improve social, community and civic participation of adults

TABLE 1 SPIDER eligibility criteria for the umbrella review.

Domain	Inclusion criteria	Exclusion Criteria
Sample	• ≥50% of participants aged 18 + years on autism spectrum, with intellectual or psychosocial disability • Living in the community, including small group homes/supported living	• People living in large group homes • People with acquired intellectual or cognitive disabilities • ≥50% of participants were secondary school students.
Phenomena of Interest	• Interventions, supports or programs in a community setting or that aimed to influence social, community or civic participation or capacity to participate.	• Interventions with a medical basis or focused on management of symptoms for delivery in the health system, consistent with previous reviews (17). • Reviews of Cognitive Remediation interventions as a separate evidence snapshot specific to this topic was already underway. • Reviews of employment or residential interventions that did not include an active social or community participation component as separate reviews were already commissioned to evaluate that evidence. • Interventions targeting the environment or community, which were beyond the scope of this review.
Design	• Systematic, scoping, rapid reviews of quantitative or qualitative research • Studies measured ≥1 capacity or participation outcome or intervention acceptability/feasibility	• Primary research studies not included in the systematic and scoping reviews.
Evaluation	• Quantitative and case study designs must evaluate a within or between group change in participation or capacity • Qualitative studies explored participation experiences, acceptability, barriers and facilitators or outcomes	
Research type	• Published in English from 2010 to 2020 • Journal articles, dissertations/theses, and grey literature	• Books, book chapters, editorials, letters, conference abstracts, organizational website content, or publicity materials from disability services due to potential conflicts of interest.

on the autism spectrum, or who have intellectual or psychosocial disabilities. Broad inclusion criteria were defined; however, in accordance with the registered protocol, the study was conducted as an umbrella review given that many systematic reviews were identified. The review aimed to answer three overarching research questions:

1. What interventions are effective for who, how, under what conditions, for which activities, and for what outcomes?
2. Where the evidence is sufficiently strong and consistent for implementation: (a) what is the acceptability of the interventions; (b) what are the barriers and facilitators of intervention implementation; (c) what resources are required for implementation; and (d) is there evidence of cost-effectiveness?
3. What are the gaps in evidence?

2. Method

The protocol for the review was registered to PROSPERO on 6th January 2020 (CRD42021229580). Minor protocol deviations are outlined in **Supplementary File 1**.

2.1. Eligibility criteria

As this review sought to synthesise evidence from studies that used a broad range of methods, we used the Sample, Phenomena of Interest, Design, Evaluation, Research type (SPIDER) framework (16). Detailed eligibility criteria are provided in Supplementary File 1. Publications were eligible for inclusion if they met the criteria outlined in **Table 1**, and if they were published between 2010 and 2020. This timeframe coincides with the increased use of systematic review methods and allows for the identification of contemporary empirical evidence as well as older studies published since deinstitutionalization and the independent living movement.

Publications could use umbrella, systematic, scoping, or rapid review methods, if they (a) included a clear statement of the purpose of the review; (b) described the search strategy, searched two or more databases, described the search terms used and the inclusion/exclusion criteria; (c) presented data on search and screening results, and presented all findings relevant to the main purpose of the review.

Primary outcomes were aspects of social participation (e.g., communication, social relationship maintenance, participation through telecommunications or online platforms, convivial encounters); social networking (e.g., friendships, relationships, networks); navigating or accessing the community (e.g., access or skills to use public or private transport); participation in recreation, sports and leisure activities in the community (e.g., sports, art, music, community or cultural events, libraries, tourism); or civic involvement (e.g., voting, volunteer work, advocacy, committee or club memberships, or political engagement). Studies that only measured housing or employment outcomes, or leisure activity participation with no social and community participation potential were not eligible. *Secondary outcomes* included aspects of psychosocial functioning (e.g., self-determination, autonomy, choice, decision-making, self-advocacy), physical or mental health, or quality of life.

2.2. Search strategy: Databases and search terms

Medical Sub-Heading (MeSH) and keyword search terms for autism spectrum disorder, intellectual disability, and psychosocial disability and social, community and civic participation were adapted for eight search engines (**Figure 1**; the search terms are available in **Supplementary File 2**). Grey literature was identified using the DuckDuckGo search engine, which does not track search terms, and reduces the chance that reviewers are presented with biased website results based on previously viewed sites. Web screening was limited to the top 50 results for each disability population. Reference lists of included publications were hand searched to identify additional reviews, and an expert panel was consulted to identify any missed literature.

2.3. Study selection

Screening was conducted using Endnote, Covidence and Abstrackr. Abstrackr is a web-based platform that uses an active machine learning algorithm of reviewer judgements to predict the relevance of remaining citations, which are then sorted by predicted relevance to enable rapid identification of relevant records (18, 19). Search results were first consolidated in an Endnote library, and duplicates were removed. Due to the breadth of the review results once duplicates were removed, clearly ineligible citations were omitted when reviewing citation title, based on studies with ineligible disorders, paediatric populations, questionnaire validation methods, biomarker and neurophysiology studies, and document type, consistent with previous large-scale reviews (20). Study selection was undertaken in accordance with Cochrane Rapid Review methods (21) as follows. Reviewer 1 screened all citations in Endnote, and all full text articles in Covidence. Reviewer 2 screened in Abstrackr for the first 20% of citations predicted to be relevant, or until no further citations had > 50% relevance, whichever threshold was reached first. Systematic Review authors were contacted for additional information to determine eligibility of full texts, where necessary. If the reviewers were unsure about full text eligibility, a final decision was made in consultation with a third reviewer.

2.4. Data extraction

Data were extracted into excel spreadsheets (**Supplementary Data file 1** and **2**). Consistent with rapid review methods data extraction was completed by a single reviewer, and extraction accuracy and completeness were discussed between authors. Original study papers were accessed, or authors contacted, if key details were not provided in the systematic review.

Data extraction was conducted in two phases. In phase one summary information on the systematic reviews was extracted including: the review aim, design, key theoretical frameworks, and review inclusion and exclusion criteria; study selection and the number of studies that met our SPIDER criteria; pooled sample characteristics in each review for eligible studies (number, age, sex, disability types, countries); summary of interventions; design of eligible studies; type of control groups; and overall quality or risk of bias of the included studies. Overall effects on participation capacity, participation, quality of life, and secondary outcomes were recorded, and summarised as positive effects if ≥60% of studies had positive effect, negative effects if ≥60% of studies had negative effect, null effects if ≥60% of studies had null effect or inconsistent effects if no effect direction met the threshold for positive, negative or null effects. For meta-analyses, the inclusion of sensitivity analyses and identified biases were recorded. Study heterogeneity and whether review authors disclosed funding sources and conflicts of interest were recorded.

In phase two, information about the individual studies included in the systematic reviews was extracted, including: first author, publication year and country; study recruitment strategy and sample demographics (e.g., sample size, age, sex, disability or diagnoses); study design (e.g., descriptive or cross-sectional, mixed or multi-methods, multiple baseline case study, randomised controlled trial (RCT) or quasi RCT, non-randomised controlled trial with (NRCT-CG) or without a control group (NRCT-NoCG), qualitative, or review); intervention and control conditions (e.g., design, mode of delivery, the agent providing the intervention, the services provided, the duration and frequency of sessions in the intervention); and whether the intervention was in a disability-specific or mainstream setting. Effects of each intervention were extracted for social participation, capacity, and “other” outcomes, with complete data on the measures used and any effects of the intervention (available in Supplementary Data File 2). Data regarding cost-effectiveness and barriers or facilitators were documented where possible.

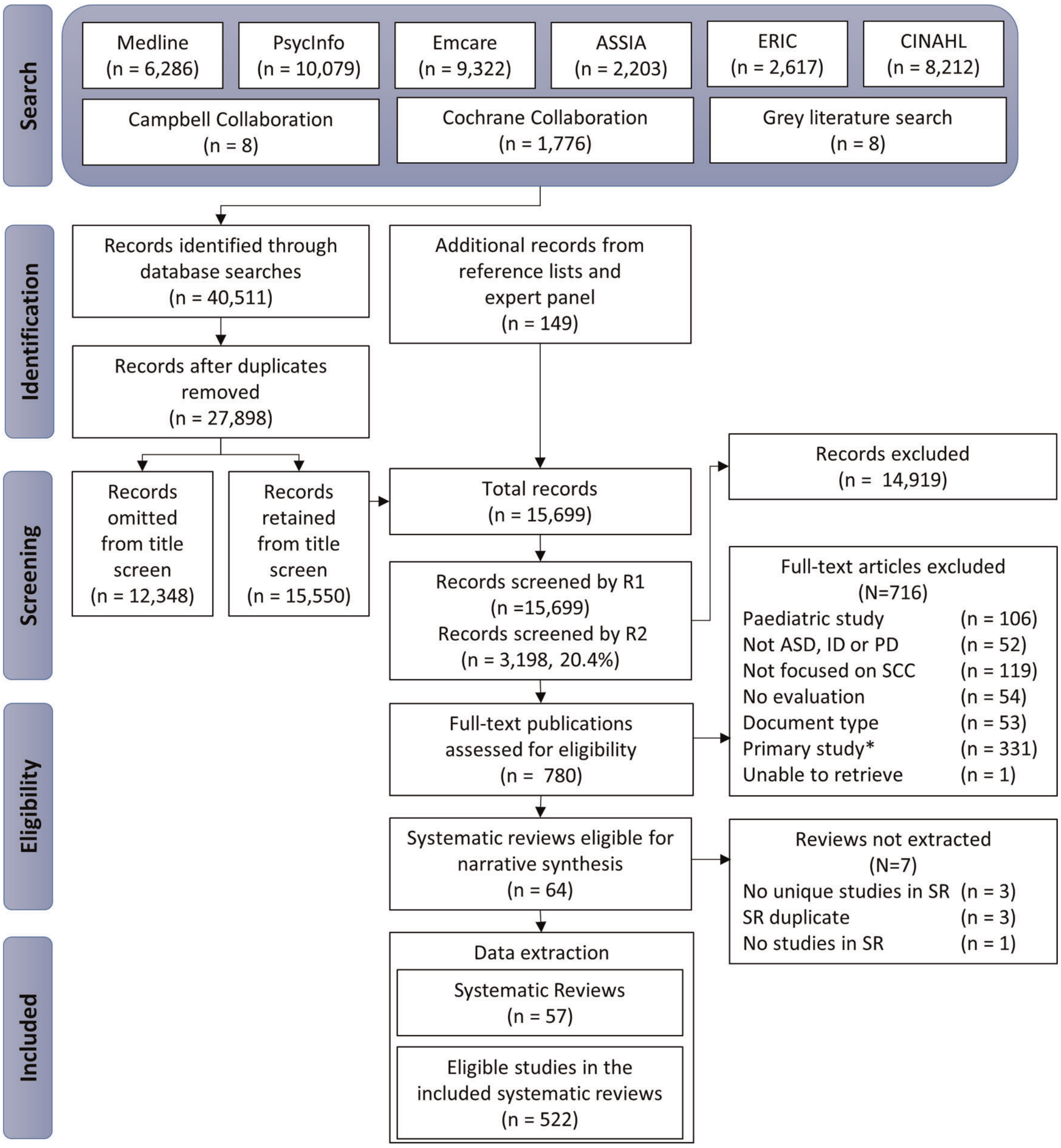

FIGURE 1
PRISMA chart. * These studies met the SPIDER study design eligibility criteria but did not meet the criteria for inclusion in an umbrella review.]

2.5. Quality assessments

Study quality, certainty, or risk of bias were extracted from included reviews where possible. Review quality was assessed using the 16 quality criteria in the checklist for Assessing the Methodological Quality of Systematic Reviews Version 2 (AMSTAR; 22). The proportion of relevant AMSTAR criteria that were met was calculated to summarise overall review quality. Review and study quality were generally classified as low, moderate, or high according to the original study classification, or based on tertiles of AMSTAR summary scores, respectively (e.g., studies that met <33.3% of quality criteria were considered low quality, but those that met >66.7% of quality criteria were considered high quality).

2.6. Data synthesis

The findings across systematic reviews are presented in a narrative synthesis of the characteristics of the included reviews, interventions, outcomes and effects on outcomes, and evidence quality (Research Question 1). To determine whether there was sufficiently strong and consistent evidence to support each intervention type we considered the consistency in the effectiveness of studies for the respective intervention category relative to the quality of the studies (Research Question 2).

As multiple systematic reviews may have included data from the same original studies, the Corrected Covered Area (CCA) was calculated across all studies, and studies on similar broad topic areas (e.g., social skills training) to provide insight into the level of overlap of original publications (23). The level of overlap was considered slight (<5%), moderate (6%–10%), high (11%–15%) or very high (>15%; 23). The following formula was used to calculate the CCA:

$$CCA = 100 \times \frac{(N - R)}{((R \times C) - R)}$$

N = number of publications including double counting; R = number of index publications; C = number of reviews

3. Results

3.1. Study selection

Study selection is summarised in **Figure 1**. A total of 40,644 records were identified from the search that was executed on 22 December 2020, including 149 records from reference lists and expert guidance. Data were extracted from 57 reviews that did not completely overlap with other included reviews (Supplementary Table 1). These reviews included a total of 1,170 original studies, of which 522 met the SPIDER inclusion criteria. Most included publications were systematic reviews (40 reviews), followed by meta-ethnographies (6 reviews), meta-analyses (5 reviews), Cochrane reviews (2 reviews), scoping reviews (2 reviews), and umbrella reviews (2 reviews).

The studies included in each review included various research designs, including: RCT or quasi RCT (167 studies); qualitative (112 studies), mixed or multi-methods (43 studies); non-randomised controlled trials with (42 studies) or without a control group (64 studies); cross-sectional studies (13 studies); and descriptive (17 studies) or multiple baseline case studies (35 studies). Sixteen of the 522 studies were systematic reviews from the two umbrella reviews (24, 25). Study design was not clear for 13 studies.

3.2. Study characteristics

3.2.1. Corrected covered area (CCA)

The overall CCA was 0.29%. Studies in four intervention categories had no overlap (i.e., travel and navigation training, art interventions for psychosocial disability and intellectual disability, parenting role training and vocation focused interventions). The remaining interventions had slight overlap (CCA median = 1.8%). Three topics that had high or very high overlap were social skills training for people on the autism spectrum (CCA = 10.2%; 17/44 studies included in 2–5 reviews), transition programs for people with intellectual disability or on the autism spectrum (CCA = 16.7%, 2/3 studies included in two reviews), and animal interventions for people with psychosocial disability (CCA = 33.3%, 2/6 studies included in two reviews).

3.2.2. Intervention settings and outcomes

The 522 eligible studies included a pooled sample of 28,154 people with disability. Interventions focused on psychosocial disability (31 reviews, 311 studies), intellectual disability (23 reviews, 139 studies), or the autism spectrum (15 reviews, 85 studies). Eleven reviews (13 studies) included people with more than one disability type.

Thirty-nine (68.4%) reviews reported the country of 346 studies. The most common global regions were North America (31 reviews; 153 studies), the United Kingdom and Ireland (26 reviews; 96 studies), Europe (22 reviews; 38 studies), Australia (18 reviews; 37 studies), Asia (10 reviews; 7 studies), Middle East (7 reviews; 11 studies), South America (1 review; 1 study) and New Zealand (1 review; 1 study).

The most common social and community functioning outcomes were: loneliness (22 studies), isolation (11 studies) or inclusion (13 studies); social functioning (17 studies), social disability (3 studies), social acceptance (3 studies), socialisation (6 studies); social networks, including network size (17 studies) or composition (4 studies), interpersonal, social or peer relations (33 studies) friendships (22 studies), contact with friends (6 studies) or other social interactions (15 studies), and social support (21 studies). Assessment of actual participation in the community was less common, but included social (7 studies), leisure (5 studies) or community activity participation (3 studies); community involvement or participation (8 studies), access to community venues (4 studies), and confidence to be in the community (3 studies).

The most common capacity-focused outcomes were theory of mind (i.e., the ability to recognize and understand the mental states of others; 22 studies), affect recognition (20 studies), attribution style (9 studies), and empathy (eight studies); social (15 studies) and communication skills (six studies); and dating knowledge or sex-related behaviours (18 studies), and social knowledge (eight studies).

The most common "other" outcomes were: psychiatric (50 studies), depression (31 studies) or anxiety symptoms (9 studies); quality of life (43 studies) or general wellbeing (9 studies); mental health (10 studies) or emotional wellbeing (7 studies); physical health (5 studies); self-esteem (20 studies), confidence (17 studies); self-value (11 studies); self-efficacy (10 studies); self-determination (8 studies); empowerment (8 studies); choice (4 studies); challenging behaviours (13 studies); adaptive behaviour (4 studies); cognitive functioning

(6 studies); employment (5 studies); and fitness (4 studies), sporting skill (4 studies) or other health-related outcomes (4 studies).

No studies examined cost-effectiveness.

3.2.3. Quality of the evidence

Study quality is summarised in **Table 2**–**4**, and Supplementary Table 2. Forty-four reviews (77.2%) met <50% of AMSTAR quality criteria (median proportion of criteria met = 0.41; Q1 = 0.19, Q3 = 0.46). Nineteen reviews (33.3%) met <25% of quality domains, 25 (43.9%) met 25%–49% of domains, seven (12.3%) met 50–74% of domains and four met 75%–100% of quality domains. Quality domains that were most often not described or that were low quality were: lacking a-priori protocol (51 reviews, 89.5%); poor or lacking explanation of study selection procedures (31 reviews, 54.4%); single author screening records (31 reviews, 54.4%) or extracting data (44 reviews, 77.2%); no report of the full text records excluded (50 reviews, 87.7%); no report of the study funders (55 reviews, 96.5%); no risk of bias assessment or failure to account for risk of bias in the synthesis (45 reviews, 78.9%); and no examination or discussion of heterogeneity (32 reviews, 57.9%).

The evidence included in each review was predominantly low (22 reviews; 51.2% of reviews reporting study quality) or moderate quality (14 reviews; 32.6%). Only seven reviews predominantly included high quality evidence. Studies evaluating interventions for people with psychosocial disabilities were generally higher quality than studies of interventions for people with intellectual disability or on the autism spectrum. Quality was not assessed in 14 reviews and was unclear in one review (26).

3.3. Intervention design and outcomes

There were three broad types of intervention: (1) 12 interventions that helped people identify and connect with participation opportunities; (2) 14 interventions that were a participation opportunity; and (3) 31 interventions that focused on building skills or capacity to participate. Intervention outcomes are summarised in **Tables 2**–**4**, and detailed information on each intervention, study design and quality, and effects are provided in **Supplementary Table 3**. Facilitators and barriers are summarised in **Supplementary Table 4**. Category numbers below correspond to the intervention category in all tables.

3.3.1. Interventions to help people connect with social, community or civic participation opportunities

Interventions focused on supporting people to connect with social or community participation opportunities used processes like person centred planning and individualized support for social functioning and participation; befriending or peer-based supports to broaden social networks; and transition supports for younger and older adults (**Table 2**).

Person centred planning (PCP) involves developing individualised plans with the person in partnership with their circle of support, with a focus on meaningful participation and goals (27). The PCP interventions (Category 1, low-high study quality) were primarily assessed in people with intellectual disability or on the autism spectrum in residential or day centre settings. PCP reduced loneliness and improved self-determination, interpersonal relations, social inclusion, contact with friends, and sense of connection and social contact, participation and involvement in community settings (e.g., restaurants, museums) and in community activities. PCP increased the variety of community locations visited, and level of access to community settings. There were inconsistent effects on social network size, no impacts on friendships with peers or social networks beyond close family and staff. There were 2.8-fold higher rates of participation in choice-making for short-term goals (e.g., whether to participate in specific activities) (28), but no impact on involvement in major life decisions (29). Plans were not developed for 30% of people in one study (28). Study quality varied enormously, and successful implementation required support from frontline staff providing individualized support through to service planners and managers (29).

Skilled individualised supports (Category 2, study quality not reported) for people on the autism spectrum or with intellectual disability encompassed *active support*, positive behaviour support, behavioural and residential assessments and modifications, functional communication training, planning, respite care, and crisis responses. Individualised support led to increased time in community settings and activities, facilitated convivial encounters in community settings, and reduced barriers to community interactions. Individualised support increased participation in employment or training but did not affect the incidence of challenging behaviour.

A previous umbrella review (25) summarised effects of interventions supporting *choice-making and asset-based approaches, social skills, setting goals and peer support* from one systematic review (Category 3, moderate quality study; frequency n/a). These interventions enhanced social inclusion, connectedness and quality of life, and reduced depression. Asset-based approaches improved self-esteem and health outcomes but had inconsistent effects on self-determination. Goal setting interventions required strong relationships between staff and participants, and were not as effective as interventions with asset-based approaches or that built social skills (30).

Animal-based interventions (Category 4) included a dog walking program alongside a dog handler for people with intellectual disability (high quality study), and short-term animal *companionship* for people with schizophrenia or depression (low-moderate study quality). Walking a dog

TABLE 2 Overall effects of intervention processes and supports to help people connect with social, community or civic participation opportunities, including AMSTAR quality rating of the SRs and quality of the original studies.

Intervention • ***No. of studies*** • ***A* for each SR reporting studies***	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social Skills**	**Other outcomes**
1. Person Centred Planning • 10 studies • A*: L L L M	ID, $N = 640$, 13–85 years, sex nr	≥1 individual or group meeting, no set frequency	+** [L-H]		+** [L-H]
2. Individualised behaviour and participation support • 5 studies • A*: L M	ID, $N = 65$, 14–39 years, sex nr	As needed to generate & review plans	+ [nr]	o/+ [nr]	+ [nr]
3. Choice-making and "asset-based" approaches • 1* study • A*: L L	ID, $N = 2$*, 69 years, sex nr	Nr	+ [M]	o/+ [M]	+ [M]
4. Animal companionship with dog walking or in home • 4 study • A*: L L H	ID, $N = 106$, aged 18–64 years, sex nr	14 × 1-hr walking sessions Continuous or 50 min to 3-hrs p/wk	o/+ [L-H]		+ [M]
5. Community group participation linkage supports • 4 studies • A*: L L M M	ID or PSD, $N = 13$, N nr for 2 studies, age, sex	30-min staff introduction or 30 h of meetings with a recreational therapist over 9-10 weeks	+ [M]		
6. Social prescriber and "connecting people" interventions • 20 studies • A*: L L M M	PSD, $N = 357$, sample was nr for 16 studies, age, and sex nr	≥1 assessment plus 1–5 additional contacts, over 3–18 months	+ [L-H]	+ [nr]	+** L-H]
7. Befriending interventions with a non-disabled volunteer • 2 studies (ID), 5 studies (PSD) • A*: M M (ID), L M M (PSD)	ID, $N = 38$, age, and sex nr PSD, $N = 637$, age and sex nr	nr 2 h/wk for 6wks to 12 months	-/o/+ [L] o/+ [M-H]		 o/+ [M-H]
8. Peer-based friendship programs • 5 studies • A*: L L M	PSD, $N = 489$ with a range of mental health diagnoses, age nr, 60% male	35–38 × 3-hr sessions (nr for most studies)	o/+ [L-M]		o/+ [L-M]
9. Peer support in the community • 9 studies (PSD), 1 study (ASD), 1 study (ID) • A*: L L M M M M H H (PSD), M (ASD), M (ID)	PSD, $N = 1,337$, age and sex nr ASD, $N = 35$, 24–77 years, 69% male ID, $N = 10$, 19–48 years & 30% male	1.5 to 2-hr sessions for 4-weeks up to 12-months	o/+ [M-H] + [H] + [M]		o/+ [M-H]
10. Peer support in mental health services • 36 studies • A*: L M H	PSD, $N = 340$ PSD mentors, $N = 2,152$ PSD mentees, $N = 138$ staff, age, and sex nr	Typically 2.5 h per week	o/+ [L-H]		o/+ [L-H]
11. Transition to young adulthood • 13 studies • A*: L L M M	ID or ASD, $N = 210$, aged 17–33 years, sex nr	Camp Campus for 1 wk or 10-month program (frequency & length nr)	+ [L-H]		
12. Transition to retirement • 2 studies • A*: L M	ID, $N = 17$, aged 48–62 years	weekly for 5–10 months (session length nr)	+** [M]		o/+ M]

Notes: Blank cells indicate no evidence available. Detailed summaries of the study and sample characteristics and outcomes are provided in Supplementary Table 4, the numbered intervention categories in column one corresponds to the numbered paragraphs below and the numbered rows in Supplementary Table 4.

Abbreviations: L, low quality; M, moderate quality; H, high quality; nr, not reported; A*, AMSTAR (A MeaSurement Tool to Assess systematic Review); ASD, autism spectrum disorder; ID, Intellectual Disability; PSD, Psychosocial Disability; SR, systematic review; UR, Umbrella Review, h, hour; hrs, hours; min, minutes; nr, not reported; N, number; wk, week.

Symbols:+positive effect (green); o null effect (red); - harmful effect (red); / indicates mixed effects (amber); * sample size does not include the participants included in SRs within URs; ** effects in most studies were positive, but some studies showed no effect.

TABLE 3 Overall effects of taking up opportunities for participation on participation and other outcomes, including AMSTAR quality rating of the SRs and quality of the original studies.

Intervention • No. of studies • A* for each SR reporting studies	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social skills**	**Other outcomes**
Participation in existing groups, activities or programs in the community					
13. Community group participation • 4 studies* • A*: L L M M	ID or PSD, $N = 58$, mean age 56–59 years, 72%–100% male	1-3 times per week for an average of 3.6 h per week	+ [L]		
14. Music • 1 study (ASD), 30 studies (PD) • A*: M (ASD), M H (PSD)	ASD, $N = 22$, age, and sex nr	nr	+ [nr]		
	PSD, $N = 1819$, 15–60 years, 56.5% male	1 to 6 sessions for a total of 45 min to 2 h p/wk	+** [L-M]		+** [L-M]
15. Dance • 6 studies • A*: L L M M M	ASD, $N = 233$ (ASD) including 151 with ID and 3 with PSD, 14–53 years, 72% male	weekly for 1 to 1.5-hrs p/wk for 7-10 wks		o/+ [M-H]	o/+ [M-H]
16. Drama • 13 studies • A*: M M	ID or PSD, $N = 31$ (ID), $N = 171$ (PSD), age and sex nr, but 4 programs for men only	Most 10–11 sessions or over 4–6 months (session length & frequency nr).	+ [nr]	+ [nr]	+ [nr]
17. Art • 1 study (ID), 8 studies* (PSD) • A*: M (ID), L L M (PSD)	ID, $N = 5$, aged 21–27 years	Two days p/wk	+ [L]		+ [L]
	PSD, $N = 60$, age, sex, and quality nr	2-hr p/wk to unlimited access to an open studio	+ [nr]		+ [nr]
18. Farm, ecotherapy, gardening and horticulture interventions • 14 studies • A*: L L L L H H	PSD, $N = 405$, aged 20s to 70s, 22% male	1 to 3 sessions of 1.5–3 h p/wk 2 × to 3-hr sessions p/wk for 12 wks	+ [M]		+** [M]
19. Outdoor nature experiences and camps • 7 studies • A*: M H H	PSD, $N = 211$, 18–65 years, 20%–39% male	1-5 weekly 1-3 h sessions	+ [M-H]		+ [M-H]
Sport or physical activity interventions					
20. Motivations to participate in sport or physical activity • 37 studies • A*: M M M	PSD, $N = 6{,}466$ psychosocial disability, N = 80 clinicians, aged 19-67 years, 71% male	1–2 weekly sessions of 45 min to 2-hrs	o/+ [M]		o/+ [L-H]
21. Sport or physical activity programs • 11 studies • A*: M	PSD, $N = 552$, mean age 25-45 years, sex nr	2-3 × 1–2 h p/wk for 8-24 wks	o/+ [M]		o/+ [L-H]
22. Mainstream sport/ physical activity in community • 5 studies • A*: L L M M	ID, $N = 356$, aged 11–83 years, sex nr	Nr	o/+ [L-H]		+** [L-H]
23. Unified Special Olympics participation • 6 studies • A*: L M M	ID, N not clear, average age 25–31 years, sex nr	Nr	+ [L]		+ [L-M]
24. Disability-specific physical activity programs • 12 studies • A*: L L L M M	ID or ASD, $N = 448$ (ID) & $N = 89$ (ASD), 13–77 years, 56.3% male	2-3 × wk for total of 1.5-hrs to 3-hrs p/day for 8 wks to 10 months	+** [L-H]		+** [L-H]

(*continued*)

TABLE 3 Continued

Intervention • ***No. of studies*** • ***A* for each SR reporting studies***	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social skills**	**Other outcomes**
25. Disability-specific Special Olympics participation • 7 studies • A*: M M M	ID, $N = 181$ intellectual disability, N = 101 parents, siblings coaches or caregivers; aged 12–50 years, 52% male	3 × 1.5 h p/wk	+ [L]		+ [L]
26. Special Olympics participation (setting nr) • 10 studies • A*: L M	ID, $N = 1{,}247$ intellectual disability, $N = 746$ other people, aged 9--69 years, 50% male	Nr	+ [L]		+ [L]

Notes: Blank cells indicate no evidence available. Detailed summaries of the study and sample characteristics and outcomes are provided in Supplementary Table 4, the numbered intervention categories in column one corresponds to the numbered paragraphs below and the numbered rows in Supplementary Table 4.
Abbreviations: L, low quality; M, moderate quality; H, high quality; A*, AMSTAR (A MeaSurement Tool to Assess systematic Review); AAT, Animal Assisted Therapy; ASD, autism spectrum disorders; ID, Intellectual Disability; PSD, Psychosocial Disability; SR, systematic review; UR, Umbrella Review, h, hour; hrs, hours; min, minutes; nr, not reported; N, number; wk, week.
Symbols:+positive effect (green) o null effect (red) - harmful effect (red) / indicates mixed effects (amber); * sample size does not include the participants included in SRs within URs; ** effects in most studies were positive, but some studies showed no effect; *** study examined acceptability and experiences only, not effects on participation or skills.

increased convivial encounters and confidence to engage socially. One passive animal companionship study found improvements in social-adaptive functioning, and two others reported reduced depression symptoms or improved self-esteem, self-determination, and psychiatric symptoms. Animal companionship did not improve social support or loneliness. It was not clear if benefits were specific to animal companionship or participation in structured activities (31).

Two types of intervention focused on *enhancing community linkage* for people with psychosocial disability or intellectual disability by linking the person with community-based recreation or interest-based activities (Category 5, study quality nr). For these *"connecting people"* or *"social prescriber"* interventions people at risk of chronic health conditions, including psychosocial disabilities, were referred to a "navigator" who assessed their social participant and/or mental health needs, interests, and preferences and helped them connect with community programs or activities (Category 6, low-high study quality). Community linkage and social prescribing interventions led to increased social network size, including non-paid contacts, social connections and friendships, and reduced loneliness. Linkage supports improved community involvement and participation but had little effect on social activity and interactions in community settings. These interventions improved interpersonal skills, self-esteem, confidence and feeling worthwhile, but had inconsistent effects on mental health and general wellbeing. Building trust was vital in establishing relationships (32). Few evaluations compared linkage support interventions with control conditions, and low uptake in several studies suggests low acceptability.

Befriending interventions matched a person with psychosocial (study quality not reported) or intellectual disability (moderate-high study quality) with a volunteer befriender from the community (Category 7). Most befriending matches were based on shared characteristics and interests for people with psychosocial disability, and some also included stipends to support activity participation costs. For people with psychosocial disability befriending increased perceptions of social support, but did not affect loneliness, social functioning, social networks, general wellbeing, or psychiatric symptoms. A monthly stipend to the group receiving befriending support and a control group that received a stipend without befriending had similar increases in social functioning and network size. There was limited engagement for some participants, however, given that 23% (33) to 36% (34) of people with psychosocial disability never met their befriender. People with intellectual disability experienced few benefits to community participation and social network size, had little choice about the nature and frequency of interactions, and some reported negative effects on existing social networks when befriending activities interrupted regular schedules.

Other *friendship interventions* matched people with psychosocial disability to a peer with psychosocial disability (Category 8; e.g., the "Buddy Care" intervention), or focused on re-establishing connections with existing friends. Peer-based befriending increased social contacts and perceived social support, and improved overall mental health but had no effect on loneliness or social network size, psychiatric symptoms or service use. Friend-oriented psychoeducation successfully re-established social networks and increased social contacts.

TABLE 4 Overall effects of interventions to build skills, psychosocial wellbeing, and broader capacity to participate socially and in the community, including AMSTAR quality rating of the SRs and quality of the original studies.

Intervention • ***No. of studies*** • ***A* for each SR reporting studies***	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social Skills**	**Other outcomes**
Social skill and communication interventions					
27. Social Skills Training that did not include a "cognitive" focus • 14* studies • A*: L L L M M M	PSD, $N = 549$, 17–51 years, 76% male	45 min to 1.75 h/wk for up to 18 months	+** [L]	+ [L]	o/+ [L]
28. Social Cognition training focused on loneliness & self-control • 3 studies • A*: L M	PSD, $N = 269$, mean age 20–50 years, 64% male	30–60 min/wk for 9-12 wks	+ [M]	o/+ [M]	
29. Individual or group Social Cognition and Interaction Training (SCIT) • 13 studies • A*: L M M M	PSD, $N = 719$, M = 33–51 years, 68% male	1–2 × 1.5 h p/wk for 16–24 wks	+ [L-M]	o/+ [M]	
30. Group social skills training • 4 studies (ID), 7 studies (ASD) • A*: L M M (ID), L L L M M (ASD)	ID, $N = 71$, 17–48 years, 38% male	2 h/wk for 12–14 wk s	+ [M]	+ [M]	
	ASD, $N = 78$, 16–55 years, 85% male	30 min to 3-hrs sessions over 4–6 wks or up to 18-wks	o [nr]	o/+ [L]	
31. PEERS-YA social skills training program • 4 studies • A*: L L M M	ASD, $N = 97$, 20–24 years, 80% male	1.5 h sessions over 14–16 wks	o/+ [L]	o/+ [L]	
32. Individual social skills training • 7 studies • A*: L L M M	ASD, $N = 31$, aged 17–20 years, 77% male	40 min to 1-hr for up to 33 wks	+ [L]	+ [L-H]	o/+ [L]
33. Intensive Interaction Support for specific communication skills • 8 studies (ID), 6 studies (ASD) • A*: L M (ID), L L M M M (ASD)	ID, $N = 27$, 28–53 years, 59% male	Frequent, usually daily, short-interval training	+ [L-H]	+ [L-H]	
	ASD, $N = 57$, 17–32 years, sex nr	10-50 min 1–2 times each week for 4-9 wks	+ [H]	+ [L]	
34. Theory of mind/ emotion/ social cognition training • 5 studies (ASD) 11 studies (PSD) • A*: L M (ASD), M (PSD)	ASD, $N = 146$, age and sex nr	30-min to 2 h/wk for 5–10 wks	o [L]		
	PSD, $N = 495$, mean age 25–44 years, 64% male	12 to 20 × 1-hr sessions	+ [M]	+ [M-H]	
Psychosocial wellbeing and capacity building support					
35. Telehealth-based supports or SMS prompting • 4 studies • A*: M M M	PSD, $N = 178$, 61–92 years, 15% male (nor for 2 studies)	12 weeks to 9 months (frequency nr)	+** [nr]		o/+ [nr]
36. Psychoeducation • 9 studies • A*: L L M M M	PSD (1 study PSD + ASD), $N = 912$, mean 32–38 years 85% male	1-2 sessions per week for 8 weeks to 2 years, from 30-40mins to 1.5-3 h p/week	o/+ [L-H]		o [L-H]
37. Mindfulness for social anxiety • 2 studies • A*: L	ASD, $N = 91$, age, and sex nr	2.5 h/wk for 9 wks	+ [L-M]		+ [L-M]

(*continued*)

TABLE 4 Continued

Intervention • ***No. of studies*** • ***A* for each SR reporting studies***	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social Skills**	**Other outcomes**
38. Cognitive Behavioural Therapy (CBT) based interventions targeting social functioning • 5 studies (ASD) 2 studies (PSD) • A*: L M M M	PSD, *N* = 87, 14–45 years, 100% female ASD, *N* = 147, age and sex nr	Weekly or monthly 1-4hrs sessions for 6 to 24 weeks	o/+ [L-M] + [M]		+ [M]
39. Cognitive reframing • 4 studies • A*: L M M	PSD, *N* = 204, age and sex nr	1–2 or 14–22 sessions for 1–2 h each	+** [L-M]	+ [M]	+ [L-M]
40. Meta-cognitive training, Cognitive Enhancement Therapy • 4 studies (PSD) 2 studies (ASD) • A*: L M M	PSD, *N* = 143, mean 26–40 years, 66% male ASD, *N* = 68 ASD, mean 25 years, 86% male	36–45 sessions 18 months (session frequency/ length nr)		+ o/+ [L-M] + [L-M]	+ [M] + [M]
41. Behaviour activation • 2 studies • A*: M	PSD, *N* = 113, age and sex nr	Up to 12 sessions	o [M]		o/+ [M]
42. Recovery-oriented clinical therapy • 2 studies • A*: L M	PSD, *N* = 56, mean 37–43 years, 45% male	20–45 min 1–2 times per week for up to 21 sessions	+ [L/nr]		
Vocational social skills interventions of supports					
43. Vocational internships and training and volunteering • 2 studies, 1 SR from an UR • A*: L L M	PSD, *N* = 112, M = 28–31 years, 64% male	nr	-/o/+ [M]		+ [M]
44. Vocational social skills or coaching programs • 3 studies • A*: L	ID, *N* = 15, 18–26 years, 60% male	3 h/wk for 12 weeks or during unpaid internships for 4–8 h/wk	+ [H]	+ [H]	+ [H]
45. Aspirations Program – vocational skills • 3 studies • A*: L L L M M	ASD, *N* = 71, 19–22 years, 86% male	4 to 20-hrs p/wk until independent in the workplace	o/+ [L]	+ [L]	+ [L]
46. Job interview and conversation skills training • 3 studies • A*: L L M	ASD, *N* = 150, M = 24–25 years, 86% male	1.5 to 2-hrs p/wk for up to 12 weeks		o/+ [L-M]	o [L]
47. Broad vocational social skills programs • 7 studies • A*: L L L M M	ASD, *N* = 153, 18–27 years, 85% male	∼15-min sessions to learn specific skills, 6-26 × 1–1.5-hrs sessions, or daily support for up to 6m		+ [L-M]	+ [L]
Relationship-focused interventions					
48. Dating, sex, and relationship programs, mostly in group settings • 7 studies • A*: L L L M M M M M	ASD, *N* = 201, 18–60 years, 62% male	1–2 or 1.5–2 h × 10–20 sessions	+ [L-M]	+ [L-M]	
49. Sex, relationship & family planning (group or individual programs) • 15 studies • A*: L M M	ASD or ID, *N* = 722, 12-59 years, sex nr	25–30 min to 2.5–3 h sessions for a total of 6–30 group sessions or 13–30 individual sessions	+ [L]	+** [L]	o [L]

(*continued*)

TABLE 4 Continued

Intervention • ***No. of studies*** • ***A* for each SR reporting studies***	**Disability cohort, pooled sample size, age range, sex**	**Typical intervention duration and/or frequency**	**Effects on outcomes [study quality]**		
			Social functioning	**Social Skills**	**Other outcomes**
50. Abuse prevention programs • 8 studies • A*: L M	ASD or ID, *N* = 175, 11-57 years, 50% male	40–60 min for 2–5 or 18–40 sessions		+** [L]	
51. SexG program on sexual health and responsibility • 4 studies • A*: H	PSD, *N* = 595, M = 37-40 years, 100% male	brief (6 × 1-hr session) and enhanced (13–15 × 1-hr sessions)		o/+ [M-H]	
52. Relationship and AIDS/HIV-prevention interventions • 9 studies • A*: H	PSD, *N* = 1,268, aged 22–59 years, 47% male	1.5–2 h × 4–10 sessions		+** [L-M]	
Life skill focused interventions					
53. Life Skills Training • 2* studies • A*: L M	PSD, *N* = 272, M = 48–52 years, 65% male	two × 2-hrs p/wk for 12 weeks		+ [L]	+ [L-M]
54. Parenting skills, knowledge, and safety training • 6 studies • A*: L H	ID, *N* = 155, aged 16-49 years	1–2-hrs × 4–5 or 10-26 sessions with home-visits in one study	+** [M-H]		
55. Digital literacy skills training • 4 studies • A*: L M	ID, *N* = 67, 18-23 years, sex nr	Duration/ frequency nr	o/+ [L-H]	+ [H]	
56. Navigation and travel training interventions • 11 studies • A*: L	ASD or ID, *N* = 171, M = 19-32 years, sex nr	Frequent short sessions for a total of 30–60 min	o/+ [nr]		
57. Life Story work • 2 studies • A*: L	ID, *N* = 71, aged 55–63 years, sex nr	2 individual 1hr sessions or 16 individual + group 1.5–2 h sessions	+ [M]		o/+ [M]

Notes: Blank cells indicate no evidence available. Detailed summaries of the study and sample characteristics and outcomes are provided in Supplementary Table 4, the numbered intervention categories in column one corresponds to the numbered paragraphs in the main text, and the numbered rows in Supplementary Table 4.
Abbreviations: L, low quality; M, moderate quality; H, high quality; A*, AMSTAR (A MeaSurement Tool to Assess systematic Review); ASD, autism spectrum disorder; ID, Intellectual Disability; PSD, Psychosocial Disability; PEERS-YA, Program for the Education and Enrichment of Relational Skills for Young Adults; SR, systematic review; UR, Umbrella Review; h, hour; hrs, hours; min, minutes; nr, not reported; N, number; wk, week.
Symbols:+positive effect (green); o null effect (red); - harmful effect (red); / indicates mixed effects (amber); * sample size does not include the participants included in SRs within URs; ** effects in most studies were positive, but some studies showed no effect.

Peer support groups in the community facilitated access to peers through the internet, mobile applications or face-to-face settings. Some peer support programs included a mental health professional facilitator alongside routine case management (Category 9). Community-based peer support groups were reported to be a welcoming community where people with disability could be themselves, share coping strategies, fill their free time, and interact with others. People with psychosocial disability experienced improvements in social belonging, connectedness, wellbeing, empowerment, hope and self-efficacy. Some studies reported limited impacts on social relationships with peers, no change in isolation, loneliness or connections with friends or family, and only short-term improvements in satisfaction with getting along with others that was not maintained to 6-months post-intervention. Peer support had inconsistent effects on quality of life and psychological wellbeing. While positive effects were observed for people who attended regularly in some studies, an internet-based peer support program found higher participation was associated with higher levels of distress (35). In that study, adverse effects on distress were attributed to potential overwhelm from the volume of interactions or "absorbing" distress from others *via* online discussion pages. Alternatively, people with higher distress may have engaged more with the peer group. Effectiveness was driven by having opportunities to participate in activities in the community (36). While only four of 10 studies had a quality appraisal, all were moderate to high quality, and seven studies compared peer support with control conditions.

Thirty-six studies assessed *peer support programs integrated into statutory mental health services* where peer mentors worked alongside clinicians to support people with psychosocial disability (Category 10, low-high study quality). A meta-ethnography by Walker and Bryant (37) examined mentor, mentee, staff, and service provider experiences, and other studies examined social and health effects of statutory peer supports. For service users with psychosocial disability, peer support reduced feelings of alienation, improved community reintegration, recovery, wellbeing, hope, motivation, friendships and social networks, and illness management skills. Peer support had no impact on social network support, social functioning, psychiatric symptoms, or quality of life. For peer workers with a psychosocial disability, providing peer support improved their own recovery, increased social networks, and led to other opportunities. Non-peer staff reported that peer workers could help service users belong in the community beyond being a "patient".

Transition programs for young adults with intellectual disability or on the autism spectrum predominantly focused on adjustment to post-secondary education or learning social and academic skills, and goal setting (Category 11, low-high study quality). Transition support improved social participation with friends and other people both with and without disabilities. Transition programs also improved participation in leisure activities, and learning self-advocacy skills.

Transition programs for older adults with intellectual disability supported *transitions into retirement* through active participation in community groups aligned with the older adult's interests (Category 12, moderate study quality). Transition to retirement supports improved intimate relationships and awareness of rights, but had inconsistent effects on interpersonal relationships, social inclusion, self-determination, and emotional wellbeing. Effects were limited if programs did not support maintenance of existing networks or building new networks, or if people had insufficient resources to continue to participate after the research project ended (38).

3.3.2. Interventions offering participation opportunities

These interventions included direct opportunities to participate socially or in the community (**Table 3**). Most activities were in disability specific settings, except for community groups (e.g., men's shed; category 13), some gardening interventions (category 19), and some sport-based interventions (categories 20–23). Effectiveness did not appear to differ between mainstream and disability-specific settings.

Three studies supported people to join *existing community groups* (e.g., Men's Sheds) that matched the participant's interests, sometimes training existing group members to support the participation of the person with a disability (Category 13, low study quality). Community group participation improved social satisfaction, social network size and time spent with new social contacts, but did not change loneliness, depression, physical health, or quality of life, possibly because the new relationships typically did not extend beyond the group setting (39, 40). Men's sheds offered the opportunity for meaningful participation and establishment of camaraderie to build a support network (41). Effects were enhanced if groups enabled genuine involvement in activities and social interactions with other group members through active mentoring (39).

Music programs (Category 14, low-moderate study quality) included the *Soundscape* program for people on the autism spectrum, which enhanced peer relations and self-esteem. Music activities for people with psychosocial disability included group singing in the community and music therapy focused on receptive (e.g., music appreciation and discussion) and/or active processes (e.g., music production or improvisation, singing, playing instruments). Group-based singing improved social functioning, belonging and connection to community. Attrition from choirs was influenced by changes in employment, worsening mental health, family problems, accommodation issues, and anxiety about singing ability (42). Music programs increased short, medium, and long-term social functioning, and had inconsistent effects on perceived social support, with superior effects from programs comprising group processes compared with education-focused programs. Music programs reduced anxiety, but had inconsistent effects on depression, cognitive functioning, psychiatric symptoms, and quality of life. Music therapy was particularly effective at improving negative symptoms such as affective flattening and blunting (i.e., a lack of emotional reactions), social relationships, and motivation (26). However, most of these interventions were provided in clinical inpatient or outpatient settings, and their acceptability and effectiveness in community-based settings was not clear.

Dance programs for people on the autism spectrum with or without an intellectual disability, and for people with psychosocial disability, focused on social skills (e.g., perspective taking or mirroring others; Category 15, moderate-high study quality). Dance programs improved interaction, imitation, emotion expression and regulation but did not affect social skills, self-other awareness, empathy, cognition, communication, or psychological wellbeing. People with psychosocial disability in dance programs felt valued by others and reported feeling empathy from others.

Drama activities for people with intellectual disability and/or psychosocial disability were predominantly provided in therapeutic programs that fostered storytelling, self-awareness, and building positive relationships with others. Two programs developed a performance presented to mainstream audiences (Category 16, high study quality or nr), which increased social acceptance and relationships with other participants and community members. The social aspects of group drama programs improved personal organisational and social development and established group harmony. Drama group participation increased social inclusion, acceptance, relationships, and friendships with other participants and community members; increased engagement with others and leisure activity participation; and reduced isolation. Drama group participants reported motivation to continue meeting other participants in a peer support group after completing the program. Drama participation also led to improved communication and social skills, self-awareness, awareness of others, and impulse control; and reduced challenging behaviours. Participants reported increased creativity, empowerment, confidence, self-worth, self-esteem, resilience, quality of life, mood, and recovery, and reduced perception of discrimination and self-stigma. As no evaluations compared drama with a control condition the mechanisms of benefit could have simply been related to the group setting or social interactions and not drama *per se* (43, 44).

Qualitative evaluations suggested that drama groups affected social functioning in two ways. First, group settings and activities enhanced *support and trust* (43), including fostering the ability to share and collaborate with others (45) and building relationships (46). Participants enjoyed observing others' resilience and resourcefulness in a crisis (47). Participating with others with similar experiences was helpful (47, 48) and resulted in a sense of safety to explore socially inappropriate behaviours (49), although drama groups led to increased sense of vulnerability for some (50). Second, through drama people *learned more about themselves and built their self-concept, confidence and empowerment* (45, 46, 50, 51). Participants explored their individual experiences to learn more about themselves (47), which improved their self-awareness and sense of control (52).

Arts-based activities (Category 17, low study quality) for people with intellectual or psychosocial disabilities focused on developing artistic skills and creating art. Some programs included community exhibitions to display and sell artwork. Participants typically created art alongside artists without disabilities or received instruction and guidance from an instructor. Programs for people with psychosocial disability included art studio programs in community centres or psychiatric rehabilitation settings for people with a mental health problem or as "arts on prescription". Art studio participation enabled creation of a "community of artists" that fostered links with the broader community, including convivial encounters with community members (53). Participation increased social inclusion, sense of belonging, engagement, mutual support, social connections, friendships, meaning in life, self-esteem, happiness, and confidence. Participants enjoyed receiving praise for their work from community members. Selling artworks gave artists a presence and voice in the community, fostered a sense of achievement, and positively contributed to identity-related outcomes. Art participation led to broader positive life outcomes (e.g., employment, housing, recovery, quality of life, wellbeing), and reduced distress and psychiatric symptoms. Studio facilitators who worked alongside participants fostered a sense of equality, inclusion, belonging and intersubjectivity (54). People enjoyed being able to offer and share with others (54). However, arts interventions were not compared with a control condition, and the social inclusion benefits may be due to the broader collaborative and creative group settings rather than art-based processes (55).

Farm, ecotherapy, gardening and horticultural interventions and groups for people with psychosocial disabilities (Category 18, low-high study quality) included short-term interventions, vocational training programs or long-standing community "allotment" programs. Farm-based programs involved working with farm animals including feeding and grooming animals, milking cows, and riding horses. Interventions led to reduced loneliness, increased social participation, new friendships, and improved self-efficacy, coping, mood and general mental health. The evidence was weak and limited as no studies compared the interventions with a control condition and studies were predominantly low-moderate quality.

Outdoor recreation and leisure programs for people with psychosocial disability included structured programs (e.g., information sessions, personal development workshops, self-help groups, community walks and forums), and nature-based programs (e.g., camps or dolphin therapy; Category 19, high study quality). Interventions led to improved social connectedness, relationships, interpersonal relationships, personal growth, confidence, wellbeing, self-determination, and empowerment, and reduced loneliness and depression.

Sports and physical activity participation opportunities were evaluated primarily for people with psychosocial disability and intellectual disability. Three meta-ethnographic reviews examined motivations and barriers to physical activity participation for people with schizophrenia (56), participant experiences when starting community-based group physical activity (57), and physical activity participation experiences

(58) (Category 20, moderate-high study quality). Other studies evaluated social outcomes after *physical activity programs* or *sport participation* for people with psychosocial disability such as soccer training and games, learning and practicing yoga, outdoor or nature-based recreation (e.g., white-water rafting) and fitness programs (e.g., aerobic, interval, resistance, and strength training; Category 21, low-high study quality).

Only 27% of people endorsed that the social aspect of exercise was a motivator (socio-ecological motivators) (56). Physical activity participation led to several outcomes, including:

1. **Psychosocial**: Improved socialization, social/emotional support, empathy, sense of warmth, companionship, sense of control, sense of achievement, self-appreciation, confidence, self-esteem to engage in the community, and autonomy. Yoga led to improved psychosocial functioning, and nature-based therapeutic recreation and soccer and football participation had positive impacts on relationships, social inclusion, and isolation; however, interval training had no effect on psychosocial functioning.
2. **Mental health and recovery**: Fewer hallucinations, psychiatric symptoms and improved overall functioning, but only for participants who attended ≥50% of exercise sessions (59–61); improved mood, relaxation, and mental health, but only for studies with aerobic with resistance training methods with ≥90 min per week of moderate-vigorous exercise (62, 63). Programs that enhanced cohesion and relatedness between participants led to reduced anxiety.
3. **General health**: Improved fitness, sleep, and quality of life. Weight loss was motivating and considered "a yardstick for recovery" (58).

Mainstream sports programs or physical activity in the community for people with intellectual disability included team sports, active recreation, or walking with a person without intellectual disability (Category 22; low-high study quality). *Unified Special Olympics* (SO) programs included athletes with intellectual disability and age and ability matched people without intellectual disability who played in the same teams (Category 23, low-moderate study quality). Mainstream sport participation led to increased opportunities for convivial encounters but had inconsistent effects on interpersonal relations. Unified SO team participation led to improved friendships, social inclusion, access to community venues and sense of community belonging, and alliances within local communities. Programs provided "a platform for the development of social relationships", and participants reported that they felt like they were "a part of society" (64). Wilhite and Kleiber (65) found more improvement in community involvement for people with moderate-severe intellectual disability, perhaps because people with mild intellectual disability already had relatively good community involvement. Participants enjoyed learning and playing sports and receiving praise or acknowledgement from others, and reported improved emotional wellbeing and physical activity levels. Participation in unified SO led to better social self-perception and acceptance and reduced maladaptive behaviours.

Disability-specific exercise, physical activity, and leisure programs for people on the autism spectrum or with intellectual disability primarily focused on strength, balance, fitness, and health (Category 24, low-moderate study quality). A leisure program for people on the autism spectrum used a PCP approach and focused on fostering social collaboration and support, and provided 2-hours of leisure activities in the community five days each week. The intervention led to improved interpersonal relationships, social support, belonging, life satisfaction, quality of life, self-efficacy, psychosocial wellbeing, quality of life, empowerment, and employment. There were inconsistent effects on community integration and adaptive behaviour, and no effect on social integration, leisure needs, engagement, or satisfaction. A lack of transport and psychosocial support limited continued participation (66).

Seventeen studies evaluated *traditional SO* training and participation for people with intellectual disability (Category 25, low study quality), or did not report whether the SO setting was unified or disability-specific (Category 26, low study quality). Traditional SO participation was associated with improved social self-perception, meeting people, making friends, community awareness, inclusion and involvement, independence in the community, social behaviour; and reduced challenging behaviour. Participation was associated with improved social skills, exercising choice, receiving social approval and acceptance; having fun, happiness and enjoyment; and physical health and sport skills. SO participation was described as playing an important role in the lives of individuals with intellectual disability, their families and the community (67).

3.3.3. Interventions to develop skills or psychosocial capacity to participate

Capacity-focused interventions aimed to improve social, communication and relationship skills, psychosocial wellbeing and life skills, and navigation of digital information or the community (**Table 4**). Most interventions were in disability-specific settings except for vocational social skills interventions in the workplace (category 43–47). Interventions targeting psychosocial wellbeing were included only if they addressed social participation, linkage, capacity, or outcomes.

3.3.3.1. Group-based social and communication skills training

For people with psychosocial disabilities, studies evaluating group-based social skills interventions were low-moderate quality and included:

- *Social Skills Training* (SST; Category 27) of interpersonal skills, social problem solving, social perception, theory of mind, social information processing, interaction skills, understanding social norms, and applying skills to everyday situations;
- *Social Cognitive Training* (SCT; Category 28) to reframe loneliness perceptions, and build self-control, coping strategies, sense of belonging and stress management; and
- *Social Cognition and Interaction Training* (SCIT; Category 29) of social cognitive dysfunction, sometimes using augmented reality simulation and cognitive remediation approaches.

Most social skill interventions were delivered alongside other clinical supports (e.g., case management, illness management, family-oriented psychoeducation). SST and SCIT improved social functioning, role functioning, social relations, and social activity participation, and reduced social isolation. SCT improved affect recognition, but only reduced loneliness after more intensive programs. SST improved behavioural skills, social skills, theory of mind, goal attainment and recovery, had small effects on non-verbal social skills, and no effects on perceived social support. SCIT had inconsistent effects on social capacity, theory of mind, affect recognition and attribution style and no effect on interpersonal communication. Interventions that provided frequent contact with a therapist (68), used a range of methods to enable transfer of learned skills into everyday life (69), and provided elements of training in community settings (70) were most effective at helping people to apply learned social skills.

Group-based social skills training for people with intellectual disability targeted social awareness and competencies including interpersonal communication and listening skills (Category 30; e.g., a TEACCH-based program; moderate-high quality studies; SCIT program, “Putting feet on my dreams” and “Problem Solving Skills 101”; low study quality). Training reduced social withdrawal and improved relationships with partners and friends; increased confidence and knowledge to participate in the community and joining or establishing support/social groups; and improved self-concept and quality of life in people whose understanding of civil rights and engagement also improved after group training.

Group-based programs for people on the autism spectrum used instruction, discussion, and rehearsal of social and communication skills with video feedback, including the *PEERS-YA* program (Category 31; low study quality). Interventions increased invitations to social get-togethers, but had inconsistent effects on social responsiveness, socialisation, social skills and behaviours; conversation skills (e.g., initiating and maintaining conversation, reducing inappropriate utterances, attention, and feedback to questions), and emotion identification. There were similar improvements in social functioning and theory of mind in the intervention and control groups who also participated in a social interaction group without training, and there were no effects on hosting get-togethers, loneliness, broad social communication skills, social performance, empathy, and social body language (e.g., eye contact, gestures).

3.3.3.2. Individual social skills training

Individual multifaceted social skills interventions for people on the autism spectrum or with intellectual disability provided psychoeducation, coaching and training to use tools like a digital planner to schedule activities (Category 32, low-high study quality). Interventions increased social event attendance, peer interaction satisfaction; social skills (e.g., initiation and maintenance of interactions, social skill performance, and timely responses to questions), and employment and quality of life.

Fourteen studies evaluated individualised interaction support training of *specific communication impairments* for people with intellectual disability or on the autism spectrum (Category 33, low-high study quality). Interventions improved social behaviour and the targeted social skills, while also reducing challenging behaviours. There were limited effects for people with severe and chronic challenging behaviours following short-term interventions (71), and gains were not consistently maintained post-intervention for people with more severe intellectual disabilities. One RCT compared a Virtual Reality (VR)-integrated computerised training program with an active control group who also received computerised training and found no differences in improvement between groups. Barriers to implementation included inconsistent capacity or maintenance of individualised interaction support by support workers over time (72–75).

Sixteen studies evaluated interventions targeting *specific social competencies* like theory of mind, emotion perception, and social perception (Category 34) for people on the autism spectrum (low study quality) or with psychosocial disability (moderate-high study quality). Interventions did not affect social functioning for people on the autism spectrum, but intensive interventions improved social and occupational functioning, social perception, theory of mind and affect recognition for people with psychosocial disability.

3.3.3.3. Psychosocial wellbeing interventions to enhance participation capacity

ehealth interventions provided people with schizophrenia, schizoaffective disorder, or depression, and people on the autism spectrum support to manage symptoms and enhance socialization through telephone or SMS-based prompting (Category 35, study quality nr). Interventions increased social interactions and leisure activity participation but did not change loneliness.

Psychoeducation for people with psychosocial disability, including people on the autism spectrum who also had a psychosocial disability, to learn problem solving and coping skills, illness management and encouraged social participation through computer or web-based programs or in-person programs (Category 36, low-high study quality). Some interventions also included family therapy. Interventions improved social functioning, social contacts, and loneliness, but effects were not consistently maintained. There were inconsistent effects on quality of life and no effects on psychological wellbeing, depression, or perceived social support.

Group-based *mindfulness* programs for people on the autism spectrum focused on awareness and management of social anxiety (Category 37) and led to reduced anxiety, depression, rumination, agoraphobia, and somatisation, and improved positive affect.

Individual or group-based *Cognitive Behavioural Therapy* interventions focused on behaviour activation, social interactions, and social anxiety for people with psychosocial disability or on the autism spectrum (Category 38, low-moderate study quality). *Cognitive reframing and remediation* interventions for people with psychosocial disability targeted cognitive strategies to analyse social situations and increase social interactions (Category 39, low-moderate study quality). Interventions did not influence loneliness after brief interventions (e.g., two 30-minute cognitive reframing sessions), but did reduce loneliness after for a more intensive intervention (e.g., five 4 h sessions). Interventions improved social cognitive processes, attribution style, empathy, theory of mind; schizophrenia, depression and anxiety symptoms, and daily functioning; and personal and social performance. One intervention led to reduced perceptions of social support in ex-military officers with PTSD (76), which may have been a spurious finding given that participants also reported improved reactivity to criticism of family members; however, these poorer outcomes suggest that interpersonal skills interventions may require more supported practice than what the brief intervention offered.

Cognitive Enhancement Therapy and *meta-cognitive training* for people with schizophrenia or on the autism spectrum were delivered in individual and group sessions targeting impairments in social and non-social information processing, cognitions and problem solving (Category 40, low-moderate study quality). These interventions improved global social functioning and perception, cognitive style and social cognition, and reduced disability, but had inconsistent effects on theory of mind and affect recognition.

Behaviour activation interventions taught people with depression to assess, prioritise and practice their values and goals (Category 41, moderate study quality), and led to decreased depression symptoms but did not change perceived support.

Integrated Psychological Therapy for Schizophrenia and *Interpersonal Community Psychiatric Treatment* are clinical therapies that focus on recovery and enhancing community participation (Category 42, study quality low or not reported). Treatment led to improved social perception knowledge, social networks, and social activity.

3.3.3.4. Vocational social skills training

For people with psychosocial disability, vocational interventions focused on creating occupational opportunities for people with psychosocial disability to work in mental health services (e.g., the *Empowerment of Mental Illness service users: lifelong Learning, Integration and Empowerment* project; Category 43, moderate study quality), or to do volunteer work (Category 43). The internship intervention improved social life, social contacts, and networks for most people, but maintaining relationships was difficult. Volunteer work increased social inclusion, social ties, and social engagement opportunities, but also put people at risk of stigmatising experiences in the community (77).

For people with intellectual disability vocational interventions targeted social skills at work (Category 44; e.g., Walker Social Skills Curriculum, covert job coaching or video-based instruction; high study quality). Interventions increased social interactions over time; improved social competence, interpersonal skills, social skill mastery and social participation; improved employment rates, job security, and ability to perform work roles; and reduced challenging behaviours.

Programs for people on the autism spectrum focused on *social and vocational skills education*, and support to find and maintain employment (the Aspirations Program; Category 45, low study quality), *job interview conversation skills* for people on the autism spectrum (Category 46; e.g., The Molly Porter Job Interview VR training program, or Social Skills Curriculum for job interview-related skills, low-moderate study quality), or *training of social skills* for vocational settings (Category 47; e.g., social skills required for a work role, such as gestures like waving, while dressed as a mascot; low-moderate study quality). Training improved empathy but did not improve peer relations or socialisation despite anecdotal reports of improvements. Job interview training improved interview communication skills, but did not improve interview performance in one study, and did not affect confidence or adaptive behaviour.

3.3.3.5. Relationship-focused skills, knowledge, and behaviour training

Relationship programs for people on the autism spectrum without intellectual disability (e.g., Ready for Love), or for people with intellectual disability (e.g., Friendships and Dating Program, Early Dating Skills Training, or Dating Skills Program; Category 48, low-moderate study quality) were predominantly group-based programs. Interventions improved social skills, dating skills and knowledge; and increased empathy, social responsiveness (i.e., autism-specific social impairments and skills), social functioning, and endorsement of dating behaviours (e.g., kissing, gay and lesbian relationships, sexual intercourse values and morals, keeping secrets). While social network size increased there were no changes in network composition. Participants wanted training that was relevant to their own relationship and sexuality aspirations including lesbian, gay, bisexual and transgender (LGBT) issues and concerns (78), and that included their partner if they were already in a relationship (79).

Fifteen studies evaluated *sex, relationship and family planning* for people on the autism spectrum and with intellectual disability in group or individual programs (Category 49, low study quality). Interventions had broad curricula, including anatomy, puberty, reproduction, sexually transmitted diseases, sexual intercourse, relationships, dating/romantic skills, safety/consent/abuse, self/other in sexuality and relationships, and private/public appropriate/inappropriate behaviours. Program participation improved "social entertainment"; understanding of friendships, interactions with people of the opposite sex; dating problem solving skills; knowledge of sexuality rights, responsibilities, and vocabulary; and endorsement of dating behaviours. There were inconsistent effects on sexual knowledge, improved social skills, and self-protection skills. All studies were low quality, eight of which did not have a control group, and the control group conditions were not described for four RCTs.

Relationship abuse prevention interventions (Category 50, study quality low or not reported) for people on the autism spectrum or with intellectual disability taught decision-making strategies to resist sexual, physical, and verbal abuse. Training increased knowledge of abuse concepts, empowerment, and recognition of inappropriate touching requests. There were inconsistent but mostly positive effects on decision making ability, and no effects on appropriate touching requests. Some people required booster training to maintain and generalise abuse prevention skills. Younger people and those who found the program more difficult had the biggest improvements in relationship knowledge and behaviour (80).

Sex and relationship programs for people with psychosocial disability focused on increasing safe and responsible sex behaviours and attitudes (e.g., SexG group-based interventions; Category 51, moderate-high study quality) or targeted prevention of AIDS and HIV risks (Category 52, low-moderate study quality). In the SexG interventions with men, discussion and role play of safe sex, responsibility, and knowledge, confidence, and motivation to use condoms had inconsistent (but mostly positive) effects on risky sexual behaviours. Interventions targeting knowledge and behaviour to prevent HIV and AIDS increased sexual assertiveness, knowledge and confidence to deal with high-risk situations, and contraceptive use, and reduced risky sex acts, the number of casual sex partners, total number of sex partners and unprotected sex.

3.3.3.6. Life skill interventions

Life skills training interventions included broad programs on medication management, organisation and planning, transportation, and financial management for people with psychosocial disability (Category 53, low-moderate study quality; e.g., Functional Adaptations and Skills Training program). Life skills training improved social skills but did not affect quality of life.

Parenting skill, knowledge, and confidence training programs for people with intellectual disability (Category 54, moderate-high study quality) were delivered individually to improve parenting safety and interpersonal and communication capability and led to improved childcare skills that were maintained over time, and health knowledge (e.g., life threatening emergencies and using medicine). The evidence was moderate to high quality; however, two studies did not include control groups.

Four studies evaluated *digital literacy skills training* for people with intellectual disability to use email or participate in social media (Category 55, most studies high quality). Training improved participants' ability to complete tasks in social media platforms (i.e., Facebook) and email training reduced social isolation. Blogging training did not affect social capital (i.e., the resources that one can access through their social connections).

Independent *travel and navigation skills* training for people on the autism spectrum or with intellectual disability (Category 56, quality not reported) was provided using augmented reality, multimedia, smartphone applications and maps. Augmented reality training led to reduced travel planning time, and improved navigation skills and public transport use. Multimedia and video-based travel skill training improved pedestrian bus route navigation skills that were maintained over time. It was not clear whether skills learned in virtual environments would transfer to natural environments (81, 82), or when a support person is not present (83). Participants benefited more from interventions that meaningfully blended real world experiences with digital information (84).

People with intellectual disability were supported to share their personal history using *Life Story* work (Category 57, moderate study quality) when changing residential locations or joining a new social group. Life Story work improved interpersonal relationships, rights, social inclusion, and self-determination, but had inconsistent effects on emotional and physical wellbeing.

3.4. Intervention implementation considerations

3.4.1. Acceptability, implementation, and maintenance

The literature highlighted that existing staff skills, attitudes and policies can negatively affect implementation (85, 86), and programs in residential or community settings needed to be embraced at all levels of the organisations from frontline support workers to service planners or managers (29). Staff need training (72, 73) and dedicated time and resources to provide (28) and maintain planning support over time (71–74). Moreover, staff or family members sometimes ignore, reinterpret or misinterpret the preferences of people with intellectual disability (87), so focus on the individual and their changing needs and preferences over the lifespan must remain a central focus (88).

To enhance intervention acceptability and maintenance both the intervention facilitators (89) and people with lived experience should contribute to intervention development and delivery (89, 90). A codesign approach can help ensure that the content is relevant to participants' needs or aspirations (78). As participants with disability attending training opportunities may know more (or less) than they seem to, information should be presented in multiple formats using simplified and accessible language (91–94), with information and questions read aloud to improve program acceptability and effectiveness (93). It is important that facilitators gain an understanding of existing relationship skills, knowledge, and interests of people with intellectual disability (86). People with poor digital literacy skills face greater barriers in connecting with others (94). Therefore, programs should provide digital and text-based literacy support and adapt materials for people with different levels and types of impairments (91–93, 95). People with no experience with computers or gaming may find it difficult to use virtual and augmented reality-based interventions (96), and rarely used video prompts (83). Interventions need to address essential life skills (e.g., social skills, literacy, time management, problem-solving, and other cognitive skills) that are needed for participation and establishment of relationships that extend into everyday life (97–99). Moreover, to participate in the community (e.g., in sport or physical activity), people needed to feel "well enough", the activity must be affordable and in an appropriate location for "people like us", and people may only participate if they expect positive outcomes (e.g., access to support, talking with others with similar experiences, seeing/making friends) (57).

3.4.2. Facilitators and barriers

The key facilitators and barriers that generally applied to all three disability cohorts were predominantly related to (a) attributes of the program or intervention; (b) Carer, staff, facilitator, or peer mentor attributes; (c) participant attributes; and (d) community-specific characteristics.

3.4.2.1. Attributes of the program or intervention

Success of participating in group programs often depended on the skills of the facilitator. Programs that effectively engaged participants had facilitators that provided multiple types of support, such as active mentoring to support participation in activities and social interactions (39, 100), and positive leadership and acceptance of people with disability (101). Participants liked programs with structured approaches, rules or policies (102), and homogeneous group characteristics (e.g., similar age) (103) with minimal participant turnover (104, 105). Participants liked having choice about which activities they could participate in, and having regular breaks, rewards, and positive feedback (38, 105–107). People often need support to maintain existing networks or to build new networks (38). Some people with psychosocial disabilities preferred individual over group formats and reported that attending new environments was challenging (58). Adherence was enhanced when facilitators had lived experience of the same disability (108), and being around others with similar disabilities could enhance the sense of community and opportunity to interact with and learn from other adults with similar experiences (47, 48, 98).

Participation was facilitated in programs that use person-centred, strengths-based approaches, and included supports to enable people to have freedom of choice (106, 107), and to set their own goals (90, 104, 106, 107). It was important that needs and priorities were reviewed over time to ensure participation opportunities remained relevant (88). Relationship-focused interventions benefited from tailoring interventions to participant needs (79), and adapting content to each person's circumstances (109). Fostering choice was a facilitator of outcomes; however, several studies noted that choice making for people with intellectual disability was often ignored, misinterpreted (87), or overpowered by caregivers, staff, family (87, 110) or community volunteers (111).

Participation was enhanced when people could have frequent contact with the intervention provider (68). Interventions were more effective when they incorporated real world contexts (84) including opportunities to practice skills or participate in community settings (68–70, 104, 112–115) with a trained facilitator (69, 112). While a single session might be enough for some participants to learn new social skills, other people required booster support or continued training to maintain skills (116). Moreover, homework was considered to be helpful by participants in some programs (117), and helped to generalize skills into everyday life (91, 92).

Participation in sport or community groups was diminished for people with poor access to transport, lack of psychosocial supports or staff to encourage participation, and limited financial resources to continue to participate (38, 57, 58, 66, 105). For people with psychosocial disability feeling dependent on others (e.g., needing reminders) was also a significant barrier to participation (57, 58).

3.4.2.2. Supporter, staff, facilitator and mentor attributes

Staff were one of the most important sources of emotional and instrumental support to facilitate goal attainment in person-centred planning interventions (118). In art programs, moderators working side-by-side with participants facilitated inclusion and belonging (54). Physical activity participation was more successful if supporters consistently encouraged participation (56, 105–107, 119).

Befriending and peer mentorship were more successful when volunteers were matched to the participant attributes including personality, hobbies and interests (e.g., sports), age and gender (120). Successful peer mentors were understanding, empathic, punctual, flexible, and professional. Participation was improved when mentors took time to get to know their mentee and to establish a comfortable relationship (121), and set boundaries where necessary (e.g., in the study by Curtin, Humphrey (122) one mentee thought that they were dating their mentor). For some people it was important to be matched to a mentor with or without the same type of disability (123).

Building trust was integral to establishing a sense of safety in group programs (49, 50) and when working with social prescribers (32, 124, 125). People with psychosocial disability reported disengaging from activities if they felt unsafe, feared injury (32), had social anxiety, were apprehensive of strangers, or if they had negative expectations (e.g., feeling vulnerable, embarrassed, disliking feeling controlled by others, having to interact with others, or pain) (57, 58).

3.4.2.3. Participant attributes

Community and social participation was reported to be easier for people with friendly dispositions and relatively good social skills (101) or literacy (94, 96), but was hampered for people with low social capital, including low levels of education, literacy, and family finances (126). Conflicts with existing personal commitments or valued activities can impede physical activity participation in new programs (57, 58, 127). Some people reported being teased by other participants (102) or peers (128), which diminished their sense of belonging. Social prescribing interventions needed to establish realistic expectations as failure to achieve expected benefits could have negative impacts on confidence (129). Lower participation in physical activity for people with psychosocial disability was affected by lifestyle factors (e.g., smoking, diet, sleeping patterns, fitness level and confidence), intrusive or fluctuating psychiatric symptoms, fatigue, sedative effects of medications, and low self-esteem (56–58). For people with intellectual disability, continued physical activity participation can be hampered by advancing age of the participant or ageing parents (130).

3.4.2.4. Community-specific characteristics

Planning, linkage and befriending programs often faced difficulties with engaging people in the community who could foster community connections (131), or could not reliably recruit volunteers who could provide befriending support (111), leading to lower levels of engagement (132). Peer support workers (37), and volunteers (77) with psychosocial disability are at risk of experiencing stigmatising attitudes in the community, strain from over-commitment, and social exclusion.

4. Discussion

Interventions were identified that (a) support connection with social, community or civic participation opportunities, (b) provide participation opportunities that increase the sense of inclusion, belonging and participation, and (c) build capacity to enhance social skills and wellbeing to enable social and community participation. While most interventions successfully improved capacity and skills to participate, or actual levels of participation, some interventions had the potential to lead to worse participation or had negative impacts on quality of life. The findings reinforce the importance of individualised planning and support to identify and link people with participation opportunities, and to account for existing skills, social networks, and confidence to participate socially or in the community, as per the socio-ecological (133) and Quality of Life models (1).

4.1. Interventions for people with intellectual disability

For people with intellectual disability, interventions that consistently improved participation used individualised and person-centred approaches. While asset-based approaches provided excellent opportunities to enhance participation, other successful interventions targeted specific deficits, such as communication or social skills, or important life domains such as dating or parenting roles. The following interventions and supports effectively supported social and community participation for people with intellectual disability:

- Strengths or asset-based interventions to support choice making
- Person centred planning
- Skilled individualised interaction support
- Receiving support to link with or participate in community groups
- Peer support and transition programs both for youths transitioning into post-secondary opportunities and older people transitioning to retirement
- Group-based social skills interventions
- Vocational social skills support
- Relationship and family planning programs
- Parenting skills and knowledge interventions
- Helping people to create a “Life Story” to share their history
- Dog walking in the community
- Art and drama participation

- Participation in physical activity (e.g., Special Olympics programs)

Interventions with inconsistent effects on participation included digital literacy and participation programs, abuse prevention training, and travel or navigation training. Participating in physical activity and sports events helped people to build acceptance, confidence, sharing and friendships (134); however, there were inconsistent effects for sport-based activities in several studies. Befriending interventions were poorly implemented for people with intellectual disability, with the individual having limited choice and control over their interactions and experiencing potential negative impacts on existing social networks. To enhance effectiveness, befriending programs need to (a) define the target population; (b) balance frequency, length and modality of befriending activities; and (c) ensure there is appropriate infrastructure in the befriending services to support training and maintenance (132). Most of the evidence for interventions for people with intellectual disability was low or moderate quality, and many programs or supports needed to be adapted to each person's individual impairments, comorbid conditions, needs and preferences.

4.2. Interventions for people on the autism spectrum

Most interventions for people on the autism spectrum focused on building social, communication and relationship skills. The following interventions effectively supported social skills and participation of people on the autism spectrum:

- One-on-one training in social, conversation and communication skills
- Dating and relationship skills and knowledge programs
- Vocational social skills programs targeting social behaviours at work, including daily coaching
- Peer support, mentoring and support to transition into post-secondary education
- Music programs, and mindfulness training to reduce social anxiety

While people on the autism spectrum liked meeting with other people with similar disabilities and experiences, group-based social skills training had limited effects on social and communication skills. Instead, one-on-one training and individualised strategies targeting social and communication impairments were more effective. Interventions targeting higher order social competencies, such as theory of mind or social cognition, were not effective. Other interventions with inconsistent effects on social skills and participation included dance-based programs, and some vocational social skills programs (e.g., the Aspirations program, or training in job interview or conversational skills). Therefore, our findings oppose the NICE (135) guideline recommendation for social skills groups as a first-line treatment for people on the autism spectrum given that only individualized programs were consistently effective for adults. Finally, there were inconsistent effects on participation from disability-specific sports and physical activity programs, use of telehealth or SMS-based supports, psychoeducation, and interventions targeting meta-cognition (i.e., thinking about thinking) for people on the autism spectrum. Most studies were low quality, and few compared interventions with control conditions.

4.3. Interventions for people with psychosocial disability

For people with psychosocial disability, interventions enabling linkage with participation opportunities and building skills and psychosocial wellbeing to enable participation were effective, particularly when provided alongside illness management in line with the recovery framework (136). Interventions that effectively supported social and community participation of people with psychosocial disability included:

- Social prescribing, community linkage and "connecting people" interventions
- Befriending when matched with volunteers with common characteristics and interests
- Peer support, as a mentor or mentee, in community and clinical settings
- Social skill training in individual or group settings
- Training specific social competencies (e.g., theory of mind)
- Sex, relationship, and life skills training
- Art and music participation
- Ecotherapy, gardening, horticulture and outdoor nature-based activities and camps
- Sport and physical activity participation
- Vocational and internship programs with a focus on social skills or participation
- Psychoeducation and ehealth or SMS-based supports
- Recovery-oriented supports (e.g., psychoeducation) with a focus on social functioning and participation

While peer support and volunteer participation had several benefits, being a peer mentor in a clinical setting, or a volunteer in the community, could also lead to the experience of prejudice and stigma. Therefore, these opportunities need careful facilitation to minimise potential negative impacts.

Social skills training was not recommended in the most recent NICE guidelines for people with schizophrenia due to insufficient robust RCT evidence (137). However, we found consistent evidence of positive effects of social skills training on social functioning and social skills for people with psychosocial disability, with most evaluations published after 2017 using

RCT designs. Unlike studies with people on the autism spectrum, training higher order social competencies in people with psychosocial disability improved social skills. While most of the studies in both populations used RCT methods, the autism studies had small samples and were predominantly low quality whereas the psychosocial disability studies had large samples and were predominantly moderate-high quality, which may explain the different outcomes in each population. Behaviour activation, which is an approach that emphasises scheduling enjoyed activities, had no effects on social functioning.

4.4. Practical considerations for implementing interventions

Overall, the effectiveness of interventions was impacted by a range of factors. In particular the setting within which the intervention or support was provided, and the attributes both of the program and the attributes and behaviours of the supporters, staff, and/or facilitators. In brief, programs were more effective and acceptable if they were person-centred, used strengths-based strategies with supporters or facilitators who worked in partnership with the individual to enable them to exercise choice and self-direction. Moreover, it is important to note that the attitudes and behaviours of people in the community can impact positively (e.g., feeling like they are welcome and valued in a community group) or negatively (e.g., experiencing stigma or social exclusion when in community settings) on the experiences of people living with disability. Strategies targeting both specific settings (e.g., training and mentorship for community or sporting groups) through to broader education and integration of people with a range of disabilities into civic life could help to overcome some of these experiences.

Participant attributes also need to be considered when building social and community participation given that people with relatively good social skills, friendly disposition or literacy skills were better able to benefit from the supports offered. Therefore, it may be that people need multiple sources of support to build their capacity, or to ensure that community settings are welcoming, in order to enable people on the autism spectrum, and those with intellectual or psychosocial disabilities, to participate meaningfully in social or community settings.

4.5. Evidence gaps

Several types of intervention were not included in systematic reviews for some or all disability cohorts, despite growing evidence of their effectiveness. These include transition programs focused on independent living, supported education/transition support (138) and person-centred planning (139) for people with psychosocial disability. The utility of training to use communication support tools was limited to social media or email use. The effectiveness of augmentative and alternative communication aids and strategies has been studied and reviewed extensively in paediatric populations (140), but not in adults and therefore could not be included in this umbrella review. Interventions to support civic participation were limited to volunteering for people with psychosocial disability and building civic rights awareness in people with intellectual disability. Interventions targeting other types of civic participation such as voting or advocacy were not identified. Finally, interventions targeting inclusive community settings or environments (141, 142) are important in the social model of disability (14), but were not identified.

4.6. Limitations

This umbrella review was limited by the level and type of details reported in the respective systematic reviews, which were predominantly low to moderate quality. Most reviews provided little information about factors affecting the feasibility, acceptability, and effectiveness of different interventions. Most reviews did not report specific outcome measures used in each study, or the magnitude of effects. Moreover, few reviews described the resources or funding required to deliver the interventions. Cost-related impacts were only noted for two of the 522 studies, which highlighted that a social skills program for people on the autism spectrum was not expensive or time consuming (143), and that providing participation support did not increase overall support costs (144). An additional 260 studies published between 2010 and 2020 were identified that evaluated interventions that met our inclusion criteria, but had not been included in the systematic reviews, suggesting that some systematic reviews missed eligible studies.

The low-quality evidence in this review is likely to have been driven by several factors. First, disability research has historically been under-resourced, making it difficult to conduct large-scale robust RCTs. Moreover, social, communication and participation-related impairments in intellectual disability, the autism spectrum and psychosocial disability often vary substantially both within and between cohorts, and many people need individually tailored supports. Therefore, designs such as multiple baseline or case study approaches are often more suitable than RCT evaluations of standardised interventions. Studies evaluating interventions for people with psychosocial disability were typically better quality, and more often used RCT designs.

4.7. Conclusions

Overall, interventions that support people to have both the capacity and access to social and community participation opportunities improved participation for adults on the autism spectrum, with intellectual disability, and psychosocial disabilities. It is important that people have access to personalised supports, where possible, and that they are given the opportunity to practice skills with active support or mentoring in the community in real-life settings.

Data availability statement

The datasets for this study are included in the article/Suplementary Material. Further inquiries can be directed to the corresponding author/s. The study only included existing published data, and therefore did not undergo review by an ethics committee.

Author contributions

MJG designed the protocol, conducted the search, screening, data extraction and synthesis, and drafting and finalising the manuscript. IR contributed to study design, screening and reviewing the manuscript. LOB contributed to study design, data synthesis and reporting. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors were employees of the National Disability and Insurance Agency (NDIA) while conducting this project. The NDIA, oversees the National Disability Insurance Scheme (NDIS) in Australia. The NDIS provides information and connections to services in the community for all Australians with disability. People who have a permanent and significant disability who are under the age of 65 are eligible to enrol in the NDIS to receive funding for reasonable and necessary supports, equipment, and services to support their independence and to support their social and economic participation. The research was undertaken to help the NDIA to identify ways of better supporting social and community participation of people living with disability.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fresc.2022.935473/full#supplementary-material.

SUPPLEMENTARY DATA 1
Data extraction of SRs and AMSTAR ratings.

SUPPLEMENTARY DATA 2
Data extraction of studies

SUPPLEMENTARY FILE 1
Detailed eligibility criteria, including deviations from protocol

SUPPLEMENTARY FILE 2
Search strategies for each database

SUPPLEMENTARY TABLE 1
Review characteristics

SUPPLEMENTARY TABLE 2
AMSTAR quality appraisal

SUPPLEMENTARY TABLE 3
Intervention characteristics and effects

SUPPLEMENTARY TABLE 4
Facilitators and barriers to participation

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Physical activity programs for cardiovascular outcomes in community wheelchair users: A systematic review

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Purpose: Physical inactivity is one of the important factors leading to chronic diseases including cardiovascular disease (CVD) in individuals with disabilities. However, not many Physical Activity (PA) interventions are available for improving the efficacy of PA and cardiovascular outcomes among community wheelchair users. Therefore, this systematic review will appraise the existing PA interventions for the community dwelling wheelchair users; we especially examined features of the PA programs that showed the improvements in PA and the CVD outcomes compared to the interventions that did not show any improvements in these outcomes among these population. The study also aimed to provide some recommendations for future research.
Materials and Methods: A comprehensive and systematic search of literature published between 2015 and 2020 using the databases Scopus, Pubmed, Embase, and Cochrane CENTRAL was conducted. This review has followed the Preferred Reporting Items for Systematic Review (PRISMA) guidelines. The quality of the evidence was assessed by Using Joanna Briggs Institute's critical appraisal tool. Studies that tested the efficacy of PA interventions for community-dwelling adult wheelchair users and published in English were involved. Two reviewers reviewed the literature and any disagreements among these reviewers were resolved by a third reviewer.
Results: Fourteen articles were selected for this review. Most of the studies reported improvements in PA. A few studies followed up the participants and majority of the studies have looked at the CVD outcomes.
Conclusion: Large-scale studies with follow-ups, and community participatory research that evaluates the effect of PA interventions on PA and CVD outcomes among wheelchair users are needed.

KEYWORDS

wheelchair users, physical activity, cardiovascular disease, technology use, community

Introduction

In the United States, an estimated 2.7 million adults require the use of a wheelchair based on their physical disabilities (1). Cardiovascular Disease (CVD) is a major cause of morbidity and mortality in these populations (2). There is compelling evidence that wheelchair users with physical disabilities have an increased incidence of chronic diseases, including CVD, cancer, diabetes, and osteoporosis (3). Physical inactivity, unhealthy diet, and alcohol consumption are major risk factors for increased risk of chronic diseases including CVD in wheelchair users (4). The majority of hospitals and clinical facilities focus on short-term rehabilitation services to improve wheelchair user skills for mobility (5). However, after discharge from the medical facilities, the improvement in daily activity in this population remains a concern as there are limited physical rehabilitation services available for continuing the recommended physical activity (PA) in residential communities (6). Additional factors, including disease severity and adaptations, add to the burden of physical inactivity in these individuals. For example, wheelchair users with multiple sclerosis indicated reduced PA participation based on dependence on the mobility device, disease severity (7), and environmental adaptations (8). Half of the wheelchair users with chronic spinal cord injury (SCI) reported no leisure-time PA (6) due to the disease severity. This underscores a critical need to focus on PA promotion among wheelchair users to decrease the burden of CVD and improve their overall well-being and quality of life.

The center for disease control (CDC) has recommended moderate to vigorous-intensity PA and muscle strengthening exercises for individuals with disabilities (CDC, 2019). In addition to the reduce risks for chronic disease, PA is essential to regain mobility, improved walking ability, balance, fitness, proper gait, and functional-ability in individuals with disabilities (9). Therefore, these individuals should perform a PA on regular basis that involves bodily movement produced by skeletal muscle contraction resulting in more energy utilization than at resting levels (10).

Wheelchair-users living in community settings engage in insufficient amount of PA for health benefits (3). This may be explained by inaccessible and unaffordable care, low education, and environmental obstacles as critical barriers for engaging in PA (11). This has underscored the importance of designing and delivering feasible and efficacious PA programs for community-dwelling wheelchair users. To that end, we conducted a systematic review to summarize the literature related to PA programs for community-dwelling wheelchair users and understand the features of the PA interventions that improve the PA and CVD outcomes compared to the PA interventions that did not show any improvements in these outcomes for this vulnerable population. We also aimed to identifying gaps in the literature for informing future research.

Methods

Overview

Literature search strategy

We conducted a comprehensive and systematic search of literature published between 2015 and 2020 using the databases Scopus, Pubmed, Embase, and Cochrane CENTRAL. This time span was selected as most of the research on the efficacy of interventions for PA in wheelchair users was done during this time period. Articles were located using keywords: exercise, physical-activit*, pilot, yoga, sport*, martial-art, recreation, garden, sports, leisure activities, community health services, wheelchairs, wheelchair*, initiative, project, program, plan. These keywords were used as they helped to catch almost all the research studies involving any kind of physical activity interventions for our target population. A professional librarian at the University of Alabama at Birmingham helped with the Literature search. The appendix lists search strings and terms used for different databases (Appendix A).

Data extraction and analysis

Two reviewers independently vetted each abstract and full article to ensure the validity and suitability of each study for inclusion. These reviewers have received an intense training in developing systematic reviews and have published some reviews before. Any disagreements among these reviewers were resolved through discussion. If a consensus was not reached, a third party who has expertise in behavioral and physical activity interventions, and has published several reviews, independently reviewed the material and resolved disputes for the articles.

Inclusion criteria

Articles describing experimental research were included. This included single subject design, RCTs, multisite RCTs, and pre-post designs of feasibility, pilot, and efficacy studies that examined the effect of PA interventions delivered among community dwelling wheelchair users in the community settings. Database searches were limited to articles written in English and continued until December 20, 2020. Studies that looked at PA for wheelchair users outcome variable, and the population residing in community settings were included for review. Studies involving PA, including leisure time PA, exercises, gardening, sports, recreation, yoga, and martial arts, as well as robotic exoskeletons were included in the review.

Exclusion criteria

Studies involving populations below 18 years of age and not involving wheelchair users were excluded. Studies conducted in any setting other than the community settings were excluded. Studies published in any language other than English were excluded.

Quality rating

Using Joanna Briggs Institute's critical appraisal tool, the two reviewers assessed the quality of studies selected for this review (12). The tools for critical appraisal were selected based on the study design. A score of 6 was given to the quasi-experimental study and a score of 9 was given to the randomized controlled trials. Any disagreements for critical appraisal were resolved by a third-party consultation.

Results

There were 304 articles identified in our initial search. We removed four duplicate articles. After reviewing the titles and abstracts for the remaining 300 articles, 161 articles were excluded and 139 articles were selected for full text review. After full text review of 139 articles, 125 articles were excluded for the failure to meet criteria, which resulted in the inclusion of 14 articles; **Figure 1** provides the flow of article inclusion for this review (PRISMA flow diagram).

Table 1 provides the selected study characteristics. All of the studies were conducted in home/community settings. Study designs include single group intervention design (13, 14), randomized clinical trials (15–22), or multisite randomized clinical trials (23–25).

Ten studies were conducted on wheelchair users with spinal cord injuries (SCI) (13, 14, 16, 17, 20, 21, 23–26), Three studies had participants with immobility due to any disorder (15, 18, 27), one study involved persons with multiple sclerosis (19).

The duration of PA programs ranged between 6 and 12 months. The interventions included exercise interventions (16–18, 23), rehabilitation programs/physiotherapy programs (26), locomotor training programs with robotic exoskeleton (13, 20), hand cycling high intensity interval training (14), low intensity wheelchair training with treadmill propulsion (21), wheelchair and propulsions skills training (19), behavioral interventions including coaching, motivational interviewing (19, 23–25, 28).

These interventions were delivered in different modes including, face-to-face delivery, online delivery, and hybrid of both face-to-face and online sessions. Some of the interventions were delivered among participants individually, some in groups and some both involved both individual and groups sessions. The dose of the PA interventions also varied between these studies. Majority of the studies have delivered interventions weekly once or twice and the duration of a session varied between 25 (1), 30 min (6), 60 min (13). These PA programs are also varied in terms of the total time duration. The shortest intervention was 7 days long (17), and others varied from 6 weeks (24, 29) to a year long intervention (27).

Although the interventions were delivered for the community residential areas; however one of the studies had the intervention delivered in a rehabilitation center near the community residential areas, the participants were coming from their homes to attend the intervention sessions (13). Two studies delivered a component of intervention in the hospital setting and rest of the intervention was delivered in the participants homes (13, 30). All other studies have used participants' homes or community facilities to deliver the interventions in person or online.

The primary outcomes for this review is PA and CVD related outcomes. The secondary outcomes for this review were pain, aerobic endurance, cardiovascular fitness, wheelchair skills, body mass index, motivation, self-efficacy, mood, anxiety and depression, resilience, quality of life, participation and satisfaction with the intervention. Majority of the studies did not have any follow-up after cessation of the programs; three studies had one year follow up (18, 27, 30) and three studies had 3–4 month follow ups (19, 21, 26).

The PA outcomes were assessed differently in these studies, including leisure time PA, moderate to vigorous PA, minutes of walk, wheelchair propelled time and distance, standing time, self-reported PA (sports, hobbies, house hold and work related activities), PA measured as metabolic equivalents. Most of these studies reported improvements in PA (14, 16–19, 24, 27). Some of the studies further reported improvements in exercise self-efficacy (24, 27), depression and anxiety (16, 17), fatigue (19, 24), endurance (14), strength (19) quality of life (24) and the compliance with the PA intervention (17).

Nine studies have looked at the cardiovascular outcomes (1, 16–18, 20, 23, 27, 29, 30). The cardiovascular outcomes that were assessed include, heart rate, blood pressure (systolic and diastolic), body mass index, body weight, serum cholesterol, triglycerides, low-density lipoproteins, high-density lipoproteins, plasma glucose, insulin resistance assessment, cardiorespiratory fitness, Serum fasting insulin, lean and fat mass and visceral adipose tissue area.

Only three studies reported improvements in blood pressure, body mass index (27, 30), heart rate (1) and cholesterol, low density lipoproteins (30).

The studies involved in this review had high quality of methodology, however, due to the small sample size of most of these studies and the inconsistencies in PA measurement, the level of evidence for this review will be of low level.

Discussion

Community based PA programs for wheelchair users

Community-based programs for promoting PA for wheelchair users represents a prime opportunity for improving chronic diseases and other outcomes. These programs are scalable and sustainable, yet may not be as effective as supervised center-based programs. Unfortunately, there are not many community-based programs available for wheelchair users living in community settings. The studies included in this review were three multisite randomized controlled trials (RCTs) and seven RCTs, including four pilot

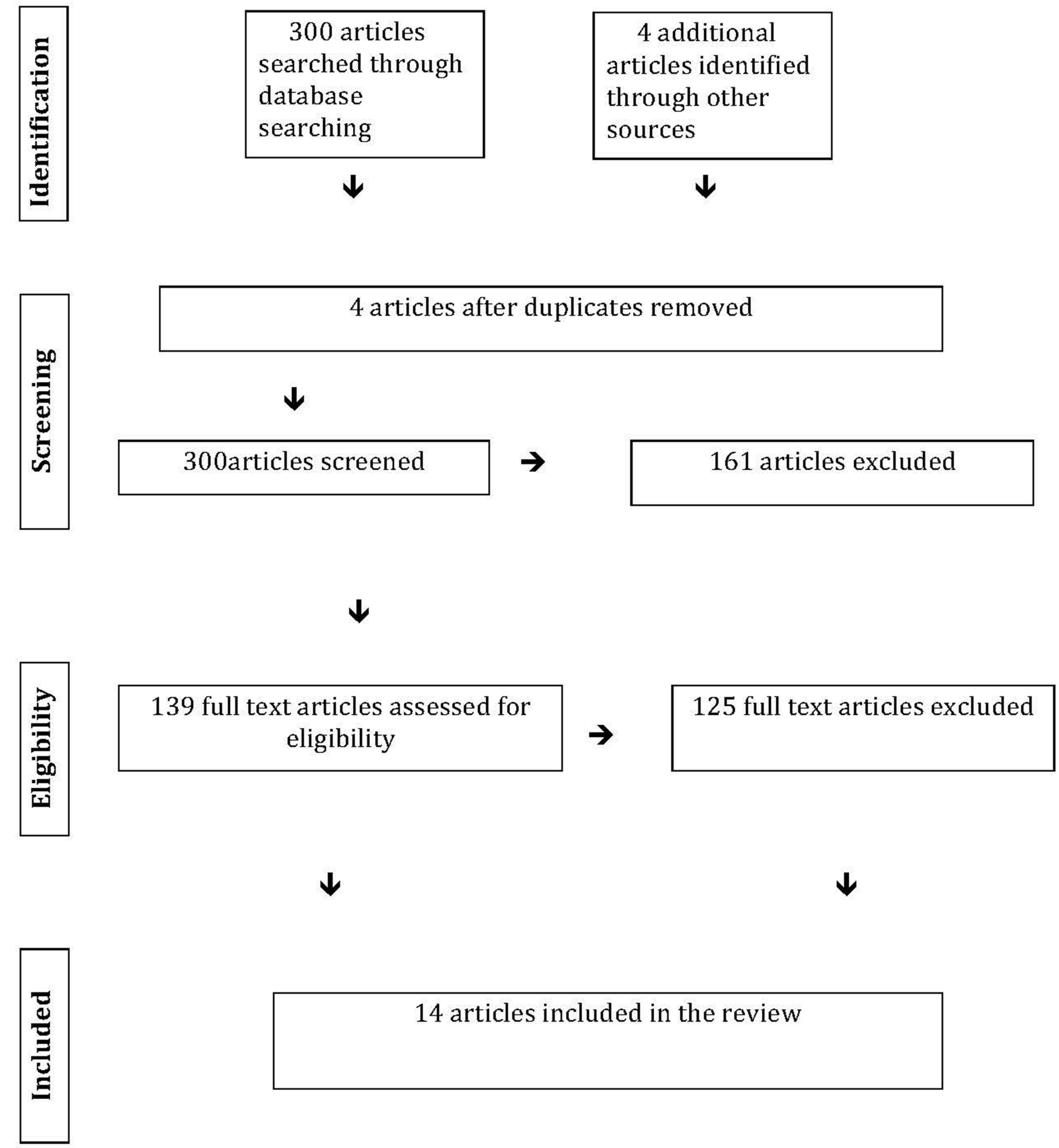

FIGURE 1
Show the data identification and stepwise data extraction including screening of the articles, removal of duplicates, and selection of the articles based on the inclusion and exclusion criteria.

RCTs. The majority of RCTs, including one multisite RCT (n = 6), reported an increase in PA from before to the after intervention phase; however, some RCTs ($n = 4$) did not report any improvements in PA among wheelchair users. The interventions in these studies differ for instance, behavioral and life style intervention, skills training, low and high intensity exercise training interventions. The interventions further differ in features including type of PA, individual vs. groups sessions, in-person vs. online delivery of intervention, length of whole intervention and duration of intervention sessions, use of technology, theory based vs. no theory-based interventions. These differences in interventions may explain the differences in efficacy of improving the PA among this population.

TABLE 1 Characteristics of the studies (PICO).

Study	Sample/ type of community setting	Study design	Experimental group Intervention type, dose, length	Comparison group intervention	Outcome variables	Results	Follow up
Best et al. (2017)	Manual wheelchair users living in the community ($N = 38$) Setting: Participant homes in community residencies	Randomized controlled trial (Pilot study)	The experimental group received a theory-based intervention that includes the existing PA guidelines for wheelchair users, a toolkit with recommendations for PA, and a mobile phone (with phone number, data plan, and preloaded Facebook page). They received a Smart Phone PA Counseling (SPPAC) program. These sessions were delivered by the trainer *via* mobile phone device. The intervention uses motivational strategies to make the program suitable for the participants' individual preferences. Dose: The fourteen SPPAC sessions were delivered to participants in 10 weeks and each session was of 30 min duration.	The control group received the PA guidelines same the as that of the experimental group. However, no SPPSC program was delivered to this group.	**PA Outcomes:** - PA measured by actigraphy - Leisure time PA **Others** - PA motivation - Self-efficacy to overcome barriers to PA participation - Anxiety and depression - Satisfaction with the psychological need for PA - Satisfaction for participation in the study - Wheelchair skills - Wheelchair use confidence	Study does not report the results yet.	Ten weeks Three months
Bombardier et al. (2020)	Underactive manual wheelchair user adults with SCI for more than a year with at least 2 cardiometabolic diseases risk factors ($N = 15$) Setting: Participants' homes or community facilities.	Randomized controlled trial (Feasibility study)	The experimental group receives the treatment *via* phone. It is adapted from a 16-session diabetes prevention program. This is a multi-component intervention that includes a home exercise tool kit (exercise bands with soft grip, ankle strap and door anchor), 16 sessions of PA counseling curriculum by a psychologist *via* phone and a DVD for verbal instructions and a video about stretching, aerobic exercises, and strength training specifically for individuals with paraplegia or tetraplegia. The PA counseling was done by psychologists using motivational interviewing techniques and SMART goal setting to promote the adherence to PA program.	The control group received the usual care control condition. This group received a letter advising them to seek medical care to make lifestyle changes in diet and exercise to address their cardiometabolic risks.	**PA Outcomes:** - PA: minutes/wk for leisure time PA **CV outcomes** - Cardiorespiratory fitness ($V_{Peak}O$) - BMI - Waist circumference - Lean and fat body mass - Insulin sensitivity index - Lipid panel: LDL, HDL **Others:** Depression - Health-related QOL - Pain related to PA and wheelchair use	Post-test at 6 months showed the following non-significant changes: - improvements in MVPA - decrease in depression - increase in pain in both groups but less pain in the experimental group	Six months

(*continued*)

Table 1 (Continued)

Study	Sample/type of community setting	Study design	Experimental group Intervention type, dose, length	Comparison group Intervention	Outcome variables	Results	Follow up
Coulter (2017)	Individuals with SCI ($N = 24$) Setting: participants' residential homes	Randomized controlled trial (Pilot study)	The experimental group received a web-based physiotherapy for 8 weeks twice a week. Each exercise page has a video, audio, and a written explanation of the exercise. Individual exercise programs (aerobics, strengthening, balance exercises) were prescribed to the experimental group participants according to their abilities. They also completed their online exercise diaries. The physiotherapists delivered the intervention electronically *via* website. They also contacted participants *via* email and phone calls every 2 weeks. Dose: Participants were supposed to perform 30-min exercise sessions twice a week for 8 weeks.	The control group received usual care consisting of self-management of their condition. If these participants were performing any exercises or gym, they were asked to maintain a daily diary and record the exercises and activities that they participated.	**PA Outcomes:** - PA: 6 min push test and 6 min walk test - PA compliance **CV Outcomes:** - Aerobic endurance - HR (work HR and resting HR) **Others:** Acceptance and feasibility of the intervention - Quality of life - Anxiety and depression - Mobility - Perceived exertion - Muscle strength	- A non-significant improvements in the PA of experimental group as indicated by 6 min push test and 6 min walk test - PA compliance was improved in intervention group compared to control group (1.4+_0.8 times/ week) - Participants were highly satisfied with the program - Anxiety depression improved in intervention group (effect size = 0.23) - Improvements in pain, strength and quality of life of the experimental group.	None
Divanuglou (2019)	Individuals with SCI ($N = 7$) Setting: Participants residential homes	Prospective cohort study (Protocol for A pilot study)	Active Rehabilitation (AR) training program which is a 7 day program will be delivered to the participants. AR consists of an online wheelchair skills program for wheelchair users	No comparison group	**PA Outcomes:** - Moderate and vigorous PA - Leisure time PA **Others:** - Wheelchair skills test - Wheelchair using confidence and capacity - Self efficacy (personal functioning, social functioning, general self-efficacy) - Life satisfaction - Resilience - Health related QOL	The study does not report the results yet.	Three months

(*continued*)

Table 1 (Continued)

Study	Sample/ type of setting	Study design	Experimental group Intervention type, dose, length	Comparison group intervention	Outcome variables	Results	Follow up
Froehlich-Grobe (2020)	Individuals with immobility. The cause for immobility included SCI, spina bifida, multiple sclerosis, amputation and age related immobility (*N* = 23). Setting: Some intervention sessions were delivered telephonically in participants' homes and some sessions were in-person in a hospital settings.	Randomized controlled trial with wait list	The experimental group received a Group Lifestyle Balance Program adapted for Individuals with Impaired Mobility (GLB-AIM). It is a 12 month weight loss intervention in which 23 sessions were delivered weekly for 13 weeks, followed by two biweekly sessions. Afterwards monthly sessions were delivered. The session were delivered online and once a month sessions were delivered face to face. The intervention prompts weight loss through reducing calorie intake and by eating healthy diet; It emphasizes walking to increase the PA.	The control group was on waitlist and received the same intervention as the experimental group after 6 months.	**PA Outcomes:** - Walk and wheel time **CV Outcomes:** - Weight - BMI - Waist circumference - BP - Cholesterol - HbA1c **Others:** - Self-efficacy of health behaviors (nutrition, exercise, health promotion, physical well being).	- Experimental group participants had an increase in their MPA minutes of physical activity/ week, whereas it was decreased in waitlist on control group - The experimental group also had increase in walk/wheel time minutes per week. They had a significant decrease in their weight, BMI and waist circumference at 3 months, 6 months and 12 months time point. No changes in these outcomes were seen in the control group at these time points. - Both groups had significant increases in self-efficacy of health behaviors.	Three months Six months Twelve months
Gagnon (2017)	Longterm wheelchair users with SCI (*N* = 14) Setting: Out patient rehabilitation center for community residents	Single group intervention study (Feasibility study)	The participants in experimental group received two familiarization sessions followed by eighteen locomotor training sessions with robotic exoskeleton that were completed in 6–8 weeks. During each training session, participants completed sit-stand transitions, performed quasi-static and dynamic standing balance exercises, and walked with assistance of a rollator walker or with assistance under supervision. These sessions were based on the individual participant's ability to perform these activities. Dose: Participants had two to three 60 min session per week.	No comparison group	**PA Outcomes:** - Standing time, walking time and number of step **Others:** - Perceived motivation to engage in physical activity - Learn to perform sit-to-stand and walk with robotic exoskeleton. - Perceived health benefits, risks and fears associated with engaging in PA. satisfaction with robotic exoskeleton locomotor training	- Participants provided positive feedback for engaging in PA (sit to stand and walking) with exoskeleton. - The standing time, walking time and number of steps were increased by 45.3%, 102.1%, and 248.7% from before to after intervention phase. - health benefits including overall health status, limbic strength and endurance were perceived to be with locomotor training program.	None

(*continued*)

Table 1 (Continued)

Study	Sample/type of community setting	Study design	Intervention type, dose, length	Comparison Group Intervention	Outcome variables	Results	Follow up
Kooijman (2017)	Individuals with SCI for atleast 10 years who are using wheelchair and were physically inactive ($N = 64$) Setting: Participants' homes in community residential areas	Blinded multicenter Randomized controlled trial	A theory based intervention called "HABITS" was delivered to the experimental group participants. HABITS involves one home visit, five individual and five group sessions delivered in 16 weeks. This intervention facilitates active lifestyle and development of self-skills management by guidance from a counselor, peer support and experiencing task accomplishment to strengthen self-efficacy. These consisting of group meeting, individual counseling and a book. Motivational interviewing was used in counseling the participants.	Control group received information about active lifestyle in a group meeting and a book "How to Stay Fit with SCI."	**.PA Outcomes:** - Self-reported PA (sports, hobbies, house hold and work related activities) - The amount of self-propelled wheelchair driving measured by using accelerometer **CVD Outcomes:** - Aerobic capacity - BMI **Others:** - Exercise self-efficacy - Proactive coping (dealing with possible future situation) - Social support - Functional independence - Mood - Perceived behavioral control - Exercise self-efficacy - Attitude towards behavior change - Readiness to change with regards to regular exercise - Fatigue - QOL	No significant within and between-group difference was found in any of the primary and secondary outcomes.	- Sixteen weeks - Forty two weeks

(*continued*)

Table 1 (Continued)

Study	Sample/type of community setting	Study design	Intervention type, dose, length	Comparison Group Intervention	Outcome variables	Results	Follow up
Koontz (2020)	Wheelchair users with SCI ($N = 7$). Setting: Participants' home in community residential areas	Non-randomized clinical trial (pilot study)	Participants received a Hand Cycling High Intensity Interval training (HIIT) with a trainer for 6 weeks, each session comprised of 2–3 min warm ups, followed by ten intervals of cycling with a ratio of one min work at 90% peak power output (PPO) to one min recovery at 0–20% PPO, the two to three min cool down. Dose: HIIT session consisted of two weekly 25-minute sessions.		**PA Outcomes:** - Moderate to vigorous PA, - Adherence to PA sessions - Wheelchair propulsion **CVD Outcomes:** - BMI - HR Aerobic capacity - Oxygen consumption **Others:** - Fatigue - QOL - Satisfaction - Training efficacy - Perceived effectiveness of the training - Endurance - Transferability	- Participants expressed a high level enjoyment - Participants had an increase in their physical capacity HR after receiving the intervention.) - improvements were seen in participants' training efficacy, endurance, aerobic capacity, transferability, wheelchair propulsion, PA after the intervention. - Extra workout time/ week did not increase - Volume of $V_{Peak}O$ increased.	None
Nightangle (2017)	Wheelchair users with SCI for more than a year ($N = 21$) Setting: Participants' homes in community residential areas	Single blinded multicenter randomized control trial	Home-based moderate intensity upper body exercise intervention is a 6 weeks intervention. The exercise sessions consisted of moderate-intensity arm crank exercise. Dose: The participants received four forty-five-minute exercise sessions per week.	The control group received lifestyle maintenance intervention. They were asked to maintain their habitual PA behavior.	**PA Outcomes:** - Moderate to vigorous PA - Energy intake and expenditure **CV Outcomes:** - VO_{2Peak} - Cardio-respiratory fitness - Sr. fasting Insulin - Body mass - Fat and lean mass - Visceral adipose tissue area - HR - Sr. Triglycerol - Total Cholesterol - HDL - Nonestrified fatty acids - Plasma glucose **Others:** - Health related QOL - Fatigue - Shoulder pain - Exercise self-efficacy	Post-test in six months showed significant improvements in health-related QOL, and fatigue. - Compared to control group, the participants in the experimental group had significant improvements in moderate to vigorous physical activity (with moderate to large effect size) and energy expenditure. - Significant improvement in exercise self-efficacy and PV O2Peak of experimental group.Significant decrease in experimental group participants' insulin resistance, fasting Sr. insulin concentration, body mass.No changes in any other cardiometabolic disease biomarkers in participants of any of the groups.	None

(*continued*)

Table 1 (Continued)

Study	Sample/ type of community setting	Study design	Intervention type, dose, length	Comparison Group Intervention	Outcome variables	Results	Follow up
(Nooijen (2017)	Wheelchair users with subacute SCI (N39) Setting: The intervention was delivered for 2 months in an in-patient rehabilitation and for rest of the 6 months it was delivered in participants' homes in community residential areas	Single blinded multicenter randomized controlled trial	The experimental group participants received a regular rehabilitation for 2 months in the inpatient rehabilitation. They also received a behavioral intervention consists of 13 individual face to face sessions with a coach trained in motivational interviewing. The behavioral intervention sessions were delivered twice a month starting before the discharge from inpatient rehabilitation, it continued for 3 months after the discharge. After this one session per month was delivered for three months. Dose: The study does not indicate the duration of the intervention sessions.	Control group only received the regular rehabilitation during their in patient rehabilitation stay. They did not receive any intervention after their discharge from rehabilitation.	**CV Outcomes:** - VO_{2Peak} - BMI - systolic and diastolic blood pressure - lipid profile - total cholesterol, LDL,HDL - triglycerides - glucose levels **Others:** - QOL	- Significant improvements in diastolic blood pressure of the experimental group as compared to the control group. - Between and within the group improvements in BMI, Peak power output, and general health perceptions were seen; however, these changes were not significant. - Significant improvements in cholesterol, LDL in experimental group compared to the control group.	- Six months - Twelve months
Rice (2015)	Wheelchair users with multiple sclerosis ($N = 14$) Setting: Participants' home, general community setting, and university laboratory	Randomized control trial	The experimental group participants received a regular rehabilitation for 2 months in the inpatient rehabilitation. They also received a behavioral intervention consisting of 13 individual face-to-face sessions with a coach trained in motivational interviewing. The behavioral intervention sessions were delivered twice a month starting before the discharge from inpatient rehabilitation, it continued for 3 months after the discharge. After this one session per month was delivered for three months. Dose: The study does not indicate the duration of the intervention sessions.	The control group were tested for propulsion on treadmill on custom fitted ultralight wheelchair. They did not receive any technique training. They were asked to continue using their primary devices for mobility. Unlike the participants in the intervention group, these participants did not receive any technical training or behavioral intervention. Their PA was measured by the accelerometers like that of the experimental group.	**PA Outcomes:** Activity counts **Others:** - Fatigue - upper extremity strength	- Increased strength significantly in experimental group - decreased fatigue in experimental group but not significant - improved propulsion skills significantly in the experimental group. - An improvement in activity counts in the experimental group than in the control group, however, this difference was not significant.	- Three months

(*continued*)

Table 1 (Continued)

Study	Sample/type of community setting	Study design	Experimental group Intervention type, dose, length	Comparison group intervention	Outcome variables	Results	Follow up
Spungen (2020)	Wheelchair-using veterans with SCI ($N = 160$) Setting: Participants' homes in community residential areas	Non-blinded randomized control trial	The intervention group received standardized care plus exoskeleton-assisted walking (EAW) advanced training for 30 sessions. The EAW training helped the participants to learn how to be safe and competent to use an exoskeleton for walking at home/community during the intervention phase of 16 weeks. The companions of these participants had to attend at least one-third of these sessions. The participants had to pass the EAW advanced skills test after which they were allowed to take the exoskeleton home and use them for their daily mobility in addition to carrying out the usual Standard of Care (SOC) activities. They also received a multimedia presentation to prevent upper limb pain and fatigue. They were given a social cognitive theory-based behavioral intervention in their homes telephonically by a physical therapist to teach them strategies related to how to maintain the PA. The PA including distance traveled, time spent in propulsion, and propulsion speed was also measured by accelerometer for this group. Dose: The intervention was 8–9 months long including participants pretesting and measurements for device fitting measurements. The study does not specify the time duration for the intervention components including the technical training and telephonic calls.	The control group participants did not receive any EAW advanced training or exoskeleton for their walking. However, they were advised to continue the SOC activities. Like the experimental group, these participants were asked to call for help and guidance if they have any questions or problems. The control group intervention was from 7 to 8 months duration.	–**PA Outcomes:** - Ambulation indoor and outdoor **CV Outcomes:** - Total body fat mass - Sr. lipid profile: low-density lipoproteins, triglycerides, total cholesterol - Insulin resistance **Others:** - QOL - Physical medical health domain: complaints of bladder, bowel, and pain - Mental health summary scores, -Sleep disturbance - Social participation domain - Spinal cord injury functional index	The study has not reported the results outcome measures yet	None

(*continued*)

Table 1 (Continued)

Study	Sample/type of community setting	Study design	Intervention type, dose, length	Comparison group Intervention	Outcome variables	Results	Follow up
Van der Scheer (2016)	Community-dwelling inactive manual wheelchair users with SCI ($N = 29$). Setting: Intervention was delivered in the rehabilitation center	Randomized controlled trial	The experimental/ exercise group received a low-intensity wheelchair training for 16 weeks. This consists of a wheelchair, and treadmill propulsion at 30–40% heart rate reserve or equivalent. The 30 min exercise sessions were delivered twice a week.	The Control group did not receive any intervention.	**PA Outcomes:** - Physical activity measured as metabolic equivalents (MET) using PASIPD (home/week on a 0–180 scale) and propelled distance in a week in the community assessed using an odometer placed on a participants' daily wheelchair. **Others:** - PO_{peak} - Wheelchair fitness and skills, performance time, ability and strain score.	- No significant effects of the intervention on PO_{peak} of the experimental group. The Control group improved in PO_{peak}. - No significant effect of the intervention on wheelchair-specific fitness except P5–15 m. - No significant effects on wheelchair skills performance. - No significant changes in Physical activity levels.	- Two months - Four months
Froehlich-Grobe (2014)	Wheelchair users with sufficient upper arm mobility for arm-based exercises ($N = 128$) Setting: Community residential areas based on the participants' preferences (participants' homes, recreation centers)	Randomized controlled trial	Theory-based multi-component exercise intervention. The experimental group or staff-supported group received intensive support from the staff for exercise. These participants received one day of educational information, workshop, and individual exercise plans were developed. Resistance bands, instructions to self-monitor exercise, fifteen regular phone calls and handwritten cards for birthdays, holidays, and major events. They received a monthly newsletter.	The comparison group or the Self-guided group received minimal support from staff; however, these participants received exercise information which was reviewed with them during the first phone call. The rest of the 14 phone calls were made to say thank you and request to return the logs and report about exercise-related injuries. They also received the resistance bands, instructions to self-monitor exercises,, and handwritten cards similar to that of the experimental group	**PA Outcomes:** - Weekly self-reported exercise - PA measured by Accelerometer data **CV Outcomes:** Peak aerobic capacity - Body weight **Others:** - Physical fitness - Predictors for exercise participation	- Experimental group significantly showed greater exercise minutes per week compared to the control group. - Self-reported exercise minutes were moderately correlated with accelerometer data. - No significant between-group differences in aerobic capacity and strength over 12 months. - Both groups had a similar number of exercise logs returned.	- Six months - Twelve months

Abbreviations: PA, Physical Activity; QOL, Quality of life; HR, Heart rate; LDL, Low density Lipoprotein; HDL, High density lipoprotein; MET, Metabolic Equivalent of Task; PASIPD, Physical activity scale for individuals with physical disability; VO_{2Peak}, Peak oxygen uptake; BMI, Body Mass Index; SCI, Spinal cord Injury; CV, Cardiovascular.

Lifestyle and behavioral interventions focusing on self-efficacy and adherence

Lifestyle behavioral interventions involved teaching people skills and strategies for increasing PA as part of daily life and may improve PA among wheelchair users. An increase in PA in a large magnitude (vector counts at wrist) were seen in wheelchair users with multiple sclerosis after receiving a three months behavioral intervention in addition to a wheelchair skills training. This intervention focused on enhancing self-efficacy, overcoming barriers, and identifying facilitators, in addition to the behavioral strategies of self-monitoring, goal setting, planning, optimizing outcome expectations (19). Weekly follow-ups were done telephonically with the participants to teach them about initiation and maintenance of PA. Another behavioral intervention with in-person individual sessions reported a significant improvement in PA and the cardiovascular outcomes of BMI, cholesterol, blood pressure, and low-density lipoproteins at one year follow up. However, these differences were not significant between the experimental and control groups. The intervention had a component of motivational interviewing, that might have increased the adherence to the intervention (25). Hence, the interventions that targeted on improving the PA self-efficacy seem to be promising for these populations. In additions some measures to improve the adherence of intervention must be incorporated into the PA interventions for better efficacy.

Contrary to the above studies, in a multisite RCT, a sixteen-week theory-based intervention that involved a home visit, 5 individual and 5 group counseling sessions and a book did not increase the self-reported PA of wheelchair users in the experimental group. The intervention focused on developing participants' active lifestyle and their self-management skills. There were no within or between-group differences in PA at four- and ten-months follow-ups. The control group had received information about active lifestyle by one group meeting and a book (23). Like other behavioral studies (19, 25), this intervention targeted PA self-efficacy, and behavioral control, however, the baseline self-efficacy in these participants was high, indicating that the intervention was not formulate based on the needs of this population. This could be explaining the ineffectiveness of this intervention on PA. In addition, this study included individuals with SCI for more than 20 years, the authors of this study believe that the long duration of their disabilities of this sample might have made their behavioral tendencies toward inactivity, thereby challenging for them to engage in the PA.

One recent study by Froehlich-Grobe et al. (2020) delivered a 12 month weight loss program to wheelchair users. The intervention was delivered in hybrid form, *via* telephone, and in-person. The intervention had a dietary and a PA component. The waitlist control group received the intervention after six months. Experimental group participants had a significant increase in minutes of walk and wheel time per week, and further had a significant weight loss after six months of starting the intervention. Both groups had a significant increase in self-efficacy of health behaviors (27).

Previously, Froehlich-Grobe et al. (2014) had delivered a theory-based multi-component exercise intervention to experimental and control groups ($N = 128$). The staff support group received additional intensive staff support for exercise, while the self-guided group received minimal support. The staff support group significantly increased exercise (17 min/week) compared to the self-guided group. There was no significant difference in aerobic capacity and strength over twelve month time period (18). Although staff-assisted interventions may show greater efficacy in improving PA, the time and cost related to such interventions affect their sustainability, especially in socio-ecologically deprived communities. Therefore, in order to develop sustainable programs, it is crucial to develop cost-effective interventions, and utilize previously existing resources from communities into these programs.

Community involvement is critical for community participatory research so as to use the already existing resources and develop interventions that are need based and acceptable to a community (31); therefore, it is vital to involve community dwelling wheelchair users when developing health promotion intervention for them. The knowledge about their needs, preferences, resources, barriers and facilities of a community is critical for the development of PA interventions (32). Cole et al. (2019) conducted a feasibility study in which participants' ($N = 7$) opinion related to the content, delivery, and self-management strategies of an evidence-based exercise intervention was assessed. Based on this information, this study developed a six months Workout on Wheels internet intervention (WoWii), which will be evaluated in the future (33). Such community-based research in which community-dwelling wheelchair users are involved in developing PA interventions is scarce. Hence, there is a critical need for such culturally sensitive PA interventions. These interventions may potentially show better acceptability, adherence, and sustainability in community settings as seen in other disciplines (34). Thereby will be more efficacious in improving wheelchair users' PA and their cardiovascular outcome.

The behavioral and lifestyle PA interventions report mixed results with regards to improvements in PA among wheelchair users. Therefore, novel lifestyle behavior programs that are tailored to the needs of these populations should be developed and tested by studies with strong methodology and larger sample sizes. The individual, interpersonal, and environmental levels factors that are found to be related to the adherence to community-based interventions (28) can be incorporated into these programs to improve consumer compliance. Studies should also examine the ecological validity and long-term sustenance of these programs in community settings. In addition, the long-term effects on cardiovascular outcomes are scarcely studied and must be focused in future research.

Wheelchair skills training programs

Wheelchair skills training programs demonstrated mixed results regarding the changes in PA of community-dwelling wheelchair users. Two RCTs delivered the wheelchair propulsion/skills training interventions to wheelchair users with SCI reported improvements in PA (14, 19). A custom-fit ultra-lightweight manual wheelchair propulsion and skills training revealed an increase in activity counts, strength and propulsion skills, and decreased fatigue of wheelchair users with multiple sclerosis ($n = 14$) compared to the participants in the control group. The experimental group had received theory-based behavioral intervention consisting of the moderate intensity wheelchair skills training and weekly telephonic follow-ups to teach behavioral strategies for initiation and maintenance of PA. Control group participants did not receive any training (19). In contrast, a sixteen-week low-intensity wheelchair propulsion-training program did not show any significant changes in PA levels of the experimental group participants. The participants received twice a week 30-minute sessions. Also, there were no improvements in their wheelchair propulsion and PO_{peak}. The study indicates that low-intensity training to be insufficient to improve PA for wheelchair users with long term disabilities (21).

These studies suggest contradicting effects of wheelchair training on PA, and more studies are needed to confirm the efficacy of such interventions on PA of wheelchair users living in community settings. Van der Scheer et al. (2016) had delivered intervention to community dwelling wheelchair users in a rehabilitation setting; the barriers to access for such interventions must be focused especially in relation to availability of these resources in these underserved populations.

Exercise training programs

In a multicenter RCT, home-based moderate intensity upper body exercise intervention delivered in-person to wheelchair users with chronic SCI ($n = 21$) showed improvements in their PA. This behavioral intervention consisted of a PA component and a dietary component; it involved 45-minute weekly sessions for six weeks. The control group received lifestyle maintenance intervention in which they were asked to maintain their routine PA behavior. The study reported moderate ($d = 0.62$) to large ($d = 1.37$) effect sizes for improvements in PA, cardiorespiratory fitness, and exercise self-efficacy among the intervention participants. These improvements were seen after six weeks of intervention; the study did not do any follow up to see the long-term effects (24). Since the intervention was personalized to each participant's needs and delivered in the home setting, it improved exercise self-efficacy, which is reported to be a key factor in improvements in PA (35, 36). Delivering such interventions that are accessible to the community residents overcomes the barriers of the lack of transportation, time and access. These factors resulted in low attrition (11%) and increased adherence to the intervention, which might explain the large effect size of the intervention outcomes.

Recently, hand cycling high-intensity interval training intervention delivered to the wheelchair users ($n = 7$) showed an increase in the participants' PA. The participants received the intervention in three weekly sessions for six weeks. The participants also showed an increase in their PA heart rate (max.), training efficacy, endurance, aerobic capacity, and wheelchair propulsion skills. The study did not do any follow-ups. The intervention was tailored to each participant's pace and the study used some measure to assess and prompt the intervention adherence (14). These programs can be tested in future studies with large sample size to confirm efficacy in these populations.

Robotic exoskeleton to improve PA

An eight-week locomotor training program with EKSOTM (version 1.1) robotic exoskeleton was delivered among long-term wheelchair users ($N = 14$) to see its effect on their PA, motivation to engage in PA and performance capability to stand transfers. The intervention consisted of 2–3 training sessions per week in which participants completed sit to stand transitions, quasi static and standing balance exercises, and walked with assistance of a rollator walker while contact-guarded by a physical therapist. The walk with assistance of rollator walker was followed by walk with robotic exoskeleton. There was a significant increase in walking speed after the completion of intervention ($P < 0.0001$) (37). The participants provided positive feedback for the robotic exoskeleton and learning about walking capability and performance of sit-to-stand transfers. The participants' perceived an increase in their motivation to engage in leisure time PA adapted to their condition. In addition, participants also perceived positive effects of this program on their overall health, endurance, and upper limb strength (13).

Another RCT, still in the implementation phase, is planning to deliver exoskeleton assisted walking to experimental group participants (wheelchair users with SCI), in addition to standard care ($N = 160$). The control group is receiving only standard care. The study aims to see the effect of this intervention on indoor and outdoor ambulation, body fat mass, serum lipid profile, insulin resistance, social participation, and sleep disturbance (20). The results of this study are not yet reported. These studies show varied types of locomotor training with varied duration. Therefore, making it difficult to make any conclusions about the dose and duration of the locomotor training interventions. More experimental studies are needed to see the effectiveness of locomotor training with robotic exoskeleton on the PA efficacy of community-dwelling wheelchair users. Studies should be conducted to understand the duration of each type of training and the mechanisms involved in improving PA and cardiovascular health outcomes due to robotic exoskeleton training in these populations.

Technology based interventions for changing lifestyle PA

Technology offers a way to reach the increased number of populations in less time and helps eliminate the barriers (like lack of transportation and physical therapist time) that interfere with the delivery of care services. The majority of the studies included in this review ($n = 6$) used technology in the interventions. Use of technology (e.g., telephone calls, exercise videos delivered *via* web or DVD, smart phones apps.) in interventions seems to be promising for improving PA efficacy and intervention adherence among wheelchair users (16, 17, 27). Bombardier et al. (2020) adapted and delivered a multipronged diabetes prevention program to the experimental group ($n = 7$). This intervention used the telephone calls for delivering counseling sessions and DVDs videos for delivering aerobic exercise and strength training sessions to the experimental group. The control group participants ($n = 8$) received usual care involving advice to seek preventive care for cardio metabolic risk factors. Posttest at six months indicated though not significant, but there was an increase in experimental group participants' leisure-time PA (minutes of walk per week). There were significant improvements in their exercise self-efficacy and depression levels, also the likelihood of increase pain was significantly low in experimental group ($P < 0.05$) (16). Coulter et al. (2017) reported an improved adherence to web-based physiotherapy intervention in the experimental group participants ($n = 16$). The intervention was delivered for 8 weeks with two sessions per week and was tailored to each participant's individual needs. The intervention consists of strengthening, stretching, aerobic, and balance exercises as appropriate based on participants' abilities. The control group ($n = 8$) were advised to do the self-management for their condition. The participants in experimental group had moderate effect size ($d = 0.40$) improvements in their mobility and endurance. The study also reported improvements in PA compliance and depression in experimental group compared to control group (17). Thereby, increasing their PA performance in everyday life.

These pilot studies suggest that technology-based interventions yield improvements in PA and intervention adherence among wheelchair users, yet studies with large sample size and diverse sample in terms of severity of disability and wheelchair dependence should be conducted to help draw some concrete conclusions. For example, Best et al. (2017) has planned to deliver a 12-week Smart Phone Peer PA Counseling (SPPAC) program *via* phone to the experimental group participants, whereas the control group participants are supposed to receive PA guidelines only. The study aims to improve participants' autonomy to get engaged in leisure time PA as measured by Actigraph, PA self-efficacy and their motivation. The study is reported to be in implementation phase (15). Based on this study, Best et al. (2019) have developed a theory based Active Living Lifestyle Program (ALLWheel) for wheelchair users with SCI. This program is aimed to reach large number of individuals in their communities to improve their self-efficacy, motivation and autonomy, thereby improving their engagement in leisure time PA (38). The intervention is anticipated to have high participant adherence due to the benefits of technology use including flexibility in timing, independence in performing the intervention and lack of need for transportation and scheduling.

These studies have used simple technology (e.g., telephone, web delivery of home exercise videos, DVDs), which can be easily available in rural communities. However, so far, none of the RCTs that have technology-based interventions have been conducted on community dwelling wheelchair users. The use of technology could be useful to overcome the ecological barriers related to remote areas, and their sustenance in these health services deprived areas (39). Therefore, there is a need for studies with controlled designs to test efficacy of technology-based interventions to improve PA and cardio metabolic outcomes in rural dwelling wheelchair users. In addition, theory based and community partnership research should be emphasized to increase the sustenance of these programs.

Limitations

There is a possibility of some articles not being captured by the search strategies of this systematic review. The above findings should be interpreted carefully as there is a potential bias caused due to the limitations imposed by small sample size of the studies involved in this review. Limitations of this review also arise from the inconsistencies in PA measurement, and other confounding factors.

Conclusion

The purpose of this review is to gather knowledge about the availability of PA programs focused on improving PA and CVD outcomes among wheelchair users living in community settings. The knowledge gathered from this review illustrates that behavioral and lifestyle interventions have produced promising results in improving PA and CVD outcomes. Although the incorporation of technology that is mostly available to community-dwelling wheelchair users (telephone calls, DVDs, web-based exercise videos) has helped to overcome the facility barriers in community settings, none of the studies were community participatory research. As such, it is critical to involve community resources and partnerships for the ecological validity and sustainability of such PA programs in community settings. Most of the studies are small-scale pilot studies, and only a few have focused on the improvements in CVD outcomes in these populations. Large-scale studies with advanced methodology and long-term follow-ups are required to make any concrete conclusions and interpretations.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Author contributions

Data extraction, data analysis. All authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Appendix A

Embase

(exercise*:ti,ab OR ’physical activit*’:ti,ab OR gymnastic*:ti,ab OR calisthenic*:ti,ab OR run*:ti,ab OR jog*:ti,ab OR swim*:ti,ab OR walk*:ti,ab OR climb*:ti,ab OR ’weight lift*’:ti,ab OR pilate*:ti,ab OR qigong:ti,ab OR ’qi gong’:ti,ab OR danc*:ti,ab OR ’tai ji’:ti,ab OR ’tai chi’:ti,ab OR ’tai ji quan’:ti,ab OR taiji:ti,ab OR taijiquan:ti,ab OR ’tai chi chuan’:ti,ab OR yoga:ti,ab OR sport*:ti,ab OR athletic*:ti,ab OR baseball:ti,ab OR softball:ti,ab OR basketball:ti,ab OR netball:ti,ab OR bicycling:ti,ab OR cycling:ti,ab OR boxing:ti,ab OR cricket:ti,ab OR football:ti,ab OR rugb*:ti,ab OR golf*:ti,ab OR hockey*:ti,ab OR wrestl*:ti,ab OR ’martial art*’:ti,ab OR ’hap di do’:ti,ab OR judo:ti,ab OR karate:ti,ab OR jujitsu:ti,ab OR ’tae kwon do’:ti,ab OR aikido:ti,ab OR wushu:ti,ab OR ’kung fu’:ti,ab OR ’gong fu’:ti,ab OR gongfu:ti,ab OR mountaineer*:ti,ab OR tennis:ti,ab OR racquetball:ti,ab OR racketball:ti,ab OR ’racket ball’:ti,ab OR badminton:ti,ab OR lacrosse:ti,ab OR skating*:ti,ab OR skateboard*:ti,ab OR snowmobiling:ti,ab OR sledding:ti,ab OR skiing:ti,ab OR snowboard*:ti,ab OR soccer:ti,ab OR track:ti,ab OR volleyball:ti,ab OR surfing:ti,ab OR rowing:ti,ab OR polo:ti,ab OR kayaking:ti,ab OR canoeing:ti,ab OR boating:ti,ab OR surfboarding:ti,ab OR recreation*:ti,ab OR ballet:ti,ab OR ’hip hop’:ti,ab OR jazz:ti,ab OR tap:ti,ab OR salsa:ti,ab OR fitness:ti,ab OR ’exercise’/exp OR ’sport’/exp OR ’recreation’/exp OR ’fitness’/exp OR ’physical activity’/exp) AND (’wheelchair’/exp OR wheelchair*:ab,ti OR scooter*:ab,ti)

Retrieves 123

PubMed

("Exercise"[Mesh] OR Exercise* [tiab] OR physical-activit* [Title/Abstract] OR pilate*[Title/Abstract] OR yoga[Title/Abstract] OR Sport*[Title/Abstract] OR martial-art [tiab] OR recreation*[Title/Abstract] OR garden*[Title/Abstract] OR "Sports"[Mesh] OR "Recreation"[Mesh] OR "Leisure Activities"[Mesh] OR "Community Health Services"[Mesh]) AND ("Wheelchairs"[Mesh] OR wheelchair* OR scooter*) AND (initiative*[Title] OR project*[Title] OR program*[Title] OR plan*[Title])

Retrieves 79

Scopus

TITLE-ABS-KEY (initiative* OR project* OR program* OR plan*) AND TITLE-ABS-KEY (exercise* OR physical-activit* OR sport* OR recreation* OR leisure) AND TITLE-ABS-KEY (wheelchair* OR scooter*)

Retrieves 94 results

CENTRAL

ID Search Hits

#1 MeSH descriptor: [Leisure Activities] explode all trees 18458

#2 MeSH descriptor: [Exercise] explode all trees 24606

#3 MeSH descriptor: [Community Health Services] explode all trees 13872

#4 (Exercise* OR physical-activit* OR pilate* OR yoga OR Sport* OR martial-art OR recreation* OR garden*):ti,ab,kw 128302

#5 (initiative* OR project* OR program* OR plan*):ti 48466

#6 (wheelchair* OR scooter*):ti,ab,kw 848

#7 MeSH descriptor: [Wheelchairs] explode all trees 197

#8 #1 OR #2 OR #3 OR #4145083

#9 #6 OR #7848

#10 #5 AND #8 AND #932

#11 #8 AND #9324