

Physical & Occupational Therapy In Pediatrics

ISSN: (Print) (Online) Journal homepage: <https://www.tandfonline.com/loi/ipop20>

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To cite this article: Caroline Elfassy , Sarah Cachecho , Laurie Snider & Noemi Dahan-Oliel (2020) Participation among Children with Arthrogryposis Multiplex Congenita: A Scoping Review, *Physical & Occupational Therapy In Pediatrics*, 40:6, 610-636, DOI: [10.1080/01942638.2020.1754319](https://doi.org/10.1080/01942638.2020.1754319)

To link to this article: <https://doi.org/10.1080/01942638.2020.1754319>



Published online: 16 Apr 2020.



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Participation among Children with Arthrogryposis Multiplex Congenita: A Scoping Review

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ABSTRACT

Aim: To explore what is currently known regarding participation among children and youth with arthrogryposis multiplex congenita (AMC) using empirical studies, gray literature, and YouTube videos. The secondary objectives included identifying activity types, outcome measures used, interventions provided, and barriers and facilitators to participation.

Method: Empirical studies and gray literature were searched through electronic databases and videos were searched on YouTube. Articles and videos pertaining to participation and youth with AMC were included by two reviewers. Data regarding activity types, location, outcomes measures, interventions, and barriers and facilitators to participation was extracted. Data was critically appraised using specific evaluation criteria.

Result: Eleven empirical studies, six gray literature articles and 71 videos met the inclusion criteria. The most common activity types reported in the empirical studies and YouTube videos were active-physical, social, and skill-based activities. Outcome measures included evaluations and questionnaires, none of which were designed to address participation. Interventions did not target participation although the environments could affect participation.

Conclusion: The paucity of research indicates a need for future studies of participation in this population. Interventions should target participation and address environmental barriers. Videos provide insight for clinicians, youth, and families to help promote participation in the natural environment.

ARTICLE HISTORY

Received 14 June 2019

Accepted 5 April 2020

KEYWORDS

Arthrogryposis multiplex congenita; participation; rehabilitation; scoping review; online videos

Introduction

Health does not solely rely on physical function but rather is influenced by a multitude of factors including psychosocial well-being, participation and the interplay with the individual's environment (Townsend & Polatajko, 2007). The International Classification of Functioning, Disability and Health (ICF) provides a framework and classification system to measure health care needs based on a common language and definition of key words (World Health Organization, 2001, 2007). According to the ICF framework, *Body Functions and Structures* can be defined as physiological function as well as anatomical

parts of the body and its systems. *Activities* is defined as the execution of tasks or actions by an individual (e.g., mobility, self-care, communication, school related tasks). *Participation* refers to involvement in a life situation (e.g. community, social, civic activity). This can include participation in leisure activities such as sports or music, participation in paid or unpaid work, and social relationships (WHO, 2001, 2007). Participation can be influenced by the individual's environment, personal factors, and by health factors. In childhood, participation in different life roles, at home, school or in the community, is important as it contributes to the child's health and development. It allows the child to develop new competencies and skills, promotes self-confidence, and helps with developing new relationships (King et al., 2004).

Increased participation in leisure activities has been shown to have a positive effect on the overall quality of life of children with cerebral palsy, as well as for children with other neurodevelopmental disabilities (Badia et al., 2013; Dahan-Oliel et al., 2012). Additionally, the inability to participate due to a chronic disease or disability has been shown to cause marginalization, social isolation, and lowered self-esteem (Novak & Honan, 2019). These negative outcomes may be exacerbated among youth who live with a rare condition, such as arthrogryposis multiplex congenita (AMC) (Ejerskov et al., 2015).

AMC is a term that describes congenital joint contractures in two or more body areas (Cachecho et al., 2019) and affects 1 in 3000 live births (Fahy & Hall, 1990; Staheli, 1998). Deriving its name from Greek, it literally means "curving of joints" (Staheli, 1998). Depending on the underlying diagnosis, different systems in the body such as the respiratory, gastro-intestinal, genito-urinary, and central nervous system may be affected (Cachecho et al., 2019; Staheli, 1998). Contractures develop in-utero due to decrease in fetal movement resulting in joint fibrosis and stiffness (Kowalczyk & Feliś, 2016). Contractures, which vary in distribution and severity, may cause limited joint movement and muscle weakness in the involved body areas. While contractures do not progress to previously unaffected joints, they may change over time due to growth and intervention. Further, the varying severity and distribution of the contractures may impact and limit independence in mobility, activities of daily living (Amor et al., 2011; Ho & Karol, 2008; Spencer et al., 2010). Early intensive rehabilitation is warranted to provide splinting and orthosis management, range of motion exercises, strengthening programs as well as surgical interventions to correct deformities and thus promote daily function (Bender & Withrow, 1989; Bernstein, 2002; Sells et al., 1996; Sodergard et al., 1997). A recent qualitative study seeking to identify the rehabilitation needs of youth with AMC, their parents and clinicians , reported that participation was a priority by youth with AMC (Elfassy et al., 2019). Although, outcome measures assessing participation exist, this construct is oftentimes overlooked in the management and rehabilitation of children and youth with AMC (Elfassy et al., 2019; Riner & Sellhorst, 2013).

In response to concerns raised at the Second International Symposium on Arthrogryposis in St-Petersburg in 2014, an urgent need was identified for the development of guidelines for AMC regarding early diagnosis, orthopedic management, and rehabilitation (Hall et al., 2015). An emphasis was placed on the need to promote autonomy in daily activities, quality of life, and participation and integration in the community (Hall et al., 2015). Further, clinicians reported that, given the rarity and

heterogeneity of AMC, rehabilitation professionals often did not feel adequately prepared or lacked knowledge when treating individuals with AMC (Elfassy et al., 2019). Therefore, the interventions and care provided to children and youth with AMC, specifically focusing on participation, differ based on expertise and clinical settings. A need to explore what is already known about participation of youth with AMC is required in order to identify gaps in knowledge and clinical practice to promote participation.

Prior to the advent of the internet, medical journals and scholarly articles were the main source of information for healthcare professionals and patients alike (Tan & Goonawardene, 2017). Now that encyclopedic information resources are available online, 60-80% of users obtain health information on the internet (Powell et al., 2003). Users may educate and empower themselves by becoming knowledgeable about a subject and more involved in the informed decision-making for their own healthcare (Hardey, 1999). Fox (2002) stated that two thirds of those using the internet to find information report some impacts on their healthcare decisions (Fox, 2002). Although internet accessibility has the potential to provide important healthcare information, the number of educational videos created by institutions are rather small (Farkas et al., 2015). In response, health educational guides and how-to videos have been created by youth with AMC, their caregivers, support networks and online platforms. These videos are most often shared on the YouTube platform, one of the most popular video-sharing websites worldwide making it an important source of publicly available information (PAI). YouTube videos are based on lived experience and often provide day to day examples of what it is like to live with a disability. They enable individuals to demonstrate and showcase their abilities and encourage others with similar conditions to pursue their independence in completing tasks of daily living, in participating in various leisure activities, and in developing their overall autonomy. Educational videos from support networks are also a valuable and accessible source of information providing tools and resources for youth and their families. Thus, the internet now presents an opportunity for important information pertaining to participation to be transmitted directly to those interested. Similar to other sources of information, such as medical journals and scholarly articles, the content of these PAI should be critically appraised to ensure the information is valid and reliable. Therefore, the overall objective of this scoping review was to explore what is currently known regarding participation among children and youth with AMC using empirical studies, gray literature, and PAI- specifically YouTube videos.

Methodology

Design

A scoping review is a form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge (Arksey & O'Malley, 2005). This design was preferred over a systematic review as this review did not aim to answer a specific research question, rather it explores and presents the existing knowledge on the topic of participation in children and youth with AMC (Munn et al., 2018). The current scoping review used

Arksey and O’Malley’s (2005) framework in addition to incorporating recommendations by Levac et al. (2010) (Arksey & O’Malley, 2005; Levac et al., 2010). This framework consisted of five steps: i) identifying the research question(s), ii) identifying relevant studies, iii) selecting studies, iv) charting the data and v) collating, summarizing and reporting results (Arksey & O’Malley, 2005; Levac et al., 2010).

Identifying the Research Question

The objective of this scoping review was to identify and explore what is known surrounding participation for children and youth with AMC. Specifically, the secondary objectives were the following: 1) In what activity types do children with AMC report being engaged? 2) What are the outcome measures used to evaluate the level of participation? 3) What are the interventions used to promote participation? 4) What are the facilitators and barriers to participation in this population?

The five dimensions of the Children’s Assessment of Participation and Enjoyment (CAPE) tool were used to identify and classify participation throughout this scoping review. The CAPE is an assessment tool for participation in day-to-day activities outside of mandated school activities of children between 6-21 years of age (Imms, 2008; King et al., 2004). The dimensions of participation of the CAPE include diversity, intensity, with whom, where and enjoyment. Furthermore, the CAPE categorizes five activity types which include recreational (e.g., playing video games or doing puzzles), active-physical (e.g. team sports or bicycling), social (e.g., going to the movies or hanging out with friends), skill-based (e.g., taking art classes or learning to dance), and self-improvement (e.g., reading or doing a chore) (Supplemental Online Material 1 for detailed definitions) (Imms, 2008; King et al., 2004). For the purpose of objectivity and consistency, location was categorized into three of the five domains of the CAPE (i.e. home, school, and community rather than in the relative’s home, the neighborhood, at school but not during class, beyond the community) to facilitate categorization when details regarding location are limited.

Identifying Relevant Studies

In order to provide a comprehensive representation of participation, this first of its kind scoping review will utilize a search of empirical studies and gray literature as well as PAI in the format of online YouTube videos. With guidance from an expert librarian in the field of rehabilitation, a search strategy for electronic databases Ovid MEDLINE (1946 – November 2019), CINAHL (1977 – November 2019), Ovid EMBASE (1974 – November 2019), PsycINFO (1987 – November 2019), and OTSeeker (2002 – November 2019) was created. These databases were selected in order to conduct a comprehensive search that would include *participation* from a range of various healthcare professions. The databases were used to reach a broad-based search strategy of English and French literature and aimed to include quantitative, qualitative and mixed-methods studies, therefore no limitation was placed in terms of publication data and publication type. The gray literature including theses and dissertations was searched in ProQuest and OpenSigle. PAI consisting of YouTube videos, as this platform is the most popular video-sharing websites worldwide, is an important database for publicly posted videos.

Table 1. Search terms used in Ovid MEDLINE (1946-2019). Adaptations were made in other databases to incorporate search strategy.

#	Searches	Results
1	arthrogryposis.mp. or exp ARTHROGRYPOSIS/	2643
2	amyoplasia.mp.	136
3	Contracture/	7877
4	multiple congenital contracture.mp.	14
5	exp Hand Deformities, Congenital/	3767
6	exp Clubfoot/	3683
7	participation.mp. or exp PATIENT PARTICIPATION/	175511
8	play.mp. or exp "Play and Plaything"/	654351
9	leisure.mp. or exp Leisure Activities/	236200
10	school.mp. or exp Schools/	298492
11	handwriting.mp. or exp HANDWRITING/	3753
12	physical education.mp. or exp "Physical Education and Training"/	15543
13	exp "COOKING AND EATING UTENSILS"/ or exp COOKING/ or cooking.mp.	22002
14	exp "Activities of Daily Living"/	98222
15	exp EMPLOYMENT, SUPPORTED/ or exp EMPLOYMENT/ or employment.mp.	121720
16	driving.mp. or exp AUTOMOBILE DRIVING/	91959
17	exp SOCIAL MEDIA/	6610
18	TECHNOLOGY/ or exp INFORMATION TECHNOLOGY/	9468
19	exp CELL PHONE/ or phone.mp.	26669
20	computer.mp. or exp Computers/	716972
21	1 or 2 or 3 or 4 or 5 or 6	17471
22	7 or 8 or 9 or 19 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20	2309347
23	21 and 22	650
24	limit 23 to ("all infant (birth to 23 months)" or "all child (0 to 18 years)")	322
25	("20180301" or 2018030* or 2018031* or 2018032* or 2018033* or 201804* or 201805* or 201806* or 201807* or 201808* or 201809* or 201810* or 201811* or 201812* or 2019*).dt,ez,ed.	3062357
26	24 and 25	23

To identify relevant studies, members of the research team entered the search terms in English in varying combinations into the electronic databases. A complete list of search terms in all databases can be found in **Table 1** and adaptations were made for the different databases. For the YouTube videos, the following combination of search words was selected: “arthrogryposis and sport”, “arthrogryposis and art”, and “arthrogryposis and play”. These keywords were selected as they represent broad categories of activities children and youth typically engage in.

Selecting Studies

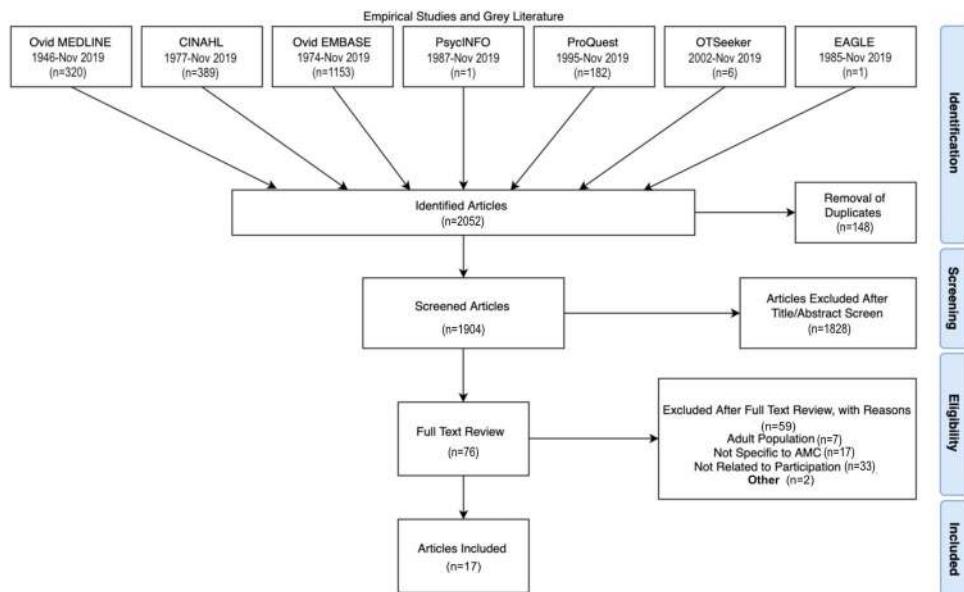
For the selection of research articles, both empirical studies and gray literature, the search strategy was conducted in two phases based on the inclusion and exclusion criteria (**Table 2**): 1) an initial title and abstract screening, followed by 2) full-text review of the articles. Two reviewers screened all titles and abstracts then reviewed the selected full texts (C.E and S.C). A third reviewer (N.D-O) was included to resolve any disagreements in the selection process. There were 16 conflicts in the abstract and title screening which represents 99% agreement. A PRISMA flow chart was used to track the number of articles at each stage of the review (Moher, Liberati, Tetzlaff, Altman, & The Prisma Group, 2009) (**Figure 1**).

For the PAI, the YouTube videos to be screened were divided between two reviewers (C.E and S.C), and were selected based on the inclusion and exclusion criteria (see **Table 2** and **Figure 2**). For the empirical studies, gray literature, and videos, two reviewers

Table 2. Selection criteria for empirical studies, gray literature, and publicly available information.

Empirical Studies and Gray Literature

Inclusion Criteria	Exclusion Criteria
Children and youth ages 0-18 years	Individuals >18 years
AMC or multiple congenital contracture, Amyoplasia, distal arthrogryposis and associated syndromes	Not AMC related diagnosis
Participation as an outcome as categorized by the CAPE (i.e., recreational, active-physical, social, skill-based, self-improvement)	Outcomes not related to participation or explicitly stated (i.e., not related to participation in a life situation)
Qualitative, quantitative, mixed methods, theses, dissertations, pamphlets, abstracts	Review articles
English and French	Other language
Publicly Available Information (PAI)	
Inclusion Criteria	Exclusion Criteria
Children and youth ages 0-18 years	Individuals >18 years
AMC or multiple congenital contracture, Amyoplasia, distal arthrogryposis and associated syndromes	Not AMC related diagnosis
Participation as main activity as categorized by the CAPE	Context other than participation
YouTube videos	None
English and French	Other language

**Figure 1.** PRISMA flow chart for empirical studies and gray literature.

independently applied the selection criteria to an initial sample of 5% of identified data and subsequently discussed any differences in order to ensure consistency and strong inter-coder agreement in applying the selection criteria. Once all members were in agreement, the selection criteria was then applied to full-text articles and online videos.

Charting the Data

A pre-defined data extraction form was created for both types of data. For the empirical studies and gray literature, the following information for each article included in the

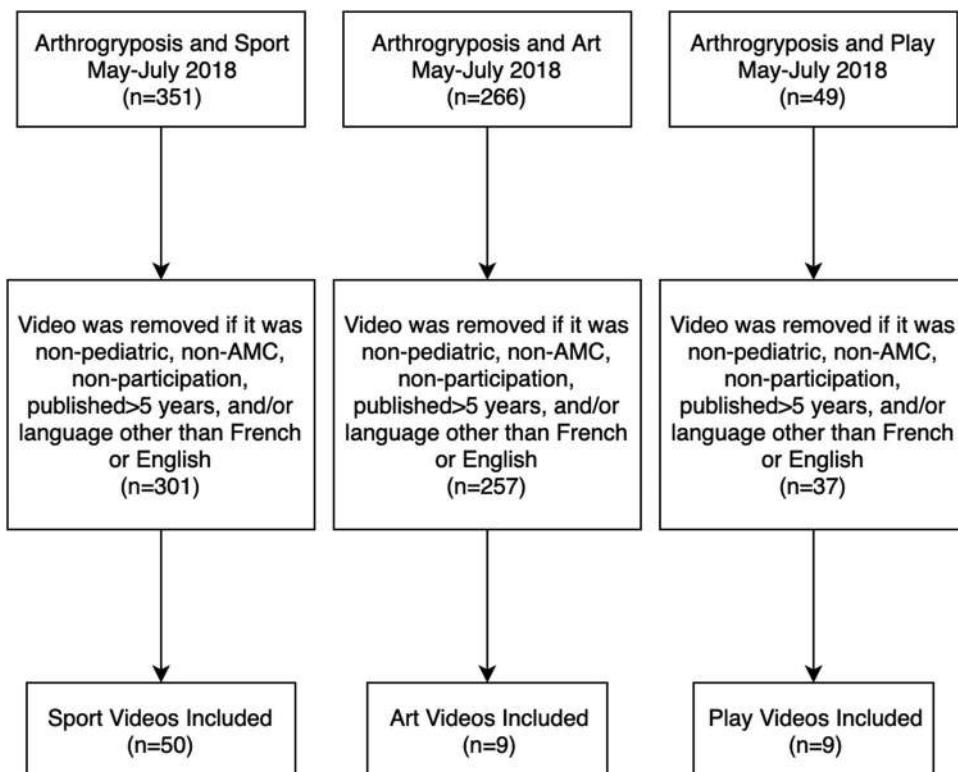


Figure 2. YouTube videos screened and included.

study was extracted by the principal author (C.E): author names, year of publication, article title, study aims/objectives, patient population, age of sample, activity type and location of participation as defined by the CAPE, outcome measures used, results of study, and key findings (Table 3). The CAPE was used to organize activity type (i.e. recreational, active-physical, social, skill-based, self-improvement) and location in a consistent manner. The CAPE provides an extensive list of activities that fall under each activity type. It also classifies different environments into categories of location (i.e., community, home, school). Therefore, this tool was used to organize and analyze the information about participation (empirical studies and gray literature) in an objective and systematic way for comparison purposes.

Methodological quality of empirical studies was appraised by the principal author (C.E) using the McMaster University's Critical Review Form for Quantitative and Qualitative Studies which evaluates the quality of the study based on eight sections: study purpose, literature, design, sample, outcomes, intervention, results, and conclusion or clinical impactation (Hand et al., 2011; Law, 1998; Letts, 2007). Two forms are available for this tool, one for quantitative studies and one for qualitative studies. This single appraisal tool was used for all the empirical studies that were included in order to compare and contrast between them, despite the differences in study designs. The AACODS Checklist (Authority, Accuracy, Coverage, Objectivity, Date, and Significance) is a critical appraisal tool specifically created for gray literature (Tyndall, 2010). Authority refers to the source of the information, the authors' credentials or organizational affiliations

**Table 3.** Characteristics of included studies.

First Author (Year)	Study Aims	Study Design	Patient Population	Patient Age	Results	Author Conclusion
Empirical Studies (n = 12)						
Amor et al. (2011)	To establish normative PODCI scores for children with Amyoplasia, and to determine whether the PODCI is sensitive to differing levels of severity of Amyoplasia and able to detect a change in the functional abilities of affected children over time	Cohort Study	Children with Amyoplasia (n = 74)	Mean age of child 8.5 ± 4.4 years	Mean scores for children with Amyoplasia were significantly lower than those without musculoskeletal disorder	The PODCI is useful in evaluating functional outcomes of children with Amyoplasia, and is sensitive to change in function over time. The PODCI shows promise as a tool to evaluate long-term outcomes of surgical management in Amyoplasia.
Azbell and Dannemiller (2015)	To document the frequency and timing of interventions for an infant with AMC following a clinical decision-making process	Case Report	Male with AMC (n = 1).	11 days to 9 months	Improvement of PROM improved, decrease of pain scores decreased during PROM, increase in PSFS score, decrease in PDMS standard score locomotion and stationary skills	Occupational and physical therapy interventions contributed to improvement of body functions and participation in some activities.
Babik et al. (2016)	To evaluate the feasibility and effectiveness of the WREX device for an infant	Case Report	Male with Amyoplasia (n = 1)	8 months to 13 months	Improvement in time spent contacting objects across a larger space looking at objects, improved unimanual and bimanual contact with objects, less use of his mouth when wearing p-WREX. AROM of shoulder flexion improved and muscle strength increased	Exoskeleton may be feasible and effective assistive devices for immediate improvements in infants' reaching and visual attention abilities, and may need to be paired with targeted intervention activities for optimal rehabilitative benefits
Babik et al. (2019)	To evaluate play in young children with AMC and to assess the assistive and rehabilitative effects of the Playskin on play with objects for young children with AMC.	Longitudinal cohort study (ABA single-subject design)	Children with AMC (n = 17)	Mean age 13.9 (±8.7 months)	The Playskin offered significant improvement to children's ability to play and reduced compensatory arm movements. While wearing the Playskin, children significantly	Overall, positive effects of Playskin on object exploration, manipulation in terms of assistive effects and rehabilitation effects

(continued)

Table 3. Continued.

First Author (Year)	Study Aims	Study Design	Patient Population	Patient Age	Results	Author Conclusion
Bagley et al. (2011)	Assess the factor structure and item-level statistics of the ASKp38.	Retrospective Study	children and adolescents (arthrogryposis ($n = 13$), cerebral palsy ($n = 105$)), fatigue ($n = 64$), limb salvage ($n = 18$))	Mean age 12 years 7 months (SD 2 years 8 months)	Item-fit statistics based on this two-factor model demonstrated adequate fit and content coverage for the AMC sample: Mean ASKp score (SD) range 68.4 (20.3) 31.6–95.1	The ASKp38 has two factors defined as activities of daily living and play. There is a need to reanalyze revised outcome instruments because the addition and modification of items may change the instrument's underlying conceptual structure.
Dalton and Hoyt-Hallett (2013)	Explore through case reports, occupational deprivation experienced by two pre-teen children with physical disabilities and the use of assistive technology to enable occupation and remove injustices	Case Report	Female with Amyoplasia and global developmental delay ($n = 1$), female with cerebral palsy ($n = 1$)	12 years	Improvement in communication, academic performance, and mobility.	Occupational enablement is facilitated through use of assistive technology and reduces social exclusion in children with physical disabilities and support participation in occupations.
Dillon et al. (2009)	To objectively describe the ambulatory and physical activity of ambulatory youth with arthrogryposis who had undergone multiple operative and therapeutic interventions and to compare their	Cohort Study	Children with arthrogryposis ($n = 13$), typically developing children ($n = 13$)	Arthrogryposis (10.91 years, SD 3.57 years), typically developing children (11.03 years, SD 3.53 years)	Youth with arthrogryposis took significantly fewer steps, spent less time at high activity levels, and had significantly lower parental report of ambulatory and physical activity than controls.	The SAM provides information related to the quantity of ambulation, as well as its quality and variability in real world situations. Coupled with other outcomes tools it can provide a more complete



<p>activity with an age- and sex-matched control group using the Step Watch Activity Monitor (SAM) and the Activities Scale for kids</p>	<p>To determine the long-term functional outcome of patients treated for arthrogryposis with surgical knee releases</p>	<p>Retrospective Study</p>	<p>Children with arthrogryposis (n = 32)</p>	<p>Mean age at time of knee release was 2.7 years (0.8-6.8 years)</p>	<p>Improvement in knee flexion and extension contractures , on FMS decreased WeeFim, PEDI and PODCI over time</p>	<p>Ambulatory ability improved in the short term but may decline over time with growth</p>
<p>Lobo et al. (2016)</p>	<p>To describe the design process and initial testing of a novel exoskeletal garment, the Playskin lift.</p>	<p>Case Report</p>	<p>Male with arthrogryposis (n = 1)</p>	<p>23 months</p>	<p>Playskin is inexpensive, easy to use, comfortable, and attractive. While wearing the device the child was able to contact objects more often throughout an increased play space, to look at toys more while contacting them, to perform more complex interactions with toys and for each toys at chest and eye level</p>	<p>Playskin is inexpensive, easy to use, comfortable, and attractive. While wearing the device the child was able to contact objects more often throughout an increased play space, to look at toys more while contacting them, to perform more complex interactions with toys and for each toys at chest and eye level</p>
<p>Pritchard-Waert et al. (2019)</p>	<p>To describe modified toy car use by children with physical disabilities in home and community settings</p>	<p>Mixed-Methods Case Series</p>	<p>Children with cerebral palsy (n = 4), child with AMC and hypotonia (n = 1)</p>	<p>Age range from 13-58 months (SD = 19)</p>	<p>Daily driving time ranged from a mean of 1.3 to 2.9 days per week, 12-63 min/session,</p>	<p>Modified toy cars are feasible and accessible option for exploring early assisted mobility with young children with mobility limitations who do not require extensive seating supports or adaptations.</p>

(continued)

Table 3. Continued.

First Author (Year)	Study Aims	Study Design	Patient Population	Patient Age	Results	Author Conclusion
Spencer et al. (2010)	To document the bone mineral density (BMD) of children with Amyoplasia and predominantly lower extremity arthrogryposis and compare it with age normative values	Cross-sectional Study	Children with arthrogryposis (n = 30)	Mean age 10.9 years (SD 4.4 years);	AMC scored lower than TDY. Early childhood is a crucial time for developing of BMD. There is linear relationship between BMD and the ambulatory level of children with arthrogryposis. Future studies needed to look at the long-term effect.	Two simple tools
Gray Literature (n = 7)	Beng Ting Lau (2009)	To examine the experiences M.A Thesis of children with physical disabilities in their play spaces and to understand what they perceived as the affordances and constraints of their play experiences. Specifically, aim was to understand these children's experiences in local school and community play spaces from their own perspectives	Dissertation	children with a range of physical disabilities (spina bifida, amputation, CP, arthrogryposis) n = 8	Physical environment: size of play space allows for more participation and access may limit participation. Social environment: variety of play partners (peers and adults) with positive play experience; attitudes and behaviors also have an impact. Play provides the opportunity to practice and achieve success. However, there is a lack of play choices and insufficient play time. Participants personal strengths and abilities are affordances to positive play experiences.- personal limitations due to lack of knowledge or experience	Two subcategories within environment emerged: physical and social, each with positive and negative aspects. Findings showcase that children should act as active beings to create their own worlds (through play participation).
Larson (1954)	To assess the adequacy of the general experience	PhD Thesis	children with disabilities	Age ranging from 3 to 6 years	The socializations score for TDY group are	For each area mean of experience score was

<p>background of children with disabilities by comparing their experiences in various critical areas with those of typically developing children of the same age and social status</p> <p>(cerebral palsy n = 74, poliomyelitis n18, other physical disabilities including arthrogryposis n = 43) and 135 typically developing youth (TDY)</p>	<p>To explore through the use of qualitative methods, the experiences of youth with physical disabilities with their peers in sport contexts using a self-determination theory lens.</p> <p>M.Sc. Thesis Dissertation</p>	<p>Adolescents (cerebral palsy n = 3, hip growth plate damage n = 1, Developmental Coordination Disorder n = 2, arthrogryposis n = 1, double below knee amputation n = 1)</p>	<p>Age ranging from 13-18 years; AMC youth 18 years old</p>	<p>significantly larger for children with; parent of youth with disability is more present and less likely to let youth away from them; parental support in terms of recognition score was the same for both groups; environmental support score was higher for TDY</p> <p>Youth with physical disabilities experience longer parental dependence and autonomy begins in later adolescence; due to low skillset in sport at first there is greater feeling of requiring extrinsic motivation from coach or family, but as participation increases so do skills. More opportunities and welcoming environments benefit youth; -The importance of social interactions for youth extends beyond sport to reduce anxiety, shyness and peer victimization, can reduce bullying and promote positive sport experiences</p> <p>Revision of the PADS and further research needed to validate the scale. The amount and duration of physical activity</p>
<p>Ortiz-Castillo and Hodge (2011)</p>	<p>1) To validate selected components of the physical activity scale for individuals with physical disabilities for use with</p>	<p>PhD Thesis Dissertation</p>	<p>Adolescents with cerebral palsy, spina bifida, muscular dystrophy,</p>	<p>Age ranging from 12 to 18 years.</p> <p>Descriptive information about participants provided. 1: strenuous activity/exercise and walk/wheel/push outside</p>

(continued)

Table 3. Continued.

First Author (Year)	Study Aims	Study Design	Patient Population	Patient Age	Results	Author Conclusion
Shum (1982)	To find similarities and differences in social interactions of children with an without disabilities with their peers and in their contacts with adults in an integrated early childhood program.	PhD Thesis Dissertation	Children with disabilities (n = 40) (arthrogryposis, ataxic telangiectasias syndrome, CP, congenital dwarfism, OI, post-polio, spina bifida, n = 20) and non-disabled children (n = 20)	Age ranging from 3 to 6 years	both groups spent a greater proportion of social participation between groups of children with and without disabilities occurs 2) children with and without disabilities do engage in more social than nonsocial behaviors, 3) children socially interact with specific individuals over large groups 4) children initiate and interact more with their peers with disabilities responded to more adult contact initiated by the adult, children with disabilities	In an integrated early childhood setting 1) social interaction between groups of children with and without disabilities occurs 2) children with and without disabilities do engage in more social than nonsocial behaviors, 3) children socially interact with specific individuals over large groups 4) children initiate and interact more with their peers with disabilities responded to more adult contact initiated by the adult, children with disabilities
					participation of adolescents with physical disabilities does not meet well-established recommended physical activity guidelines.	participation of adolescents with physical disabilities does not meet well-established recommended physical activity guidelines.



				without disabilities in a reverse mainstream setting does not result in negative behavior of one group toward another 6) reverse mainstreaming is a workable model for social integration at the early childhood level
				responded more in peer contact, both groups initiated more contacts with peers with disabilities than responded, both groups favored the initiation direction of interaction with peers without disabilities, affective tone of behaviors was found to be identical for both groups
Wingate et al. (2005)	To examine children's attitudes and perceptions toward and involvement in the recreation of their siblings with disabilities	M.Sc Thesis Dissertation	Female children (n = 7) with siblings with disability	Age ranging from 10 to 16 years

PROM = passive range of motion, AROM = active range of motion, PSFS = patient-specific functional scale, PDMS = Peabody developmental motor scales, ASK = activities scale for kids, ADL = activities of daily living, ASKP38 = 38 items activities scale for kids performance, PODCI = pediatric outcomes data collection instrument, WRREX = Wilmington robotic exoskeleton arm, P-WREX = pediatric Wilmington robotic exoskeleton arm, TDY = typically developing youth, PEDi = pediatric evaluation of disability inventory, BMD = bone mineral density, WeeFIM = pediatric functional independence measure, FMS: functional mobility scale, PADs: physical activity determinants scale.

and their qualifications in relation to the topic. Accuracy refers to the reliability, truthfulness and accuracy of the presented information. Coverage questions whether there are clear parameters that define the context that is covered. Objectivity identifies any biases in the report. The date confirms the relevance of the information provided. Finally, significance is the value judgment in the context of the research area. Each article from the gray literature was evaluated and appraised for the different items of this tool by the principle author C.E.

Similarly, a data extraction form was created for the PAI, specifically, the YouTube videos and included: uniform resource locator (URL), video title, retrieval date, year posted, length of video, total views at the time of extraction, population posting video, observed contracture locations, adaptive aids used, activity type and location of participation based on the CAPE, and methodological quality using the CRAAP Evaluation Criteria. The CRAAP is a critical appraisal form developed for online sources and evaluates videos' Currency, Relevancy, Authority, Accuracy, and Purpose (Meriam Library, 2018). Currency refers to the timeliness of the information (i.e., when was the information published and how current is it). Relevancy refers to the importance of the information in relation to the topic that is being researched and whether the level of information is appropriate. Authority refers to the author of the work to determine if the information is to be trusted, while Accuracy refers to the level of evidence and trustworthiness of the source. Finally, Purpose refers to the reason for which the information was presented or published. Similar to the AACODS checklist, each video was evaluated on whether it met the requirement for different items of this checklist.

The data extraction forms were tested independently by two members of the research team (C.E and S.C) on a random sample of five articles and five videos and revised iteratively by the research team while completing the search. Differences in data charting were resolved by discussion, and remaining disagreements were resolved with involvement of the third reviewer (N.D-O). The three methodological quality rating tools (i.e., McMaster University's Critical Review, AACODS, CRAAP) were used to inform the strengths and limitations of this current body of evidence and guide future directions, but did not serve as selection criteria, as per the methodology suggested for scoping reviews (Arksey & O'Malley, 2005; Levac et al., 2010).

Collating, Summarizing, and Reporting Results

Once the data extraction was completed, it was synthesized according to three steps: (1) analyzing the data, (2) reporting the findings, and (3) discussing the implications (Arksey & O'Malley, 2005). The descriptive numerical analysis highlights the nature and distribution of the research articles and videos included. Secondly, the study purpose and major findings were the primary units of analysis.

Results

Empirical Studies and Gray Literature Articles

The empirical study and gray literature searches were conducted between January and November 2019 and generated a total of 1885 articles. Eleven empirical studies and six gray literature articles were included. Detailed information regarding included and

excluded literature can be found in the PRISMA flow chart in [Figure 1](#). [Table 3](#) provides information on characteristics of the included studies such as study aims, study design, patient population, results and conclusions. Study designs for the empirical data included four case reports, five retrospective cohort studies, one case-series, and one cross-sectional study, all published within the last ten years. The gray literature search found six dissertations published over a 50-year span. Nine of the 11 empirical studies focused solely on children and youth with AMC, whereas the three remaining empirical and all gray literature studies addressed children with different types of physical disabilities, including those with AMC. Participants included infants and toddlers, school-aged children, adolescents and young adults. As categorized by the CAPE, the most common activity type for the empirical studies was active-physical activities and for the gray literature it was active-physical, social, and skill-based activities. Empirical studies used a variety of standardized outcome measures. The most commonly used ones included the Alberta Infant Motor Scale (AIMS), the Activities Scale for Kids-performance (ASKp), the Pediatric Outcomes Data Collection Instrument (PODCI), the Pediatric Functional Independence Measure (Wee-FIM), as well as questionnaires developed specifically for individual studies.

In terms of the interventions provided to promote participation, occupational and physical therapy sessions including stretching, strengthening, and orthosis fabrication were the main methods reported. However, detailed descriptions on specific interventions, such as duration, frequency, and therapeutic modalities, were not provided. Five empirical studies described the use of specific assistive technology aiming at increasing participation in specific activities ([Babik et al., 2016](#); [Dalton & Hoyt-Hallett, 2013](#); [Lobo et al., 2016](#); [Pritchard-Wiart et al., 2019](#); [Babik et al., 2019](#)). Access to this assistive technology, feasibility of it, cost and ease of use all constituted facilitators to participation in play, social and academic settings. Barriers and facilitators to participation in this population were not always explicitly mentioned; nonetheless, the physical environment such as accessibility or available space was considered to have an impact on the ability to participate ([Beng Ting Lau, 2009](#); [Pritchard-Wiart et al., 2019](#)). The social environment, such as interactions with peers through organized sports, peer groups or integrated school settings ([Beng Ting Lau, 2009](#); [Larson, 1954](#); [Shum, 1982](#)) were the most important enablers. Families, including parents and siblings, and adults in general were also mentioned as potential factors affecting participation. Adults were presented as the ones providing the opportunity for the child to engage in different forms of participation through active involvement, support and encouragement, and modeling of healthy behaviors of participation ([Beng Ting Lau, 2009](#); [Larson, 1954](#); [Ortiz-Castillo & Hodge, 2011](#); [Wingate et al., 2005](#)). Finally, attitudes and behaviors of the youth themselves and of peers were also important factors ([Beng Ting Lau, 2009](#); [Ortiz-Castillo & Hodge, 2011](#)). [Table 4](#) summarizes extracted data about activity types, outcome measures used, interventions reported to promote participation levels, and facilitators and barriers to participation among youth with AMC. [Table 5](#) summarizes the methodological quality appraisal of the empirical studies. For the gray literature, the seven included articles met each category for the AACODS appraisal tool (i.e., Authority, Accuracy, Coverage, Objectivity, Date, and Significance).

Table 4. Description of participation extracted from empirical studies and gray literature.

	Author (Year)	Activity Type	Outcome Measures	Interventions	Barriers and Facilitators
Empirical Studies (n = 12)					
Amor et al. (2011)	Active-Physical	PODCI	Conservative treatment (no further details)	Conservative treatment has an overall positive impact on participation	
Azbell and Dannemiller (2015)	Active-Physical, Recreational, Social	ROM, MMT, AIMS, Bayley Scale of Infant and Toddler Development 3, PDMS2, PSFS, FLACC Pain scale	PT 1x/2weeks, OT on alternating weeks: stretching, splinting, serial casting, surgical management when necessary	None reported	
Babik et al. (2016)	Not enough information provided	Observations and video recording, AIMS, weight bearing, positioning, Strength, ROM	Exoskeleton 1 hour daily	Pediatric Wilmington exoskeleton arm (P-Wrex) as a feasible device	
Babik et al. (2019)	Recreational, Skill-Based	Observational based following evaluation guideline developed for the study	Children tested 2x/week with and without Playskin for floor and surface assessments	Playskin as an accessible device (low cost, simple materials), limitation in terms of minimum ROM required to benefit from device	
Bagley et al. (2011)	Active-physical, Recreational, Skill-Based	ASKp38	None reported	None reported	
Dalton and Hoyt-Hallett (2013)	Social, Self-Improvement	Observations	Adaptations made to wheelchair	Assistive technology as facilitators to occupational enablement and social interactions	
Dillon et al. (2009)	Not enough information provided	SAM, ASKp	Ambulatory activities of varied intensity and frequency (no further details)	None reported	
Ho and Karol (2008)	Active-Physical	ROM, FMS, PEDI, Wee-FIM, PODCI	None reported	None reported	
Lobo et al. (2016)	Not enough information provided	Observations, Questionnaire developed for this study	Playskin as inexpensive, easy to use and comfortable device		
Pritchard-Whart et al. (2019)	Recreational, Skill-Based	Quantitative data: family driving record (multiple categories) using 10-point Likert Scale qualitative data: Semi-structured interviews	Modified toy cars are feasible and accessible tools for mobility. Physical environment can limit mobility.		
Spencer et al. (2010)	Active-Physical	Bone densitometry, Wee-FIM, PODCI	None reported	None reported	
Beng Ting Lau (2009)	Skill-Based, Active-physical, Social	Questions developed for semi-structured interview or qualitative nature	Physical environment (space, access) can promote or hinder participation. Positive impact of social environment (interactions, opportunities, attitudes and		
Gray Literature (n = 7)					

Larson (1954)	Social, Self-Improvement	Questionnaire developed for this study	None reported
Orr and Arbour-Nicopoulos, (2016)	Active-physical, Skill-Based, Recreational	Interviews	None reported
Ortiz-Castillo and Hodge (2011)	Active-physical, Recreational	Physical Activity Scale for Individuals with Physical Disabilities, PADS Observational data	None reported
Shum (1982)	Social	Early childhood program	An integrated early childhood setting as a model for social integration
Wingate et al. (2005)	Active-physical, Social, Skill-Based	ATDP	Support and encouragement from siblings

ROM: Range of motion; MMT: manual muscle testing; AIMS: Alberta Infant Motor Scale; PDMS-2: Peabody Developmental Motor Scales-2; PSFS: Patient Specific Functional Scale; FLACC: Face Legs Activity Cry and Consolability; PT: physical therapy; OT: occupational therapy; ASQp38: Activities Scale for Kids-performance; PODCI: Pediatric Outcomes Data Collection Instrument; SAM: StepWatch3 Activity Monitor; FMS: Functional Mobility Scale; PEDi: Pediatric Evaluation of Disability Inventory; Wee-FIM: Pediatric Functional Independence Measure; AMs: Modified Active Movement Scale; AROM: active range of motion; PROM: passive range of motion; MAL: Motor Activity Log; CIT: Constraint-Induced-Movement therapy; ETCH: Evaluation Tool of Children's Handwriting; SFA: School Function Assessment; BOT: Bruininks-Oseretsky Test of Motor Proficiency; MAP: Miller Assessment for Preschoolers; QUEST: Quality of Upper Extremity Skills Test; PADS: Pain and Discomfort Scale; ATDP: Attitude Toward Disabled Persons Scale.

Table 5. McMaster University's critical review form for quantitative and qualitative studies.

Study (author, year)	McMaster University's Critical Review Form for Quantitative and Qualitative Studies						
	Study Purpose	Literature Review	Design	Sample & Justification	Outcomes	Intervention & Confounders	Results & Analysis & Drop-outs
n = 11							
Reliable							
Azbell and Dannemiller (2015)	X	X	X	X	X	X	X
Bagley et al. (2011)	X	X	X	X	X	X	X
Amor et al. (2011)	X	X	X	X	X	X	X
Babik et al. (2016)	X	X	X	X	X	X	X
Dillon et al. (2009)	X	X	X	X	X	X	X
Ho and Karol (2008)	X	X	X	X	X	X	X
Spencer et al. (2010)	X	X	X	X	X	X	X
Lobo et al. (2016)	X	X	X	X	X	X	X
Dalton and Hoyt-Hallert (2013)	X	X	X	X	X	X	X
Pritchard-Whart et al. (2019)	X	X	X	X	X	X	X
Babik et al. (2019)	X	X	X	X	X	X	X
Count	9	8	10	8	2	7	4
					2	8	8
						3	11

Note: X indicates the criteria was met.

Table 6. Information regarding participation on sport, art, and play related online videos.

Type of video	Participation location	Activity Type	Population posting video	Contracture location	CRAAP Appraisal
Sport related (n = 50)	Community only (n = 25) Home only (n = 11) School only (n = 3) Community & School (n = 2) Community & Home (n = 4) Home & School (n = 3) All three (n = 2)	Active-physical (n = 31) Recreational (n = 4) Self-improvement (n = 2) Skill-based (n = 10) Social (n = 8)	Network/Production (n = 15) Family member/caregiver (n = 21) Self (n = 6) University/Hospital (n = 5) Other (n = 3)	Knees, feet (n = 4) Shoulders, elbows, hands, knees, feet (n = 5) Shoulders, elbows, hands, hips, knees, feet (n = 10) Shoulders, elbows, hands (n = 8) Elbows, hands (n = 2) Hips, knees, feet (n = 6) Elbows, hands, hips, feet (n = 1) Hands, feet (n = 3) Shoulders, elbows, hands, feet (n = 1) Elbow, hands, knees, feet (n = 1) Spine, shoulders, elbows, hands, knees, feet (n = 1) Not observable (n = 8) Elbows, hands (n = 1) Shoulders, elbows, wrists, hips, knees, feet (n = 1) Shoulders, elbows, hands (n = 5) Wrists, hands (n = 1) Not observable (n = 1)	Currency (n = 47) Relevance (n = 49) Accuracy (n = 48) Authority (n = 16) Purpose (n = 16)
Art related (n = 9)	Community only (n = 3) Home only (n = 2) School only (n = 2) Community & School (n = 1) All three (n = 1)	Recreational (n = 2) Skill-based (n = 7)	Network/Production (n = 2) Family member/caregiver (n = 2) Self (n = 5)	Elbows, hands (n = 1) Hips, knees, feet (n = 1) Shoulders, elbows, hands, hips, knees, feet (n = 1) Shoulders, elbows, hands, hips, knees, feet (n = 2) Shoulders, elbows, hands (n = 6) Not observable (n = 1)	Currency (n = 9) Relevance (n = 8) Accuracy (n = 9) Authority (n = 1) Purpose (n = 4)
Play related (n = 12)	Community only (n = 6) Home only (n = 5) All three (n = 1)	Active-physical (n = 7) Recreational (n = 4) Self-improvement (n = 1) Skill-based (n = 2) Social (n = 2)	Network/Production (n = 1) Family member/caregiver (n = 8) Self (n = 3)	Elbows, hands (n = 1) Hips, knees, feet (n = 1) Shoulders, elbows, hands, hips, knees, feet (n = 1) Shoulders, elbows, hands, hips, knees, feet (n = 2) Shoulders, elbows, hands (n = 3)	Currency (n = 12) Relevance (n = 12) Accuracy (n = 12) Authority (n = 1) Purpose (n = 3)

Note: Some activities performed by the child encompasses different activity types (i.e. recreational and social), therefore the activity was counted in all activity types.

Publicly Available Information (YouTube Videos)

The PAI YouTube video search was conducted between May and July 2018 and yielded a total of 666 videos. Of these, 50/351 sport related, 9/266 art related and 12/49 play related videos met the inclusion criteria, therefore 71 videos were included in this scoping review. Participation for the PAI was categorized based on the CAPE description for activity type and location ([Table 5](#)). In regards to the sport related activities, children participated while using their manual wheelchair ($n = 14$), power wheelchair ($n = 1$), walker ($n = 1$), adapted tool for the sport itself ($n = 7$), and the remaining children did not use any adaptations ($n = 27$). In terms of the art related activities, only one child required a power wheelchair to participate. For the play related activities, children participated with no adapted aids ($n = 11$) except for one who used a stylus to play on a phone. Although not explicitly reported, these adaptive aids could potentially pose as facilitators to completing the activity observed in the videos. Based on the included videos, the most frequent participation location was in the community, the most common activity type was active-physical, and the most common individual posting the video was a family member or caregiver. Additionally, the authors (C.E and S.C) indicated the contracture location when possible to indicate the heterogeneity of the AMC population (i.e., various limb involvement and severity). Based on the CRAAP appraisal, the three types of videos (Sports, Art, and Play) all demonstrated a high value of Currency, Relevance, Accuracy and low use of Authority and Purpose. [Table 6](#) displays the participation details for the included videos as well as the video appraisal tool results.

Discussion

The overarching goal of this scoping review was to explore what is known on participation among children and youth with AMC. Based on the results of this scoping review, active-physical activities represented the most frequently reported activity type across empirical studies, gray literature and PAI. It has been demonstrated that regular physical activity promotes growth and development in children and youth ([Kohl & Cook, 2013](#)). Physical activity has benefits at every age, as it reduces the risk for heart disease, obesity, and metabolic syndrome; improves aerobic capacity, muscle and bone strength, flexibility; and reduces stress, anxiety, and depression ([Kohl & Cook, 2013](#)). Indeed, physical activity improves mood and self-esteem and helps children socialize and develop relationships with peers ([Baksjøberget et al., 2017](#); [Silva & Klatsky, 1985](#), [Orr & Arbour-Nicitopoulos, 2016](#)). Children with chronic illnesses are encouraged to accomplish physical activities provided they are modified to promote health and not exacerbate symptoms. However, it has been demonstrated that children with chronic conditions like cerebral palsy ([Lauruschkus et al., 2017](#)) or AMC ([Dillon et al., 2009](#)) spend more sedentary time and participate less in habitual physical activities than their peers without disabilities, which implies risks for health outcomes, physical function and metabolism. Indeed, Elfassy et al. ([2019](#)) reported that youth with AMC prioritized participating in active-physical areas, yet, access to extracurricular organized sports was very limited ([Elfassy et al., 2019](#)). Although the empirical studies and gray literature did not explicitly promote areas of participation, the YouTube videos provide the viewer with possibility to observe how children participate in physical activities that were meaningful to them.

The outcome measures used to evaluate the level of participation was only determined in the empirical studies and gray literature. As stated, the most frequently reported outcome measures included the Alberta Infant Motor Scale (AIMS), the Activities Scale for Kids-performance (ASKp), the Pediatric Outcomes Data Collection Instrument (PODCI), the Pediatric Functional Independence Measure (Wee-FIM). Although these outcome measures have been validated in the literature, they are not specifically designed to address participation but rather the body functions and structures, and activities domains of the ICF. Only the ASKp38 and the PODCI have participation subcategories: “play” in the ASKp38 (Bagley et al., 2011) and “sport and physical functioning” in the PODCI. According to Adair et al. (2018), participation encompasses several constructs such as attendance, involvement, competence and sense of self, and outcome measures may only quantify one or several of these constructs. Therefore, measures used for research on participation and in rehabilitation practice must be chosen according to the purpose of measurement and the construct of interest (Adair et al., 2018). Although participation specific measures do exist, none were utilized in the studies included in this scoping review.

In terms of intervention, most studies included in this scoping review used a “bottom up” type of approach to address separate components of a child’s skills (Babik et al., 2016; Brown & Chien, 2010). Therefore, most studies addressed participation indirectly through interventions focused more on body function. However, the empirical studies that addressed participation through the use of assistive technology (Babik et al., 2016; Dalton & Hoyt-Hallett, 2013; Lobo et al., 2016; Pritchard-Wiar et al., 2019; Babik et al., 2019) described concrete interventions and improvement in the child’s participation in different activity areas (e.g., play, social interactions, academic performance). Though not always clearly described, these interventions required practice within the child’s natural environment (e.g., home, school), addressed needs determined by the child or their parents, and was elaborated based on the child’s strength and abilities. A “top-down” approach takes a global perspective and focuses on the child’s participation in their living context to determine what is important and relevant to the child (Brown & Chien, 2010). Novak and Honan (2019) reported that “top-down” interventions confer larger clinical gains by implementing the child’s goals to optimize motivation and saliency of practice using practice in natural environments, intense repetitions to activate plasticity, and scaffold practice to the “just right challenge” to enable success (Novak & Honan, 2019). Therefore, the interventions provided to children and youth with AMC should implement a “top down” approach to intervene directly at the level of participation.

In regards to the barriers and facilitators of participation reported in the empirical and gray literature as well as the YouTube videos, one of the barriers was the physical environment. In a given situation, the activity itself may not be the issue as the child with AMC may have the physical capacity to complete the activity but the physical environment, such as accessibility and availability of space, is what restricts participation (Beng Ting Lau, 2009). The availability of activities in the community that can be adapted to a child with a disability may be a limiting factor as well (Orr & Arbour-Nicitopoulos, 2016). Access to adaptive aids or technology that are feasible and cost-effective is an important enabler for children with AMC to participate in different types of activities, as seen in empirical studies and in YouTube videos. The social

environment plays an important role in participation as children in less inclusive settings demonstrate lower levels of general health and are more likely to encounter difficulty interacting with peers (Anaby et al., 2019). Interacting with peers through team sports for example helps develop positive experiences, reduces anxiety or shyness, increase self-esteem and confidence, and reduces bullying (Orr & Arbour-Nicitopoulos, 2016). Only gray literature evaluated social interactions and their impact on children with physical disabilities, yet no studies have looked at interventions addressing social participation. Furthermore, institutional environments could pose a limitation as healthcare professionals reported that institutional factors such as budgets, reduced local resources, and time constraints often affects the care that goes above and beyond what is considered conservative treatment (Elfassy et al., 2019). Therefore, the environment, whether it be active-physical, social, or institutional, plays a large role in the participation levels of children and youth with AMC. Thus, considering the positive impact of participation on mental and physical health (Dahan-Oliel et al., 2012), and the limited number of studies pertaining to this topic in AMC, there is a need to further evaluate community specific environmental facilitators and barriers to participation. Based on such studies, interventions and areas for change or improvement at the individual, community and policy levels may be targeted. In addition, integrating new interventions such as the Pathways and Resources for Engagement and Participation (PREP) approach has the potential to improve health and well-being for children with AMC as well (Anaby et al., 2017). PREP is a strengths-based intervention aimed at enhancing participation in chosen leisure activities by removing environmental barriers and coaching youths and their parents.

When comparing empirical studies and gray literature to the YouTube videos, accessibility of information regarding participation takes an important role as YouTube allows for a greater number of people to access and view videos of other children performing the activity. Therefore, key stakeholders, such as children and youth with AMC, their caregivers, as well as local clinicians can observe how other children participate, the environment in which they participate, and the adaptations that could be required in order to promote participation. Finally, the involvement of family members as active team players working alongside healthcare professionals have been shown to be beneficial to the child (Anaby et al., 2019). The importance of families' involvement is demonstrated in the YouTube videos and highlights the central role caregivers play in promoting participation among youth living with AMC. Through their support and active involvement, families provide the opportunity for participation and encourage children in developing their skills outside of the home setting (Beng Ting Lau, 2009; Larson, 1954; Ortiz-Castillo & Hodge, 2011; Wingate et al., 2005).

Limitations of the Scoping Review

Although the search strategy for empirical and gray literature was in English, the review itself was not limited to articles published in English; however, articles that were not in English or French were excluded. Furthermore, on-going and non-disseminated studies were also not included in the selection criteria. Finally, the YouTube videos were extracted on a single day in March 2018. Since YouTube is a social media platform,

multiple videos meeting our eligibility criteria could have been uploaded daily. However, these were not considered after the extraction date in 2018 for feasibility purposes. As the number of videos included in this scoping review was significant, including additional videos would likely not affect the current findings.

Future Studies

Future studies regarding participation with children with AMC should use validated tools to evaluate participation while targeting specific participation constructs, and should evaluate effectiveness of “top-down” interventions with this population. Studies pertaining to barriers and facilitators to participation may help guide interventions and changes at the level of the individual and of the community.

Conclusion

The paucity of research found in the empirical studies and gray literature indicates a need for future studies in the field of participation and rehabilitation for this population. However, new and innovative ways of accessing PAI can be found on online platforms such as YouTube, where clinicians as well as youth and families can become familiar with methods in which to participate in their natural environment. As there is lack of knowledge on specific interventions to promote participation for children with AMC, clinicians should follow general principles promoted in childhood disability. These interventions should be top-down, activity based and include parent partnerships. Principles described in the PREP approach should be used to address potential barriers and ensure the discrepancy between the child’s capacity and motivation with the environment is minimized.

Disclosure Statement

No potential conflict of interest was reported by the author(s).

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