

Quality of life and leisure participation in children with neurodevelopmental disabilities: a thematic analysis of the literature

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Abstract

Purpose The aim of this systematic review was to document evidence of the association between leisure participation and quality of life (QoL) in children with neurodevelopmental disabilities, and to identify the main factors that further clarify this relationship.

Methods An electronic search of the literature was conducted using Medline, Embase, PsycInfo, CINAHL, ERIC, OT Seeker, and the Cochrane library using relevant MESH heading and key words. An inductive thematic analysis was used to synthesize main findings from the studies. The mixed methods appraisal tool (MMAT) was used to assess the methodological quality of the included studies.

Results Nineteen studies were included in this systematic review. All 19 studies fulfilled most of the criteria outlined by the MMAT for each study design (quantitative, qualitative, and mixed methods). Mixed methods studies had the lowest MMAT scores. Seven themes were identified that further elucidate the important relationships between leisure participation and QoL: active physical leisure participation and physical well-being; leisure participation and self; leisure participation and emotional well-being; leisure participation and social well-being; leisure participation and cognition; leisure preferences and well-being; and negative aspects of leisure participation.

Conclusions The use of thematic analysis enabled the synthesis of findings from quantitative, qualitative, and mixed methods study designs. Participating in both naturally occurring, spontaneous forms of leisure activities and participating in more structured types of leisure and therapeutic programs that focus on leisure contribute to improving the QoL of children with neurodevelopmental disabilities. A key aspect to consider is the child's ability to participate in his/her preferred leisure activities. Environmental adaptations may be required to ensure that the leisure opportunities meet the children's capabilities and contribute positively to their QoL.

Keywords Quality of life · Participation · Leisure · Physical activity · Neurodevelopmental disability · Childhood disability · Mixed methods · Thematic analysis

Introduction

Neurodevelopmental disabilities refer to a diverse group of conditions and disorders that begin in the early years of children's lives and influence their development, often for life. The traditional focus of health care services for this population has been primarily directed on rehabilitation interventions that address the underlying developmental impairments, such as abnormal muscle tone, decreased attention span, poor dexterity, or difficulties with perceptual concepts. Only recently, intervention approaches have demonstrated a shift toward promoting independence in everyday activities and participation in different life areas, including leisure [1]. Families place more value on achieving goals related to activity and participation than on impairment-based goals that are typically reflected in standardized developmental measures [2].

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Leisure participation is an important component of the World Health Organization's International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) [3]. Engaging in leisure activities may be influenced by the child's personal factors, environmental factors, and their health condition [3, 4]. By participating in leisure activities, children develop competencies, achieve mental and physical health, gain an understanding of their strengths and abilities, and form friendships and relationships. It is by participating that children and adolescents make contributions to their community, learn about the expectations of society, and develop the skills needed to become successful in their home, school, and community [4, 5]. Participation in leisure activities includes participation in sports, arts, entertainment, social, self-improvement, and religious activities [5]. Participation is increasingly considered as one of the ultimate aims of pediatric rehabilitation and is believed to contribute to child health, development, and quality of life [5, 6].

Another important concept to consider in rehabilitation is quality of life (QoL). QoL is a broad concept encompassing many components of overall health and well-being (e.g., physical, psychosocial, economic, and cultural) [7]. It is influenced by the context of the culture and value systems and relates to the individual's goals, expectations, standards, and concerns [7]. The QoL of children and adolescents who live with a neurological condition can be impacted at different levels, including physical (e.g., physical health, independence in basic functional activities), psychological (e.g., mental status, positive self-perception), and psychosocial dimensions (e.g., forming friendships, leisure time, finding a partner) [8]. Identifying the factors that are associated with better or worse QoL is important [9] so as to guide program planning and allocation of resources, thus contributing to optimizing the well-being of these youth and young adults. Findings from several studies have challenged the assumption that individuals with disabilities have a lower QoL and a diminished perception of their well-being and general satisfaction with life [10, 11]. Recent studies are exploring a variety of factors that may contribute to a good QoL in children with neurodevelopmental disabilities; however, most studies explore aspects related to developmental impairments and activity limitations such as motor functioning. Recent studies exploring level of participation in active physical and other leisure activities in children and youth with disabilities justify the importance of participation by its potential impact on QoL. Although frequently assumed, the relationship between leisure participation and QoL has not been clearly delineated [12].

The aim of this systematic review was to document evidence of the association between leisure participation and QoL in children with neurodevelopmental disabilities,

and to identify the main themes/factors that further clarify this relationship. More specifically, we aim at understanding the phenomena of the influence of participation in specific leisure and social activities on the different dimensions of QoL.

Methods

Search strategy and selection criteria

Eligible articles published in journals were identified through the following electronic databases: Medline via Ovid (1950–2011), Embase via Ovid (1980–2011), PsycInfo via Ovid (1987–2011), CINAHL via EBSCOhost (1990–2011), ERIC, OT Seeker, and the Cochrane library using the MeSH-term and/or text word search or combinations of: (1) for participation: participation; social participation; leisure; leisure activities; recreation; recreational activities; sports; physical activities; fitness; active physical activities; motor activity; physical fitness; exercise. (2) for QoL: quality of life (QoL); health-related quality of life (HRQL or HRQOL); life quality; health status; well-being. The search was then restricted to children between 0 and 19 years. The diagnostic criteria included central nervous disorders of the brain that result in developmental disabilities. Studies on asthma, diabetes, cancer, and obesity were excluded from the search, as the primary etiology of the developmental disability was non-neurologic. In addition, studies on children who are typically developing were not retained. The QoL terms were based on a search strategy established by the University of York Centres for Review and Dissemination [13]. The search was limited to original articles published in English.

Duplicates were removed. Abstracts, editorials, commentaries, review articles, letters, and study protocols were excluded but reviewed for possible references. Abstracts of potential articles were reviewed, and studies that were not focused on children (0–19 years) with neurodevelopmental disabilities, did not provide results on leisure participation and QoL, and did not look at an association between these two constructs were excluded. In the case of studies reporting on children and older individuals, authors were contacted to check if separate data on children 0–19 years was available. Similarly, authors were contacted when data were provided on both participation and QoL, but when the association between these two constructs was not provided. Inclusion criteria are displayed in Table 1. Full-text articles of the selected abstracts were sought and reviewed according to the inclusion criteria. To be included, studies had to use either a quantitative, qualitative, or mixed methods (quantitative and qualitative) methodology. Additionally, reference lists from original papers were

Table 1 Inclusion criteria

| Inclusion criteria | Inclusion criteria |
|--|--|
| Population | >50% of sampled population are under 19 years Neurodevelopmental disability |
| Research design | Quantitative Qualitative Mixed methods |
| Participation | In any leisure activity: sports, arts, entertainment, social, self-improvement, and religious activities Measured using a quantitative tool Measured using interview Measured using both of the above approaches |
| Quality of life measurement | Measured using a quantitative tool Measured using interview Measured using both of the above approaches |
| Relationship between participation and quality of life | Statistically explored in quantitative or mixed methods studies Inferred in qualitative studies |

reviewed. Google scholar was used to identify any further relevant articles.

Data extraction and synthesis

Data on the study design, characteristics of the sample (sample size, age and etiology), measurement of leisure participation and QoL, and findings of included studies were extracted by two reviewers into data extraction forms. Studies that used both a quantitative and a qualitative methodology were classified as mixed methods. For quantitative and mixed methods studies, only data from outcome measures directly measuring QoL were considered. This precluded a formal meta-analysis as the assessment methods, and the presentation of the results were heterogeneous. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist and flowchart [14] were used to verify that all aspects of the review were considered.

We used an inductive thematic analysis for synthesis of the relationships between leisure participation and QoL. Thematic analysis reflects a qualitative methodology that is appropriate for literature reviews as it provides a qualitative synthesis of original qualitative, quantitative, and/or mixed-methods research studies through the extraction of themes and sub-themes [15, 16]. Themes are built from the text of selected studies after quality appraisal is first achieved. This appraisal is performed in an inductive manner and is data driven by definition, meaning that the authors did not try to infer their own conceptions or previously established theories, but rather explored the richness of data as it is presented [16]. Thematic analysis is a valuable methodology to explore studies with different designs, as it is able to capture common themes

irrespective of the nature of the study. The results sections of all selected studies were independently manually coded to generate initial categories that organize the data into meaningful themes. These categories were then reviewed to consolidate key themes that recurred across the data by consensus. The themes represent the primary leisure activity types that are related to specific dimensions of QoL.

Quality assessment

The mixed methods appraisal tool (MMAT—Version 2011) was used for quality assessment of the included studies [17, 18]. The methodological quality criteria of the MMAT are illustrated in Table 2. There is no consensus on the criteria to be used for the concomitant critical appraisal of the methodological quality of studies retained for synthesis in the review of qualitative, quantitative, and mixed methods studies. However, the MMAT is a recently developed tool that has demonstrated an intra-class correlation of 0.8 based on pilot testing in 2009, its use is effective and practical for the quality assessment of a mixed methods review. There are four criteria for appraising quantitative and qualitative studies. For a mixed methods study, both the appropriate section for the quantitative component and the qualitative component are used. Scores vary from 25%—one criterion met, to 100%—all criteria met. For qualitative and quantitative studies, this score is the number of criteria met divided by four. For mixed methods studies, the overall quality score is the lowest score of the quantitative and qualitative study components. Quality assessment scores using the MMAT were calculated for the studies included in this review.

Table 2 Quality rating

| Study designs | Methodological quality criteria |
|--|--|
| 1. Qualitative | 1.1. Are the sources of qualitative data (archives, documents, informants, observations) relevant to address the research question (objective)? 1.2. Is the process for analyzing qualitative data relevant to address the research question (objective)? 1.3. Is appropriate consideration given to how findings relate to the context, e.g., the setting, in which the data were collected? 1.4. Is appropriate consideration given to how findings relate to researchers' influence, e.g., through their interactions with participants? |
| 2. Quantitative randomized controlled (trials) | 2.1. Is there a clear description of the randomization (or an appropriate sequence generation)? 2.2. Is there a clear description of the allocation concealment (or blinding when applicable)? 2.3. Are there complete outcome data (80% or above)? 2.4. Is there low withdrawal/dropout (below 20%)? |
| 3. Quantitative non-randomized | 3.1. Are participants (organizations) recruited in a way that minimizes selection bias? 3.2. Are measurements appropriate (clear origin, or validity known, or standard instrument; and absence of contamination between groups when appropriate) regarding the exposure/intervention and outcomes? 3.3. In the groups being compared (exposed vs. non-exposed; with intervention vs. without; cases vs. controls), are the participants comparable, or do researchers take into account (control for) the difference between these groups? 3.4. Are there complete outcome data (80% or above), and, when applicable, an acceptable response rate (60% or above), or an acceptable follow-up rate for cohort studies (depending on the duration of follow-up)? |
| 4. Quantitative descriptive | 4.1. Is the sampling strategy relevant to address the quantitative research question (quantitative aspect of the mixed methods question)? 4.2. Is the sample representative of the population under study? 4.3. Are measurements appropriate (clear origin, or validity known, or standard instrument)? 4.4. Is there an acceptable response rate (60% or above)? |
| 5. Mixed methods | 5.1. Is the mixed methods research design relevant to address the qualitative and quantitative research questions (or objectives), or the qualitative and quantitative aspects of the mixed methods question (or objective)? 5.2. Is the integration of qualitative and quantitative data (or results*) relevant to address the research question (objective)? 5.3. Is appropriate consideration given to the limitations associated with this integration, e.g., the divergence of qualitative and quantitative data (or results*) in a triangulation design? <i>Criteria for the qualitative component (1.1 to 1.4), and appropriate criteria for the quantitative component (2.1 to 2.4, or 3.1 to 3.4, or 4.1 to 4.4), must be also applied</i> |

Mixed methods appraisal tool (MMAT) criteria

* These two items are not considered as double-barreled items since in mixed methods research, (1) there may be research questions (quantitative research) or research objectives (qualitative research), and (2) data may be integrated, and/or qualitative findings and quantitative results can be integrated

Results

Search strategy and selection criteria

A total of 1,539 references were imported into Endnote X2 reference manager. The titles and abstracts of these references were reviewed, and 224 citations were retained as pertaining specifically to the relationship between leisure participation and QoL. Titles and abstracts of the 224 identified articles were reviewed, and 52 were selected based on selection criteria. The full-text articles of these 52 studies were retrieved and reviewed using the inclusion and exclusion criteria. Of these, 19 studies met selection

criteria and were included for review in this paper. The identification and selection process is outlined in a flow chart (Fig. 1). All included studies were in full accordance with ethical principles.

Data extraction and synthesis

An overview of the methods of these 19 studies is presented in Table 3: nine were quantitative studies (RCT, cross-sectional) [19–27], six were qualitative studies [28–33], and four were mixed methods studies (case-studies with interview, RCT with interview, cross-sectional with interview) [34–37]. In terms of diagnostic criteria, nine studies included

Table 3 Findings of the included studies

| Author, year, country | Methodology | Age range/ mean | Study group | Control group | Outcome measure/type of report/intervention | | Findings |
|---------------------------------|-----------------|---------------------------------|--|---|--|---|---|
| | | | | | Participation | Quality of life | |
| Quantitative studies | | | | | | | |
| Bjornson, 2008, USA [22] | Cross-sectional | 10–13 year old/ 11.8 year | 81 children with CP (GMFCS levels I–III) | 30 children with typical development/ mean age 11.9 year | -Stepwatch accelerometer -ASK self-report | -CHQ (self-report) -YQOL -R (self-report) | Activity performance (accelerometry) was positively associated with the physical functioning and role/social behavior domains of the CHQ but not with YQOL-R. ASK scores were positively correlated with the physical, behavioral, and emotional domains of the CHQ |
| Buffart, 2009, Netherlands [20] | Cross-sectional | 16–30 year old/ 21.1 year | 51 children with myelomeningocele | – | Physical activity using an accelerometer on 2 consecutive days | SF-36 (self-report) | Adjusted for sex and ambulatory status, individuals with higher physical activity levels perceived a higher physical HRQL ($P = 0.02$). No association with mental HRQL |
| Conant, 2008, USA [23] | Cross-sectional | 8–16 year old/ 10.6 year | 9 children with epilepsy | – | Karate program: 1 h/week for 10 weeks | QOLCE (parent-report) | The memory subscale of the QOLCE increased $P = 0.01$ following the karate program. There was a trend for improved QoL in all other domains |
| Gorzowski, 2010, USA [21] | Cross-sectional | 7–17 year old/ 12.5 year | 97 girls with SCI | – | CAPE | PedsQL | The relationship between participation and QoL was mediated by depression; a greater social participation context was associated with decreased depression, which was then associated with greater QoL |
| Majnemer, 2011, Canada [27] | Cross-sectional | 5.8–12.9 years old/ 9.7 year | 63 children CP GMFCS I–V | – | CAPE self-report with parental assistance | PedsQL (parent- and self-reports) | Participation in active physical activities is related with physical well-being ($r = .34$) |
| McManus, 2008, Ireland [26] | Cross-sectional | 8–12 year old | 98 children with CP (GMFCS levels I–V) | 448 children from regular schools | FPQ (parent-report) | Kidscreen (parent-report) | Participation in everyday activities had an effect on physical well-being ($B = 7.8$), moods and emotions ($B = 4.5$), and support and peers ($B = 13.9$) |
| Odame, 2006, Canada [24] | Cross-sectional | 4–23 year old/ 15.6 year | 25 children with a childhood brain tumor | – | Modified Baecke questionnaire on physical activity (parent-report) | HUI2 and HUI3 (parent-report) | Sports scores showed statistically significant correlations ($P < 0.05$) with both HUI2 and HUI3 scores for overall HRQL |
| Pirpiris, 2006, USA [25] | Cross-sectional | 8–16 year old/ 10.2 year | 90 children with CP (GMFCS levels I–III) | – | PODCI (parent-report) | PedsQL (parent- and self-reports) | There was a modest correlation between PODCI and PedsQL domain scores. The sports and physical function domain of the PODCI correlated significantly ($P < 0.01$) with the child-report QoL, physical and psychosocial scores |

Table 3 continued

| Author, year, country | Methodology | Age range/ mean | Study group | Control group | Outcome measure/type of report/intervention | | Findings |
|---|--------------------------------------|-----------------------------|--|---|---|--|---|
| | | | | | Participation | Quality of life | |
| Verschuren, 2007 Netherlands [19] | RCT | 7–20 year old/11.6 year | 32 children CP GMFCS levels I and II | 33 children CP GMFCS levels I and II | CAPE (parent-report)/ training program 45 min twice/week for 8 months | TNO-AZL TACQOL (parent-report) | Training group had significantly better scores on the motor, autonomy, cognition and social domains at the end of the 4 training months, and in the autonomy domain after the last 4 training months. No differences between groups were found 4 months after the training period |
| Qualitative studies | | | | | | | |
| Davis, 2009, Australia [31] | Grounded theory | 13–18 year old/15.5 year | 17 adolescents CP GMFCS I–V and 23 parents | – | Semi-structured interviews | Semi-structured interviews | Participating and trying new things is important for QoL. Adjustments such as eliminating barriers may have to be done to engage in activities and promote leisure opportunities |
| Mandich, 2003, Canada [28] | In-depth interviews | 7–12 year old/8.3 year | 12 parents of 10 children with DCD | – | In-depth interview (parent- report) | In-depth interview (parent-report) | Findings revealed that incompetence in everyday activities had serious negative effects for the children |
| McBurney, 2003, Australia [32] | Thematic coding | 8–18 year old/ 12.8 year | 11 adolescents with CP (GMFCS levels I–III) and their parents | – | Individual semi-structured interviews with parents and child separately | Individual semi- structured interviews with parents and child separately | Participation in a strength-training program was beneficial for overall psychological functioning |
| Shikako- Thomas, 2009, Canada [29] | Constructivist grounded theory | 12–16 year old | 12 adolescents CP GMFCS I–V | – | Semi-structured interviews | Semi-structured interviews | Participation in a variety of activities, especially leisure activities of their own choosing, was emphasized by almost all teens as contributing to QoL |
| Ronen, 1999, Canada [33] | Textual analysis | 6–10 years old/ 9.2 year | 29 children with epilepsy and 42 parents | – | Separate focus groups for children and parents | Separate focus groups for children and parents | Participation in leisure contributed to a sense of life fulfillment and time use |
| Young, 2007, UK [30] | Constant comparative method | 8–13 year old/ 10.8 year | 28 children with CP (GMFCS levels I– V) and 35 parents | – | Interview | Interview | Four themes emerged: 'social relationships', 'home and school environment', self and body', 'recreational activities and resources'. Physical activities were found to be important, and the issues of interest and safety were mentioned |
| Mixed methods studies | | | | | | | |
| Becker, 2010, USA [35] | Case study | 11 year old | 1 child with Down syndrome | – | Performing arts program 90 min once/week for 14 weeks | PedsQL/ interview (parent- report) | QoL total scores increased 51–57. Parent reported pride, happiness, improved coordination, speed, endurance, confidence, information retention, getting along with peers, keeping up with school work, decreased loss of balance and anxiety |

Table 3 continued

| Author, year, country | Methodology | Age range/ mean | Study group | Control group | Outcome measure/type of report/intervention | | Findings |
|-------------------------------|---------------------------|---------------------------|--|--|---|--|--|
| | | | | | Participation | Quality of life | |
| Davis, 2009, Australia [36] | RCT/ interview | 4–12 year/ 7.4 year | 50 children with intervention with CP (GMFCS levels I-III) | 49 children with CP (GMFCS levels I-III) | Riding program 30–40 min once a week for 10 weeks | CP-QOL and Kidscreen (parent- and self-reports), CHQ (parent-report) | Slight increase in parent-reported global HRQL Kidscreen ($P < 0.05$) in intervention group versus the control group, and in family cohesion in CHQ parent-report ($P = 0.05$) |
| Fraas, 2010, USA [34] | Case study | 14 year old | 1 child with acquired brain injury (Eastern Equine Encephalitis) | – | Mentoring program 5.5 h/ week for 10 weeks | YQOL-R and weekly interviews (self-report) | Higher QoL in each of the 4 domains (self, relationship, environment, general QoL) was observed after the mentoring program. The teen's perceived benefit from participation on QoL was rated as 'a lot' |
| Poulsen, 2008, Australia [37] | Cross-sectional/ diary | 10–13 years old/11.6 year | 60 boys with DCD | 113 boys who do not have DCD Mean age 11.8 year | LDB-S (self-report), seven-day leisure-time diary and retrospective 12-month leisure survey (parent-report) | SLSS and LSDQ (self-report) | Team sport participation has a small positive effect size on life satisfaction (.27) and negative effect on loneliness (–.2) |

ASK activity scale for kids, CAPE children's assessment of participation and enjoyment, CHQ child health questionnaire, CP-QOL cerebral palsy quality of life questionnaire for children, DCD developmental coordination disorder, FPQ frequency of participation, GMFCS gross motor function classification system, HUI2 health utilities index mark 2, HUI3 health utilities index mark 3, ICF-CY WHO international classification of functioning, disability and health, LDB-S leisure diagnostic battery-short form, LSDQ loneliness and social dissatisfaction questionnaire, PedsQL pediatric quality of life inventory, PODCI pediatric outcomes data collection instrument, QOLCE quality of life in childhood epilepsy, SCI spinal cord injury, SLSS students' life satisfaction scale, TNO-AZL TACQOL TNO-AZL questionnaire for children's health-related quality of life, YQOL-R youth quality of life-research version

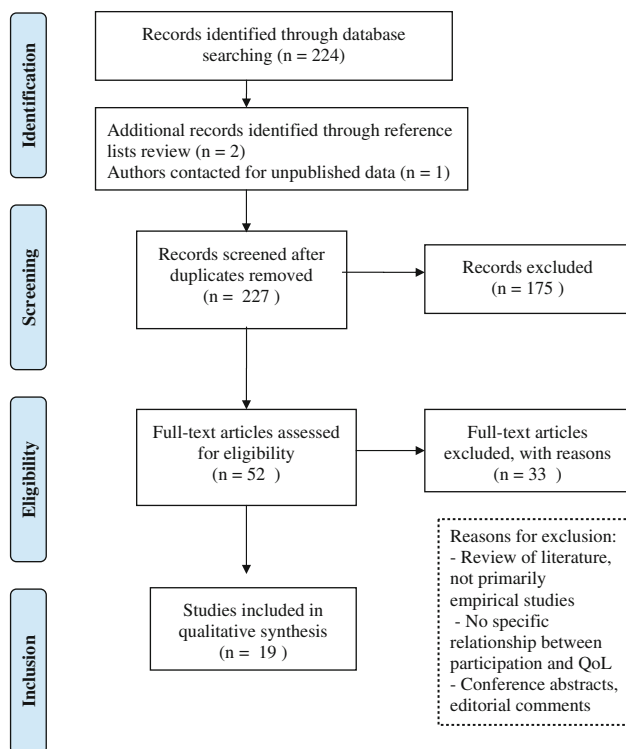


Fig. 1 Flow chart on different phases of a systematic review

children with cerebral palsy (CP), two had children with developmental coordination disorder (DCD), two with epilepsy, and one study each with children with acquired brain injury, myelomeningocele, Down syndrome, spinal cord injury (SCI), and brain tumor. All studies included both boys and girls except for two studies, one of which included only girls with SCI [21] and one which included only boys with DCD [37]. The main findings are also summarized in Table 3. The two randomized clinical trials [19, 36] evaluated the effects of participating in a leisure activity program (exercise training and horseback riding) on QoL in children with CP. Three studies [23, 24, 35] explored the effect of participating in social or structured leisure activities (e.g., karate, performing arts program) on QoL. Another eight studies [19, 21, 22, 24, 25, 26, 27, 37] measured the association between participation and QoL using standardized tools to assess both constructs. These studies used different tools to measure QoL and included generic and disease-specific QoL measures. Leisure participation was most often measured through self-report or parent-report questionnaires, but also with objective physical activity measures such as accelerometry. These studies found a positive relationship between greater participation scores and enhanced QoL. One of these studies [21] found an indirect relationship between leisure participation and QoL that was mediated by depression. All six qualitative studies [28–33] indicated that both children with neurodevelopmental disabilities and their

parents emphasized the importance of participating in everyday leisure activities in the home, school, and community on promoting well-being and a better QoL. Similarly, the respondents felt that a lack of participation or constraints to leisure participation resulted in poorer QoL.

All the texts retained were read independently by two authors (N.D-O., K.S-T.), and initial themes that related to the association between participation in leisure and QoL were identified. A face-to-face session was then held to discuss the themes identified and reach consensus on the themes that better represented the findings. The texts were then read independently again for identification of the new themes. Any new theme that emerged from the second analysis was discussed in a final consensus meeting. Seven identified themes were selected that represented an account for the phenomena of the influence of leisure participation on the QoL of children and youth with neurodevelopmental disabilities. Themes were explored to help elucidate the objective experiences that contribute to the subjective dimensions of well-being and the overall meaning that children with neurodevelopmental disabilities and their families can attribute to these phenomena [16].

Theme 1: active physical leisure participation and physical well-being

One study reported that engaging in daily physical activities such as running errands and joining organized activities outside school was positively associated with better QoL in the physical health and well-being domains of the Kidscreen [26]. Several studies remarked that both intensity and diversity of participation in different physical activities were linked with physical well-being [19, 20, 25–28, 35, 36]. Specifically, engagement in active physical activities such as bicycling and doing water sports, playing sports and other games [26, 27], doing aerobic exercises [19], and horseback riding [36] were positively related with feeling well physically. The subjective understanding of physical well-being encompassed aspects such as better endurance or ability to do certain motor tasks [35]. These findings indicate that participating in active physical leisure activities contributed to physical well-being.

Theme 2: leisure participation and self

Participating in a performing arts program, horseback riding, riding a bike and participating in a mentoring program were associated with an increased sense of self-perception, self-esteem, and self-competence, but also a sense of being treated with fairness and being respected [23, 28, 30, 35, 36]. A number of studies suggested mediators in this process such as the inclusion of friends during engagement in leisure contributing to increased self-confidence and a

sense of pride [35, 36]. Furthermore, participation in adapted activities was viewed as positive for self-competence as adaptations enhanced the possibility of successes and mastery [29, 31]. Participation in adapted activities also contributed to the feeling of identity and perceiving the disability as something positive [29]. Therefore, participating in both naturally occurring forms of leisure activities (such as riding a bike) and participating in therapeutic programs of leisure activities (such as horseback riding) contributed to improvement of the sense of self in children with neurodevelopmental disabilities.

Theme 3: leisure participation and emotional well-being

In several reports, being part of activities contributed to the emotional well-being of children with disabilities, allowing them to experience greater happiness and enjoyment in life [29, 33, 35, 36]. In general, the engagement in recreation promoted a sense of fulfillment [33] and made it possible for the children to experience success and realization of dreams that they would not otherwise experience [28, 29]. Participation in everyday activities was associated with the QoL domains of moods and emotions [26]. Other specific activities that had an emotional benefit were as follow: (a) a strength-training program was associated with psychological well-being [32], (b) sedentary activities were valued for fostering friendships [30], (c) horseback riding facilitated relaxation [36], (d) participating in a performing arts program helped to control anxiety [35], and (e) social and vocational participation was associated with fewer depressive symptoms, which in turn was associated with better QoL [21]. Another important aspect of emotional well-being, especially relevant for adolescents, is autonomy and independence. For example, children who participated in aerobic and anaerobic exercise realized benefits in the autonomy domain of the TNO-AZL Children's Quality of Life Questionnaire Parent Form [19]. Parents reported that participation in a horseback riding program contributed to the child's growth and independence [35], whereas adolescents indicated that being able to visit with their friends on their own also contributed to feelings of autonomy [30]. One study found an association between active physical leisure participation and psychosocial well-being [27]. Participating in a strength-training program was also perceived as positive for psychological well-being [32]. Another study did not support the association between physical activities measured by accelerometry and mental well-being [20]. Participation was both influenced by and had an influence on motivation. When children attempted to achieve an activity that was challenging and then finally mastered it, they felt motivated to try even harder [28, 32] and those achievements in turn contributed to a better QoL. Overall, most studies found that participating in therapeutic

leisure programs (performing arts program, strength-training program) or in naturally occurring leisure activities (sedentary activities) contributed to emotional well-being.

Theme 4: leisure participation and social well-being

Socialization and social well-being were key aspects of general QoL. Participation in recreational and social activities was felt to be a way for children to experience different situations with peers, to negotiate roles, to practice social skills, and to integrate the different life contexts [26, 30, 34, 35]. Participation in everyday activities was associated with QoL, in the social support and peers domains [26], and likely allowed for children to spend more time with peers and develop friendships. In another study, a mentoring program promoted QoL with regard to social relationships [19]. Participation in an arts program helped children to take initiatives for peer interaction and enabled them to keep up with other children in the playground [35]. It is interesting to note that participation in both integrated and segregated activities was perceived as beneficial to adolescents with CP [29]. One study [29] found that while activities with peers without disabilities contributed to feelings of acceptance and skill development, segregated activities contributed to relaxation and a sense of belonging to the group of children with disabilities. This indicates that both leisure participation arising from therapeutic rehabilitation programs and naturally occurring in the child's environment can promote involvement with children with and without disabilities. These opportunities can contribute to children's ability to navigate their social environment, strengthen significant relationships, and promote a general sense of social well-being.

Theme 5: leisure participation and cognition

Although cognitive skills can be understood as an aspect of general development and function rather than a QoL component, some QoL measures include cognition as a dimension of QoL. Participation in aerobic exercises, performing arts program and karate were beneficial for cognition, and specific skills such as memory were collectively perceived as positive for the QoL of participants [19, 23, 35]. Therefore, participating in therapeutic leisure programs may contribute to improved cognition, which in turn may lead to better QoL.

Theme 6: leisure preferences and well-being

The ability to engage in activities of their choosing was considered an important aspect that can improve QoL for children and adolescents with neurodevelopmental disabilities. The ability to participate in preferred leisure activities and to pursue their own interests helped these

children feel better about themselves and is perceived by parents and children alike as being one of the key elements contributing to a better QoL [28–30, 36].

Theme 7: negative aspects of leisure participation

Although leisure participation has several positive associations and effects on QoL, it may also have negative effects. The negative influence of engagement in leisure, however, is likely mediated by other factors, especially the interaction of physical limitations and environmental barriers. Children experienced frustration in both organized sports and less structured leisure pursuits such as playing in the playground or playing tag, especially when they encountered limitations and were not able to perform like peers [28, 30, 36]. Another negative attribute of leisure participation was when the equipment and environment were not adapted to accommodate the children's needs, thus preventing them from engaging in valued activities, or keeping up with their peers, generating feelings of fear, isolation and boredom [28–30]. Furthermore, when children faced financial constraints that prevented them from accessing recreational programs or adapted leisure activities, this had negative repercussions on their well-being [30]. We can infer that contextual factors can play an important role in the relationship between leisure and QoL.

To summarize, participating in both naturally occurring, spontaneous forms of leisure activities and participating in more structured types of leisure and therapeutic rehabilitation programs that focus on leisure contribute to improving the QoL of children with neurodevelopmental disabilities. A key aspect to consider is the child's ability to participate in his/her preferred leisure activities. Environmental adaptations may be required to ensure that the leisure opportunities meet the children's capabilities and desires and thus contribute to their QoL.

Quality assessment

The quality appraisal of selected studies is presented in Table 4. Overall, all 19 studies fulfilled most of the criteria outlined by the MMAT for each study design. Mixed methods studies had the lowest MMAT scores. Interestingly, none of the four mixed methods studies were identified as such by the authors. However, since they used both quantitative and qualitative methodology, they were classified as mixed methods in this review.

Discussion

This systematic review aimed to capture the association between participation in leisure activities and QoL in

Table 4 Mixed methods appraisal tool (MMAT) scores for included studies

| Study | MMAT score |
|------------------------------------|------------|
| Quantitative studies | |
| Bjornson, 2008, USA [22] | *** |
| Buffart, 2009, Netherlands [20] | **** |
| Conant, 2008, USA [23] | *** |
| Gorzowski, 2010, USA [21] | *** |
| Majnemer, 2011, Canada [27] | **** |
| McManus, 2008, Ireland [26] | **** |
| Odame, 2006, Canada [24] | **** |
| Pirpiris, 2006, USA [25] | *** |
| Verschuren, 2007, Netherlands [19] | *** |
| Qualitative studies | |
| Davis, 2009, Australia [31] | **** |
| Mandich, 2003, Canada [28] | **** |
| McBurney, 2003, Australia [32] | **** |
| Shikako-Thomas, 2009, Canada [29] | **** |
| Ronen, 1999, Canada [33] | **** |
| Young, 2007, UK [30] | **** |
| Mixed methods studies | |
| Becker, 2010, USA [35] | * |
| Davis, 2009, Australia [36] | * |
| Fraas, 2010, USA [34] | ** |
| Poulsen, 2008, Australia [37] | ** |

Scores vary from * (25%)—one criterion met, to **** (100%)—all criteria met

children with neurodevelopmental disabilities. A thematic inductive approach was used to synthesize common themes that explained this phenomenon based on quantitative, qualitative, and mixed methods studies.

A total of 19 studies were included in this review. Overall, the findings from the quantitative studies indicated the existence of a relationship between participation in a variety of leisure activities and different dimensions of QoL [19–27]. Qualitative data obtained from children with neurodevelopmental disabilities and their parents explored in-depth aspects of leisure and social experiences as contributing to QoL [28–33]. The main relationships between leisure participation and QoL that emerged from the 19 studies include the following: (a) physical and everyday activities enhance physical well-being; (b) participation in leisure activities has an effect on self-perception, they can promote emotional and social QoL, and may contribute to happiness and enjoyment; and (c) participating in chosen, preferred activities promotes overall well-being. This review also highlighted some negative impacts of involvement leisure activities on QoL, mostly mediated by environmental constraints related to accessibility or lack of adaptations.

Health promotion guidelines stipulate the importance of participation in activities such as physical activities for health and well-being, and the need to participate in such activities is viewed as a priority in health promotion initiatives for all children [38]. Children with neurodevelopmental disabilities may experience poor motor, cognitive, speech, social, and/or behavioral skills that negatively affect leisure participation. Individuals with disabilities often report a similar QoL as their typically developing peers. Indeed, children and parents may perceive that the disability in itself does not represent a barrier for their QoL, but rather, the child's ability to engage in a variety of preferred activities contributes to a great extent to their health and well-being. However, physical and attitudinal environmental obstacles that limit participation in leisure activities may impose constraints on QoL [10, 29, 39]. Efforts to change attitudes and enable children with neurodevelopmental disabilities to participate in the activities that matter most to them are critical. Educational and rehabilitation services and policies should facilitate the rights of these children as citizens and advocate for their participation in society in order to optimize their well-being. Defining, understanding, and intervening in problem areas as well as strengthening potential resources and supports can improve children's functioning and well-being.

This review presents some interesting points to consider in interventions and health promotion initiatives. An important finding was that most positive QoL outcomes were related to the child intrinsically, such as better physical well-being, self-perception, and emotional well-being while the negative ones were mostly related to environmental factors. This indicates that adaptations to the attitudinal and physical environment are required in order to facilitate participation of children with neurodevelopmental disabilities in activities of their choosing. In this case, the personal benefits including increased self-perception and emotional well-being may be achieved to their fullest potential.

Several qualitative studies have found that engaging in leisure activities with typically developing peers as well as with peers with disabilities had a positive effect on QoL. However, none of the quantitative studies considered this variable. It is important to acknowledge the fact that there are benefits for both segregated and integrated leisure opportunities, and therefore, children should have the chance to engage in both. Leisure programs should consider the structure and organization of activities that take into consideration different levels of ability, adaptation of equipment and materials, and training of the personnel supervising the activities to integrate and valorize the potential of all children.

Another aspect that emerges from the analysis is the difference between spontaneous leisure activities and leisure that occurs within a structured therapeutic program. The diversity of interventions and methodologies in the studies reviewed does not allow us to draw a specific conclusion as to which type of leisure activity may be better. However, we can infer that the phenomena of the relationship between a positive QoL and participation in leisure activities is present in both type of activities, supporting the conclusion that engagement in a variety of activities is beneficial for the child's QoL. Further investigation should be done to assess the difference in outcomes of both naturally occurring and organized therapeutic programs in order to establish priorities in program development and policy.

This systematic review included quantitative, qualitative, and mixed methods studies, which enabled a comprehensive appraisal of the association between leisure participation and QoL. While this methodology facilitated the exploration of the richness of data, an important methodological limitation is the inability to generalize from qualitative studies and case studies. Rather, they were used to obtain an in-depth understanding of specific phenomena. A wide array of outcome assessment tools in the quantitative studies precluded a formal meta-analysis. Furthermore, a heterogeneous etiology within the broad realm of neurodevelopmental disability, and a combination of parent-proxy and child self-report of QoL are other limitations to consider. One study used a health status assessment tool to measure health-related QoL [22]. There is no consensus as to how QoL should be measured, and the measurement of participation in children has been reported as being challenging as well [40]. This problem is partially due to the complexity and multidimensionality of both constructs. A multitude of factors contribute to QoL (i.e., an individual's feelings of well-being and contentment with life). Therefore, it cannot be inferred that participation in different leisure activities is central to promoting QoL, but rather is a piece that can be incorporated into interventions and a mechanism to approach other intervening factors such as environmental accessibility and personal attributes.

Understanding the impact that neurodevelopmental disabilities can have on children and their families is critical for professionals who provide services to these children. Professionals need to consider complex social, emotional, and physical outcomes such as leisure participation and QoL when planning interventions and developing programs [12]. Exploring the underlying components of both constructs and how they influence each other is essential to act pragmatically. It is generally assumed that increased involvement in leisure activities will enhance the

health and QoL of children and youth with disabilities. This systematic review highlights that there is a lack of empirical evidence, particularly for specific diagnostic groups or stages of development, to support this assumption. However, this systematic review and thematic analysis aimed to explore the phenomena that emerged from the interaction of the objective aspects of leisure participation and the subjective QoL dimensions in children with neurodevelopmental disabilities. Findings provided novel and clinically useful evidence on the important association between these two concepts.

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