Social determinants of health: What gives? Exploring health outcomes in pediatric populations using a social deprivation framework

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Abstract

Background: It is well established in public health literature that the environments in which children grow, learn, and develop make important contributions to the quality of their health, life, and wellbeing. To understand the specific barriers and facilitators inherent to an individual's environment or set of circumstances, the World Health Organization (WHO) categorizes these factors under the term 'social determinants of health' (SDOH). SDOHs represent the numerous economic and social factors influencing environmental living conditions, such as access to services, resources, and social support. Social deprivation is one construct that measures these determinants, commonly using census data to measure the area-level impact of socio-economic disparities in public health. Among individuals with childhood disabilities, caregivers represent the primary measurement in public health contexts as they are the first line of support in all areas of their child's development. At the same time, caregivers are known to experience the impact of their child's disability in mental, physical, and emotional domains, and also require support.

Methods: To understand the contributions of SDOH to health outcomes found among children with disabilities this thesis explored the impact of the combination of social and economic factors in two different early-onset conditions. The scoping review (Manuscript 1) investigated social deprivation in autism spectrum disorder, or ASD, which informed the investigation of indicators used to identify data points for Manuscript 2. Our scoping review included key stakeholders in the ASD community who informed at each stage of the knowledge synthesis. Our findings demonstrated the challenge of defining social deprivation in research. It also demonstrated increased levels of social deprivation in the ASD cohort, and the need for caregivers to receive support. We used these findings to inform Manuscript 2, which explored the role of SDOH in a group of rare musculoskeletal diseases, Arthrogryposis Multiplex

Congenita (AMC) using data from a hospital-wide registry. Extracting data related to social deprivation indicators, we used structural equation modeling (SEM) to analyze the strength of association and direction between personal and environmental factors (SDOH), and the health outcomes of individuals with AMC ages 8-21 (e.g. physical function, mood, pain, health perception, and peer relationships).

Results: Our results indicated that the environmental factors inherent to caregivers played less of a role in AMC than in ASD. Increased impact of AMC on physical function was most strongly associated with increased frequency of parental stress, indicating that caregivers of children with increased AMC severity likely require more attention and support in clinical contexts.

Conclusion: The context of patients included both personal and environmental factors, of which SDOH are a part. The results indicate that the degree of impact by both personal and environmental factors appear to vary based on the limitations imposed by severity of the physical condition in conjunction with cognitive and psychological barriers. To advance patient-centered care, clinicians are encouraged to 1) discuss the impact of personal and environmental factors with children with disabilities and their caregivers to reduce population-level and individual-level barriers, and 2) to include support options for caregivers in addition to the needs of the child.

Abrégé

Contexte : Il est établi dans la littérature sur la santé publique que les environnements dans lesquels les enfants grandissent, apprennent et se développent contribuent de considérablement à la qualité de leur santé et de leur bien-être. Pour comprendre les barrières et les facilitateurs spécifiques et inhérents à l'environnement d'un individu, l'Organisation mondiale de la santé (OMS) classe ces facteurs comme "déterminants sociaux de la santé" (DSS). Les DSS représentent les nombreux facteurs économiques et sociaux qui influencent les conditions de vie, comme l'accès aux services, aux ressources et au soutien social. La privation sociale est un concept qui mesure ces déterminants à l'aide de données de recensement pour quantifierl'impact des disparités socio-économiques régionales sur la santé publique. Parmi les enfants souffrant de handicaps, les aidants, étant la première ligne de soutien de ces derniers, représentent la principale mesure dans la santé publique. En même temps, on sait que les aidants subissent l'impact mental, physique et émotionnel du handicap de leur enfant et qu'ils ont également besoin de soutien.

Méthodes : Afin de comprendre les contributions des DSS aux résultats de santé observés chez les enfants handicapés, cette thèse a exploré l'impact de la combinaison des facteurs sociaux et économiques dans deux troubles différents d'apparition précoce. L'étude exploratoire (Manuscrit 1) a porté sur la privation sociale dans les troubles du spectre autistique (TSA), ce qui a orienté l'étude des indicateurs utilisés pour identifier les points de données pour le Manuscrit 2. Notre étude exploratoire a inclus des parties prenantes de la communauté des TSA qui ont contribué tout au long de la synthèse des connaissances.

Résultats : Nos résultats ont démontré la difficulté à définir la privation sociale dans les contextes de recherche. Ils démontrent également le niveau plus élevé de cette dernière au sein

des cohortes TSA ainsi que le besoin de soutien aux aidants. Nous avons utilisé ces résultats pour alimenter le manuscrit 2, qui explore le rôle des DSS dans un groupe de maladies musculo-squelettiques rares, l'arthrogrypose multiple congénitale (AMC), à l'aide de données provenant d'un registre hospitalier. En extrayant les données relatives aux indicateurs de privation sociale, nous avons utilisé la modélisation par équations structurelles (MES) pour analyser la force de l'association et la direction entre les facteurs personnels et environnementaux (DSS) et les résultats en matière de santé des personnes atteintes d'AMC entre 8 et 21 ans (par exemple, la fonction physique, l'humeur, la douleur, la perception de la santé et les relations avec les pairs). Nos résultats indiquent que les facteurs environnementaux inhérents aux aidants ont joué un rôle moins important dans l'AMC que dans les TSA. Là où la sévérité de l'AMC est plus élevée, un stress plus considérable est rapporté par les aidants.

Conclusion : Le contexte des patients comprend à la fois des facteurs personnels et des facteurs environnementaux, dont les déterminants sociaux de la santé font partie. Les résultats indiquent que le degré d'impact des facteurs personnels et environnementaux semble varier en fonction des limitations imposées par la sévérité de la condition physique en conjonction avec les barrières cognitives et psychologiques. Pour faire progresser les soins centrés sur le patient, les cliniciens sont encouragés à 1) discuter de l'impact des facteurs personnels et environnementaux avec les enfants handicapés et leurs soignants afin de réduire les obstacles au niveau de la population et au niveau individuel, et 2) à inclure des options de soutien pour les soignants en plus des besoins de l'enfant.

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Contribution of Authors

This thesis consisted of two manuscripts. The first was submitted to *Autism*, and the second will be submitted to ORPHANET after approval by all co-authors. These manuscripts are not being submitted elsewhere by any co-authors.

Chapter 1 – Rationale and Objectives: Tessah wrote the chapter with review by Dr. Noémi Dahan-Oliel and Dr. Laurie Snider.

Chapter 2 – Literature Review: Tessah wrote the chapter with review by Dr. Clarice Araujo, Dr. Noémi Dahan-Oliel, and Dr. Laurie Snider.

Chapter 3 – Manuscript 1: "*What is known about social deprivation in Autism? A scoping review"*

Dr. Snider and Dr. Dahan-Oliel guided the scoping review process by devising the theme (social deprivation), attending stakeholder meetings, and reviewing the completed manuscript. Tessah prepared the search strategy with expert librarian Jill Boruff. Translations for each database were completed by Tessah and co-author Shahrzad Nematollahi, and confirmed by Jill Boruff. Tessah and Shahrzad screened all abstracts and titles, completed full-text review, and data extraction. The selection criteria table was created by Shahrzad and revised by Tessah. All other tables and the complete manuscript were completed by Tessah. The draft was sent to all co-authors before submission.

Dr. Anita Menon provided comments regarding knowledge translation strategies, and applicability of findings to research settings. Dr. Keiko Shikako provided comments to strengthen the discussion section. All other co-authors suggested minor revisions to the introduction and methods sections that included rephrasing sentence structure and grammar, reporting style, and contributed to guiding the focus of the discussion section and interpretation of results.

Chapter 4 – Connecting Manuscripts 1 & 2: Tessah wrote the chapter with review by Dr. Clarice Araujo, Dr. Noémi Dahan-Oliel and Dr. Laurie Snider.

Chapter 5 – Manuscript 2: "*Using a social deprivation perspective to investigate functional outcomes from the AMC Registry Project: an SEM analysis*"

Dr. Noémi Dahan-Oliel was the project lead for the AMC Registry Project (Dahan-Oliel et al., 2022). She oversaw collection of data from all 8 locations. The data set was reviewed by Dr. Noémi Dahan-Oliel and Tessah for variables to include from the scoping review project and literature search. Data was extracted by Sena Tavukçu, the clinical research coordinator Sena Senha at Shriners Hospital for Children, Montreal. Statistical consultation was provided by Dr. Nancy Mayo, who met for multiple meetings with Tessah to establish the research question, research design, data preparation, statistical analysis plan, and interpretation of results.

Data scientist Dr. Stanley Hum was brought in by Dr. Mayo's recommendation to prepare the data set, construct the SEM model, test its parameters. When the model showed poor fit, Dr. Hum and Dr. Mayo met to make modifications, and presented the final model with results to Tessah. Using SPSS, Tessah evaluated the sample's distribution, and synthesized all data tables: sample characteristics, summary of outcomes, and tested all environmental variables with linear regressions. Tessah created all tables which were reviewed by Dr. Mayo for completeness and accuracy. Using the output from Mplus, Tessah illustrated each model figure in Microsoft Word. All written content in this manuscript was done by Tessah, and reviewed by Dr. Laurie Snider and Dr. Noémi Dahan-Oliel.

Chapter 6 – Discussion: Tessah wrote the chapter with all sections and infographics reviewed by Dr. Shahrzad Nematollahi, Dr. Noémi Dahan-Oliel and Dr. Laurie Snider.

Chapter 7 – Conclusions: Tessah wrote the chapter with review by Dr. Noémi Dahan-Oliel and Dr. Laurie Snider.

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Abbreviations

ABA – Applied behavioural therapy

ASD – Autism spectrum disorder

APPT – Adolescent pediatric pain tool

AMC – Arthrogryposis multiplex congenita

CAHS – Canadian Academy of Health Sciences

CFI – Comparative fit index

CG – Caregiver

CNS – Central nervous system

CRIR – Centre of Interdisciplinary Research in Rehabilitation

CSDH - Commission on Social Determinants of Health

DSM-V – Diagnostic and Statistical Manual of Mental Disorders

EHR – Electronic health records

ESDM – Early-Start Denver Model

ESI – Environmental support index

EQ-5D-Y – EuroQOL - 5 Dimensions – Youth version

FAQ – Gillette Functional Walking Subscale

HIC – High income countries

HRQL – Health-related quality of life

ICF – International Classification of Functioning and Disability

ICF/WC – International Classification of Functioning and Disability with Wilson-Cleary extension

IMD - Index of Multiple Deprivation

IRB – Institutional Review Board of Research Ethics

KT – Knowledge translation

MLE – Maximum Likelihood Estimation

PRISMA – The Preferred Reporting Items for Systematic Reviews and Meta-Analyses

PROMIS-SF – Patient-Reported Outcomes Measurement Information System – Short Form

RMSEA – Root Mean Square Error of Approximation

SES – Socioeconomic status

SDI – Social deprivation indices

SDOH – Social determinants of health

SD – Standard deviation

S.E. – Standard Error

SEM – Structural equation modelling

SRMR- Standardized Room mean square residual

TLI – Tucker-Lewis Index

WeeFIM – The Functional Independence Measure for Children

VAS – Visual analogue scale

CHAPTER 1: INTRODUCTION & LITERATURE REVIEW

1. Background

The primary focus of this thesis is social determinants of health (SDOH) as investigated in the context of childhood disability. Children with disabilities are vulnerable to the environments in which they are born, live, and grow. According to the World Health Organization (WHO), these non-medical conditions, or environmental factors, comprise SDOH (World Health Organization, 2013). In a recent review of SDOH in pediatric disability populations, SDOH were increasingly recognized as important yet understudied (Hollin et al., 2022). This warrants the attention of researchers in childhood disability to identify barriers and facilitators that benefit the health and quality of life of this large community.

To investigate this topic, two childhood disability populations were identified, and two frameworks were integrated to situate the research topics and synthesize findings. The first framework is the International Classification of Functioning, Disability, and Health (ICF) ,a well-established framework that provides a classification system of functional outcomes and contextual factors known to differentially influence health in individuals with disabilities (World Health Organization, 2013). The second framework is the Wilson Cleary conceptual model which proposes relationships between variables and represents domains of health perception, not included in the ICF. The purpose of integrating these two frameworks as a conceptual model in my thesis is for the modeling of complex factors that interact at the individual and population levels in health conditions. To advance research in SDOH, this thesis will explore environmental and personal factors related to functional health outcomes, address SDOH-related barriers at the population-level, and support applicability of evidence-based patient-centered approaches.

1.1. Social determinants of health

Over the last three decades, public health research has demonstrated robust evidence that non-medical factors like socioeconomic status, income, and area of living, are more influential to population health than medical factors, accounting for up to 55% of health outcomes (Victoriano & Gauthier, 2009; Andermann, 2016; World Health Organization, 2013). These factors, or SDOH, are the non-medical components and conditions in which people live, work, and age (What are the social determinants of health?, 2008). SDOH are driven at systemic levels by economic, political, and social policy development, and social norms that directly influence the conditions of daily life (Braveman & Gottlieb, 2014). Examples of SDOH factors include socioeconomic status, education, employment/income, area of living, housing conditions, social support, access to health care, and exposure to discrimination or social inequalities (Andermann, 2016). SDOH can either facilitate factors that are protective and support optimal health, or become an insurmountable barrier leading to diminished health, adverse events, and social disadvantage (Ostoijic et al., 2023).

1.2. Origins of Social Determinants of Health

Social determinants of health can be measured at both the population- and individual-level. In fact, SDOH research has its origins in population-level research set in the late 1960's with the Whitehall study conducted by British researcher Michael Marmot. Marmot's 1967 study was the first to develop a large-scale, area-based research design including demographic and socioeconomic factors in relation to mortality and health (Marmot et al., 1978). It established steep associations between coronary risk factors and coronary heart disease (CHD) and employment grade among 17, 530 British civil servants living in London, England over a 9-year period (Marmot et al., 1978). This study found that the men in the lower employment grades had 3.6 times the rate of CHD mortality than the highest employment grades (Marmot et al., 1978).

The second Whitehall study further established the importance of SDOH by exploring how health outcomes could vary by social gradient when poverty is not present (Marmot et al., 1991). This study observed the role of psychosocial factors such as work-related stress and work-family conflict on outcomes such as high blood pressure, smoking incidence, and mortality (Marmot et al., 1991). The results were consistent with the Whitehall study I, where a significant association was found between lower employment grade and: psychosocial satisfaction, lower health perception, high incidence of health symptoms and chronic conditions, and social support (Marmot et al., 1991).

Since the Whitehall studies, a variety of SDOH indices have been developed in high income countries (HIC) such as Canada, the United Kingdom (UK), and the United States (Pampalon, 2009; Townsend, 1987; Robert Graham Center, 2018) to observe the social and socioeconomic gradients and risk factors in health outcomes among populations (Braveman & Gottlieb, 2014). To date, the most cited SDOH index is that of Peter Townsend, who developed the Townsend deprivation index in the late 1980's (Zelenina et al., 2022). Townsend uses 4 domains (e.g., unemployment, house overcrowding, non car ownership, and non home ownership) to identify areas of the United Kingdom with unmet needs due to lack of resources (Townsend, 1987). Deprivation in this context can implicate both social and material domains, and it thus has been used to describe various forms of 'deprivation,' including socioeconomic (Wilson et al., 1999), social (McConachie et al., 2009; Warnell et al., 2015), and neighbourhood deprivation (Jivraj et al., 2021).

1.3. Social deprivation

Numerous social deprivation indices have been developed in countries around the world to identify small-areas or neighborhoods of disadvantage leading to worsened health (Zelenina et al., 2022). Social deprivation is categorized as a risk factor in the SDOH (Chen et al., 2020), and is defined as the limiting of social opportunity based on one's material and social status, and living area (Social Deprivation Index, 2022). Social deprivation is used by governments, policymakers, researchers to map out the distribution of health inequity by small-area level using various social and material indicators clustered by area-code. The resulting outcome is a statistic to quantify areas in which greater mortality, disease severity, and morbidity are found. Generally, high scores on most indices indicate high deprivation levels and are associated in many populations with decreased quality of health outcomes or quality of life (Kashem et al., 2019).

Since the COVID-19 pandemic, social deprivation and its impacts have been increasingly studied among individuals with disabilities. Among adults living with chronic conditions, social deprivation has been repeatedly associated with poorer physical and mental health outcomes, and lower quality of life (Alegría et al., 2018). In pediatric populations, social deprivation has a variety of associations depending on the category of the condition. In conditions producing long-standing movement impairments such as cerebral palsy (CP), studies have shown an association between high deprivation levels are associated with a decreased frequency in community and leisure participation in children with CP, (Hammal et al., 2007; Zeidan et al., 2021; Hollin et al., 2023); increased bilateral spasticity, and severity of comorbidities (Ostoijic et al., 2023).

In the United States, over half of those in the ASD community were reported to live in low-income neighborhoods (Anderson et al., 2022), while in the United Kingdom, higher deprivation levels were associated with increased prevalence of ASD (Delobel-Ayoub et al., 2014). For children with ASD in Canada, higher neighbourhood deprivation status was associated with increased vulnerability in learning domains, reducing likelihood of kindergarten children with ASD attaining social competence, communication skills, and general knowledge domains (Siddiqua et al., 2020). Unmet needs in the ASD community are largely associated with economic cost, level of deprivation, number of household caregivers, and level of informal social support (Roddy et al., 2020; Sapiets et al., 2023). To address these disparities at the clinical level, recommendations to reduce these barriers involve addressing economic or social disparities by providing more flexible or accessible services (Sapiets et al., 2023). However, more action is required at the health systems and policy levels to address these system-related barriers. To understand the population-level impacts of SDOH and contribute to the research used in policymaking of socially disadvantaged childhood disability populations, I conducted a scoping review using an inductive framework and stakeholder consultation, from January 2022 to May 2023 to explore what is known about social deprivation in the context of children with ASD. This comprises the first manuscript of this thesis. The second manuscript used the findings of Manuscript 1 to identify data elements representative of environmental factors (SDOH and social deprivation), and explored the extent to which environmental or personal factors were associated with functional outcomes in children with an early-onset musculoskeletal condition.

1.4. Social Determinants of Health in recent years

From 2005 to 2008, the WHO launched the Commission on Social Determinants of Health (CSDH) to raise global awareness and partner with global and community-level institutions to reduce the impact of non-medical contributors to ill health (Lee, 2005; CSDH, 2008). The aim of the CSDH was to foster support and collaboration with health organizations to target SDOH and reduce health inequity (Mensah & Riley, 2021) by addressing daily living conditions, and creating policies surrounding inequitable distribution of power and resources (Irwin et al., 2006).

1.4.1. Challenges in implementing Social Determinants of Health

Since 2008, research initiatives have greatly advanced the understanding of specific SDOH through implementation of strategies at the population- and individual-levels. Population-level strategies include spatial analysis of SDOH characteristics, such as social and material deprivation indices, where an index analyzes census data to produce a metric representing SDOH by area (Tyris et al., 2023). Individual-level strategies include collecting and integrating SDOH data in clinical settings into electronic health records (EHRs) using a variety of screening tools either digitally or face-to-face by a doctor or pediatrician (e.g. iScreen, Family Needs Screening Tool, Patient-Centered Assessment Method) (Hewner et al., 2017; Sokol et al., 2019). Although individual-level screening and inclusion of SDOH in clinical settings (e.g., merging SDOH into EHRs) been recommended and implemented by researchers (Devoe et al., 2016; Hudon et al., 2022) and select policy agendas (Baker et al., 2017), the limitations in feasibility, accuracy, ethics, and social-undesirability of discussing social factors (e.g. income, socioeconomic stability) with patients present significant challenges in clinical settings (Chen et al., 2020; Van Cleave et al., 2022). A systematic review examining the use and efficacy of SDOH screening tools noted that the tools 1) did not demonstrate reliability, 2) validity, 3) were highly variable in interpretations, and 4) did not adequately contextualize the chronology or duration for which the collected data was applicable (Sokol et al., 2019).

Although the implementation and inclusion of SDOH data into electronic health records (EHRs) has demonstrated significant predictive advantages for clinicians when EHRs included SDOHs in hospital and clinical settings, the advantages were limited by the context or type of outcome (Chen et al., 2020). For example, housing status, income, and education were significant predictors of emergency service utilization, drug use, suicide prevention, and housing stability, but were not predictive of hospital re-admission rates, SDOH-related referrals, or specified physical health outcomes (Chen et al., 2020). Overall, the benefits of individual-level measures of SDOH appear to be limited to certain contexts. Another study examining SDOH screening recorded in EHRs by clinicians found that patient data pertaining to SDOH were present in only 40% of patient appointments (Bunce et al., 2023). This indicates further research must be done to make SDOH research more understandable for clinicians and patients and advance the clarity of population- and individual-level determinants in order to optimize patient-centered care. Although integration of SDOH has progressed from the 2005 Commission, the majority of studies reported on adult populations. Therefore, research on SDOH in pediatric disability populations is still in its early stages (Hollin et al., 2022; Van Cleave et al., 2022).

1.5. Vulnerable populations

Globally, it is recognized that vulnerable individuals and those living in socially disadvantaged situations are at risk of diminished mental and physical health, health care access, mortality, and increased severity of health conditions than those living in socially advantageous circumstances (Irwin et al., 2006; World Health Organization, 2003; Ostojic et al., 2023). Pediatric populations are especially vulnerable to the impacts of SDOH in the early stages of development when the infants are care-dependent. However, the effects can be simultaneous and reciprocal, as both the child and the caregiver have an experience of the physiological limitations and psychological burden of the condition within the context of their own SDOH. In this way, SDOH can become an even greater barrier for both the child and the caregiver, as caregivers are responsible for not only providing for themselves and their family, but also physically and psychologically supporting a child with a chronic disabling condition, and facilitating their treatment. As a result, not only children with disabilities, but also their caregivers report worse mental health (e.g. anxiety, depression), parental stress (Murphy et al., 2007; Wang, 2021), and lower quality of life (Benjak, 2011). The combination of these factors over time can negatively impact employment and income stability within the home. For this reason, it is important to advance research efforts to identify the ways in which SDOH can be improved to reduce barriers specific to children living with disabilities and their families.

1.6. Terminology in Social Determinants of Health

Literature addressing SDOH in clinical contexts uses terms to identify determinants by their relationship to 'upstream' or 'downstream' factors (Anderman, 2016). Structural or population-level determinants are identified as 'upstream,' while health outcomes and individual-level determinants impacted by upstream factors are identified as 'downstream' (Braveman et al., 2014; Anderman, 2016). Although this establishes a directional understanding of factors, the categorization may not account for mediating or moderating factors that are influential to rehabilitation outcomes, diminishing applicability in clinical practice.

To gain a better understanding of the interplay between social deprivation and health outcomes in childhood disability, recent research has emphasized incorporating a dynamic, multidirectional paradigm (Thimm-Kaiser et al., 2023). Such a paradigm should incorporate determinants in a continuum in relation to their scale of influence (e.g. population or individual-level), experience in time (e.g. cumulative exposure to factors across a lifespan), and the individual's characteristics and responses (e.g. physiological characteristics, behavioural tendencies, level of resilience) (Thimm-Kaiser et al., 2023). By considering such factors, one can account for the numerous mechanisms acting simultaneously that can produce different effects based on the individual and their behavioral response to their immediate environment. Additionally, in research and clinical settings, ascertaining the extent to which population-level determinants are impacting individual-levels (and vice versa), are unclear, given that not all drivers of SDOH produce the same effects in different contexts (Thimm-Kaiser et al., 2023).

1.7. Conceptual frameworks

1.7.1. The International Classification of Functioning, Disability, and Health

To address these challenges, this thesis used the ICF framework with the Wilson-Cleary extension. The ICF framework was developed to map the impact of disability, providing a comprehensive taxonomy of health, disability, and functioning. The ICF framework recognizes that health is influenced by a complex interplay of biological, psychological, physical, and social factors, and integrates the medical and social models of disability to form the biopsychosocial model (World Health Organization, 2013).

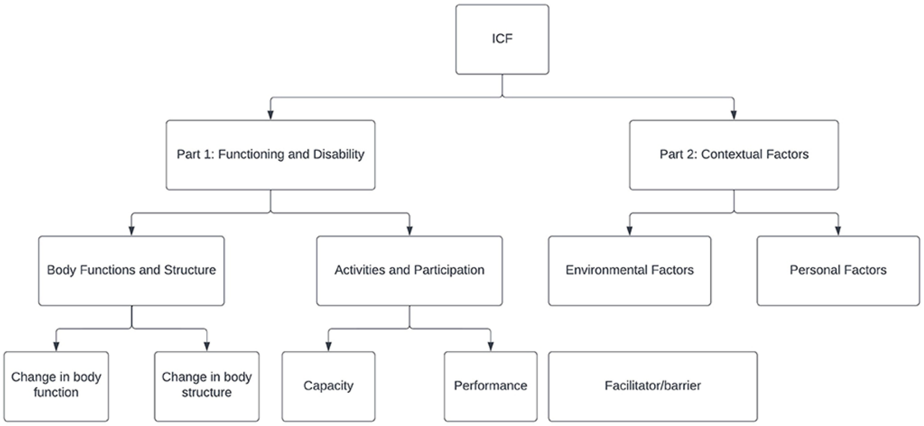

Figure 1: The ICF Framework (World Health Organization, 2013)

Illustrated in Figure 1, the ICF organizes factors by their relationship to 1) functioning and disability, and 2) contextual factors (World Health Organization, 2013). Functioning and disability involve three main categories by which to identify the impacts of health conditions: body structures and functions, and activities and participation. Contextual factors include aspects inherent to the individual, termed personal factors, and the social and material aspects of their specific environment, or environmental factors.

Personal factors are intended to provide context specific to the individual and their characteristics, but can be represented by multiple indicators applicable to the line of study. Examples of these are age, ethnicity, biological sex, perceived gender, beliefs, attitudes, and personality. Environmental factors are the physical, social, and attitudinal environments where individuals conduct their lives, and are distinguished at individual and societal levels (World Health Organization, 2006). They include the external physical, social, and attitudinal environments where children are born, live, and grow. These factors influence individuals directly, and are categorized in the Children and Youth version of the ICF (ICF-CY) according to the extent these factors create barriers or facilitators impacting body structure or function, performance of every-day activities, and participation in social opportunities.

Societal environmental factors involve structural organization at the population-level. These factors pertain to the region where the child lives, and the health service access, communication, and transportation systems in place. On the other hand, individual environmental factors pertain to the physical settings where a child engages with material environments (e.g. home, school, or neighbourhood) and social relationships (e.g. family, peers, acquaintances) (World Health Organization, 2006). Situating contextual factors in these categories is helpful to practitioners and researchers, in that addressing barriers at the level they are impactful can more effectively direct resources and advise the relevant health systems in place who are responsible for public, community, or individual health.

1.7.2. Wilson-Cleary extension

Despite the comprehensive classifications available in ICF, the gaps in hypothesis generation it presents are remedied by the integration of the conceptual model presented by Wilson & Cleary. The Wilson-Cleary conceptual model is the most well-cited framework in health-related quality of life (HRQL) literature (Bakas et al., 2012), and proposes five categories of health concepts, three of which overlap with the ICF: biological and physiological factors, symptom status, functioning. Two important concepts are added: general health perceptions and HRQL (Wilson & Cleary, 1995). Important to the methodology of the second manuscript, the Wilson-Cleary extension goes beyond taxonomy of medical and non-medical factors, and proposes specific causal relationships among these five health concepts, which are useful in statistical modeling and hypothesis testing (Ojelabi et al., 2017). The relationships proposed in the Wilson-Cleary extension are the following: symptoms mediate physiological factors and functional status; functional status mediates between symptoms of health perception, and health perception mediates between functional status and overall HRQL reports (Wilson & Cleary, 1995; Bakas et al., 2012). Figure 1 illustrates the integration of the ICF and Wilson-Cleary model (ICF/WC).

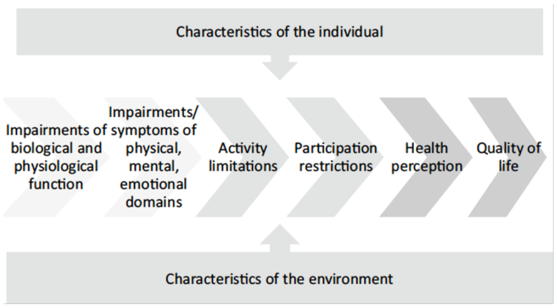

Figure 2: ICF/WC Integrated model (Mayo, et al., 2019)

Because the direction of impact is already specified, the Wilson-Cleary model provides advantages in hypothesis generation, testing, and targeting of specific mediating factors, which can promote optimized treatment options. To that end, the Wilson-Cleary model has demonstrated efficacy in the research of chronic conditions such as HIV/AIDS, chronic obstructive pulmonary disease, and arthritis, where health perception and HRQL are significantly impactful to psychosocial domains, and are impaired as a result of chronic symptoms (Ojelabi et al., 2017; Mayo et al., 2019). As changes in HRQL and health perception are highly subjective and individually based, this model includes patient-reported measures to represent the patients experience of their health. This inclusion has led to a practical understanding of health perception as a predictor of other health-related outcomes such as frequency of hospitalization (Chen et al., 2020), and health seeking behaviours (Abuduxike et al., 2020). These attributes are important to the second manuscript, where patient-reported outcome measures comprised the majority of the data for children with arthrogryposis multiplex congenita (AMC), and modeling of complex factors related to health perception were of primary interest to the research question.

1.8. The ICF in pediatric contexts

Exploring pediatric disability entails a slightly modified interpretation from adult populations as caregiver attributes directly influence a child's physical environment, health, and psychosocial development over time (Hollin et al., 2022; Scattolin et al., 2022). Studies show that SDOH of a child are associated with or co-vary with social and material attributes of the caregiver, since caregivers are primary contributors to the child's immediate environment (e.g., attitudes, norms, parenting styles, mental health status, financial resources to acquire assistive devices, and transportation) (Jiandani & Mhatre, 2023), For example, level of education and socioeconomic status of the caregiver were shown to be associated with the level of social support and anxiety/depression in children, respectively, (Scattolin et al., 2022). In addition, increased duration of employment instability has been associated with an increase of the child's physical issues such as increased frequency of migraines and respiratory problems (Victorino, & Gauthier, 2009).

Adding to this, parenting style, caregiver mental health, and conditions of the home environment also have demonstrated significant and lasting impacts on children throughout development, whether positive or negative, especially in the context of chronic disability (Scattolin et al., 2022). Caregivers are the most consistent and enduring influence in their child's development, where the child learns social and material norms, and develops personal characteristics in response to their immediate surroundings (e.g. self-perception, emotional tendencies to internalize/externalize, physical and mental health, and expectations in social relationships) (Scattolin et al., 2022). The personal characteristics in turn support complex functioning in health-related quality of life domains, implicating a child's subjective experience of their health perception, mood, and peer relationships.

1.9. Populations explored

The populations explored in this thesis and manuscripts were comprised of two different sets of conditions: a group of rare musculoskeletal conditions under the diagnostic group arthrogryposis multiplex congenita (AMC), and the more prevalent neurodevelopmental diagnosis of autism spectrum disorders (hereafter, ASD). AMC and ASD are distinct types of disabilities with inherent differences in onset, limitations on body structure and function, and etiology. However, these conditions are similar in diagnostic challenges, frequency of co-morbidities, long-term and individualized multidisciplinary rehabilitation, and high involvement of caregiver support.

CHAPTER 2: RATIONALE & OBJECTIVES

2.1 Rationale

The populations selected for this thesis were chosen to explore the ways in which SDOH can exert a differential impact on pediatric populations where one diagnosis is categorized as a visible physical disability requiring invasive surgical and physical therapy interventions, while ASDs are categorized as an invisible disability (Invisible Disabilities® Association, 2023) and require frequent psychosocial therapy. Comparing and contrasting SDOH in this way is intended to promote our understanding of SDOH in the area of childhood disability and increase the generalizability of findings. This will contribute to a more holistic and context-specific paradigm that can be used by clinicians and researchers when supporting pediatric disability populations.

2.1.1. Differences between AMC & ASD

AMC and ASD are distinct disability categories, each characterized by unique features that encompass inherent differences in terms of diagnostic onset, impact, and frequency.

AMC typically presents at birth or shortly thereafter through the identification of joint contractures, where diagnosis is established by a combination of diagnostic imaging, medical history, genetic testing, and physical examination during infancy (Dietrich et al., 2019). In contrast, ASD is commonly diagnosed by behavioural criteria from the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) which inventories delays in social and cognitive developmental milestone attainment around the ages of two or three (Gabbay-Dizdar et al., 2021). Where AMC primarily affects the musculoskeletal system impairing joint mobility, physical functioning, and activities of daily living (Dahan-Oliel et al., 2022). ASD primarily impacts neurodevelopment, resulting in difficulties with social interactions, language acquisition, sensory processing, mental health disorders, and maladaptive behaviors (Lai et al., 2014).

Finally, the frequency of these conditions in the global population differs dramatically: AMC is a group of rare conditions estimated to occur in approximately 1 in 3-5,000 live births (Lowry et al., 2010); ASD affects about 1 in 100 children (Zeidan et al., 2022).

2.1.2. Similarities between AMC & ASD

Despite these fundamental differences, AMC and ASD have important similarities in clinical and rehabilitation contexts. Both conditions are heterogeneous in nature, presenting diverse symptomatology in early development. Their diagnosis and treatment require multidisciplinary collaboration with physicians, specialists, and rehabilitation experts as ASD and AMC have high frequency of multi- or co-morbidities with physical, cognitive, or psychological conditions. Socially, both are known to experience social isolation and negative impacts to their social relationships (Cachecho et al., 2021; Umagami et al., 2022) either due to barriers in physical or social participation, or the lack of understanding of the condition (Kasari & Sterling, 2014). Among children with autism, a long-standing parental concern is in regards to their child's development of skills supporting participation in social settings, specifically peer relationships (Knot et al., 2006; Coussens et al., 2020). In AMC, parental concerns are related to the physical restrictions or limitations to attaining physical autonomy (e.g. mobility, activities of daily living, and self-care), both leading to limitations in social participation important in rehabilitation contexts (Elfassy et al., 2019;). Furthermore, due to the heterogeneity in expression, and presence of co-occurring conditions, recommended treatment strategies are patient-centered, focusing on the individual's unique symptomatology and thus require frequent follow-ups, ongoing support, and reliance upon collaboration between caregivers and clinicians, practitioners, and testing centers (Suen et al., 2021).

2.1.3. The roles of caregivers

Well established in pediatric disability populations is the critical role of caregivers in their child's care. Caregivers have multiple roles in the care of pediatric populations as they facilitate treatment-related engagement in addition to physical, financial, emotional, and social support. Examples of rehabilitation engagement are attendance and organization of multidisciplinary care appointments, adherence to interventions, transportation, and attention to individualized rehabilitation strategies from birth into adulthood. Although the above functions are directly facilitated by the caregiver, the role of socioeconomic disadvantage and urban planning can introduce insurmountable structural barriers in areas with high social deprivation, leading to worsened physical outcomes (Zeidan et al., 2020; Paget et al., 2022) and increased psychosocial stress (Emerson et al., 2004).

A 2022 review by McMullan and colleagues found that caregivers of children with rare diseases reported a knowledge gap between caregivers and clinicians, requiring more applicable and meaningful support from clinicians as well as specialized information regarding their child's condition (Mcmullan et al., 2022). As a result, caregivers reported that they were often 'obliged' to become an expert in their child's condition(s) to increase their ability to identify effective treatments and reduce its negative impacts on their child (Mcmullan et al., 2022). Subsequently, this has been associated with subsequent psychosocial stress, and a diminishing of psychosocial resources like energy, memory, and attention. The exhaustion of psychosocial resources detracts from important areas of functioning like caregiver capacity for social engagement, ability to maintain employment, and maintenance of their mental health.

Yet another role for caregivers pertains to social opportunities that entails two components: the quality of socioemotional relationship with their child, and the caregiver's facilitation of social activities with peers. Among children with ASD, for example, children who exhibit lower initiation in social activities with their caregivers or peers are more likely to experience social isolation later in life (Estes et al, 2018). Due to these challenges faced on an every-day basis, caregivers in ASD are known to experience higher levels of anxiety, depression, and stress (Wang et al., 2021, McLeod, 2023; Delaye et all, 2022). The resulting impact diminishes social opportunities, introducing barriers related to social participation for the caregiver and child (McLeod, 2023). Similar finding are also reported in AMC, children with AMC reporting more pain and fatigue were associated with diminished social participation and psychosocial wellbeing for both the caregiver and child (Cachecho et al., 2021; Elfassy et al., 2019).

Using the frameworks described to explore two different populations, this thesis will describe the population-level and individual-level health outcomes of the SDOH, with the aim of identifying the contexts in which SDOH are meaningful in childhood disability, specifically ASD and AMC. The hypothesis of this thesis is that SDOH are important and differentially impactful to the health and wellbeing of childhood disability populations. Based on the socioeconomic gradients in health observed in literature that more support and resources are linked with better health outcomes, the findings will reflect that SDOH influence health differently according to the contexts and childhood conditions in which they are explored. The implications of the findings in this thesis promote a comprehensive understanding of factors contributing to diverse health outcomes in both clinical and rehabilitation contexts. I have investigated contrasting methods of SDOH at the population-level (social deprivation) and the individual level (AMC registry data) to observe similarities and differences in health outcomes and measurement approaches. The methodology used in this thesis will identify which factors are associated with health outcomes of children and youth with disabilities and their caregivers.

Our findings will promote the clarity and optimization of considerations used in clinical decision-making specific to the pediatric disability population.

2.2. Objectives

The overall objective of this thesis is to explore how SDOH are differentially impactful in the context of chronic pediatric disability, and to propose a comprehensive paradigm including population and individual level considerations for practitioners, researchers, and those in the pediatric disability community. To accomplish this, I have performed a scoping review investigating a population-level measure of SDOH, social deprivation, in an early-onset pediatric population known to experience social and material, area-level barriers to treatment namely ASD (Manuscript 1). Then, I used the results from the scoping review to inform the SDOH indicators related to social deprivation, to investigate the impact of SDOH in a group of rare musculoskeletal conditions (i.e., AMC) using structural equation modeling in the context of a population-based registry for AMC (Manuscript 2).

2.3. Research Questions

The primary research question of my thesis was: what is known about SDOH and social deprivation among children with chronic disabilities?

The specific research questions represented by two manuscripts are:

1) What is known about social deprivation in the context of ASD? What deprivation levels are reported, how is it operationalized, and what measures are used (Manuscript 1)?

2) Using structural equation modeling from the subset of data extracted from the AMC registry representing children 8-21 with AMC and their caregivers, are functional outcomes associated with personal factors, or environmental factors (ie. the individual-level SDOH)? Secondly, which variables are most associated with functional outcomes (Manuscript 2)?

CHAPTER 3: MANUSCRIPT 1

*This manuscript was formatted for the requirements of Research in Autism Spectrum Disorders***.**

What is known about social deprivation in autism? A scoping review

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Abstract

Background: Social deprivation is a multidimensional construct that reflects the interrelated factors comprising the level of social opportunities of a family or individual. As individuals with autism are known to experience challenges in socially related domains, this scoping review aimed to explore what is known about social deprivation in autism. **Method:** Community and implications of findings. A literature search conducted in PsycINFO, EMBASE, Ovid MEDLINE, and CINHAL identified 3,146 studies. **Results:** Six studies met inclusion criteria. Five studies explored area-level social deprivation among children with autism the United Kingdom (UK). The sixth study explored a proactive intervention targeting individual-level term 'social deprivation' was found to be polysemous in both autism and deprivation literature, demonstrating the need for reporting clarity. Deprivation levels were found to be higher than the UK national average, where prevalence increased more dynamically in areas of highest deprivation. **Conclusions:** To advance research efforts, policy development, and resource allocation strategies, future studies should prioritize: 1) using social deprivation to demonstrate relationships between socioeconomic variation and health outcomes important to the autism community (e.g. social isolation); 2) including community colleagues to inform of current barriers; and 3) clearly describing or operationalizing social deprivation according to the level of observation (ie. area- or individual-level), population context (ie. general or specific to autism), and measurement instrument (ie. questionnaire or census-data).

Keywords: Autism spectrum disorders, school-aged children, social deprivation, scoping review, deprivation index, operationalization

Introduction

Autism spectrum disorder (hereafter 'autism' or 'ASD') is a set of early-onset and heterogeneous neurodevelopmental conditions, characterized by restrictive or repetitive behavioral tendencies and difficulties in social communication (APA, 2022; Lai et al., 2014). The worldwide prevalence of autism has risen from approximately 1 in 160 in 2012 (Elsabbagh et al., 2012) to 1 in 100 children in 2022 (Zeidan et al., 2022). While this may reflect the impact of considerable increase in autism research, it also points to a growing need of the worldwide population for specialized health services, guidance, and resources. Resources for autistic persons and their families are commonly provided by public health, education, and social services systems. The collaboration between researchers and policymakers facilitates the planning and implementation of research findings (Salari et al., 2022). However, several barriers to the provision of health services exist for this group, based on a combination of individual, family, and community or area-level factors, warranting the attention of researchers (Brisendine et al., 2017).

Studies have shown that the combination of barriers to care results in increased emergency service use, and risk of worsening of psychological outcomes and quality of life (Beverly et al., 2021). Social deprivation thus represents a multidimensional exposure involving area- and individual-level factors, the unique combination of which can diminish or augment quality of life and important social opportunities to long-term health outcomes. Access to services for autistic individuals often depends on an established diagnosis. The combination of heterogeneous developmental profiles in autism and multiple co-occurring conditions, such as mental health issues, hinders clear diagnostic categorization (Lai et al., 2014; Watson et al., 2011; Wei et al., 2018; Public Health Agency of Canada, 2022). For example, psychiatric conditions are reported by 70-75% of autistic children and adolescents (Guererra et al., 2022, Siminoff et al., 2008). As a result, a large proportion of children with autism have more complex needs, which can increase barriers to accessing care when services are dependent on a diagnosis, rather than a needs-based approach. (Kogan et al., 2008). The unmet need for socially focused services amplifies stress levels in children and families (Brown et al., 2011). The lack of these services can impact quality of life (Hodgetts et al., 2015; CDC 2022), mental health (Bauminger & Kasari, 2000; Bauminger et al., 2003), and interpersonal connections (Fletcher et al., 2012).

Barriers to Diagnosis & Treatment

Early identification of autism, particularly between birth and 2.5 years of age (Gabbay-Dizdar et al., 2022), is associated with improved outcomes in the child's social domains and quality of life (Van't Hof et al., 2021; Elder et al., 2017). Despite well-established diagnostic criteria, obtaining a diagnosis is often complicated by the heterogeneity of symptoms, co-morbidities, and availability of specialists in rural areas. The latter, also known as 'service deserts,' can result in delayed access to services and supports, with consequences for quality of life for the child and family (Dawson, 2008; Drohta et al., 2020; Gabbay-Dizdar et al., 2022;). It is well established that socioeconomic status (SES) influences opportunities for families to pursue health resources and social supports (Wang et al., 2021). Families with lower SES report poor access to and poor quality of services, lower quality of life, and reduced health service satisfaction (Dawson, 2008; Patten et al., 2012; Drohta et al., 2020), as not all families have equal access to diagnostic and continuous services and supports (Wang et al., 2021; Malik-Soni 2022). Further, autism occurrence is associated with high annual economic costs for both state-provided resources and out-of-pocket expenditures (Roddy et al., 2019). A study in Ireland found that, depending on autism severity, the total amount of annual funding provided by state budgets was surpassed by out-of-pocket expenses for caregivers (Roddy et al., 2019). Factors such as level of education, access to transportation, employment type, and stability of income were also found to impact the frequency and pattern of service use (Fountain et al., 2011), as low SES and high geographic distance from medical resources deters some families from seeking services (Ashburner et al., 2016).

To support autism community's right to health, researchers and clinicians would benefit from understanding the complex disposition, not only of autism symptomatology, but the socioeconomic pressures facing caregivers, which differ by individual or family (Nempewo et al., 2022). The combined caregiver-related demographic factors of living area, and social opportunities have been investigated under the construct of 'social deprivation,' which will be the focus this review.

Social Deprivation

To investigate the interrelatedness of population-based or area-level health outcomes associated with material and social opportunity, social deprivation indices (SDIs) were developed in many high-income countries (HICs) such as the United Kingdom (UK), the United States (US), and Canada. SDIs aim to quantify the area-based impact of socioeconomic factors such as employment, income, education, and marital status, which when weighted on a given index, provides a metric representing disproportionate health impacts to inform policymaking and resource allocation (Hales, 2003; Pampalon et al., 2012; Lai et al., 2014). The SDIs largely rely upon national census data with these weighted sociodemographic characteristics by area code to identify geographic areas of greatest need (Kephart & Asada, 2009). These measures demographic indicators, deprivation frameworks, and scale interpretations (e.g., high vs. low deprivation could mean a high or low score) depending on the region and context of the measure. Together, the factors have introduced a level of ambiguity social deprivation deprivation (Smith et al., 2020). Thus, given the differences between scales, regions, and included domains, we expect the included studies to have diverse findings.

Social Deprivation in Autism

In autism literature, the construct ‘social deprivation’ has typically been used outside the ecological contribution to area-level social determinants of health. It has been investigated in neurodevelopmental and psychological contexts, referring to a neurological stimulus deficiency in early development, or as an individual’s psychological experience of feeling socially excluded or isolated. Social isolation, or subjective feelings of loneliness are well documented in autism literature, and are seen to mediate socially and materially related domains in autism (e.g., social skill, attention, communication). Social isolation is also associated with increased unemployment rates and severity of mental health distress (e.g., anxiety, depression, and suicidal ideation) (Schiltz 2021; Umagami et al., 2022). In these ways, the area-level contribution of social deprivation on an individual or family’s experience of social opportunity thus represents an important gap which can be improved at health systems levels to advance the enjoyment of health rights for those in the autism community.

Operationalizing “social deprivation”

As the term ‘social deprivation’ is investigated in a variety of contexts in both deprivation and autism research, the operationalization of this review was informed by 1) the definition provided by the American Psychological Association, 2) definitions reported by developers of social deprivation indices, 3) descriptions from published literature, and 4) reports from our panel of community colleagues on conceptualization of social deprivation in real-world and research settings. Therefore, the term ‘social deprivation’ explored in this review will be defined as: the limiting of access to society’s resources (APA, 2022) and social opportunities identified by several family-level socioeconomic factors or individual characteristics in social, physical, and mental health contexts (Pampalon, 2012). Social deprivation can therefore be identified where area-level deprivation indices or measures capturing area-level contributors are explored, and where individual-level measures are used. At the individual-level, social deprivation includes contributions from area-level influences (e.g. distance from services, home conditions), and an individual's psychological experience of diminished of social opportunities (e.g. barriers to social developmental milestones, decreased social participation, mental health status). In order to include both contexts, intervention studies focused on the improvement of social deprivation were also included. Because this review focused on the area- and individual-levels of social deprivation among the autism community, other contexts of social deprivation autism-related research were importantly identified and distinguished by literature review. These contexts include psychology or neuroscientific studies that investigated causal factor identified in institutionally-reared (or socially deprived) children, where deprivation of social stimuli in early development results in pseudo-autistic features (Cepanec et al., 2010), which were not explored in this review.

Objectives

The overall aim of this scoping review was to explore what is known about social deprivation among individuals with autism. The specific aims of this review were to:investigate the operationalization of social deprivation

ii. describe social deprivation levels among individuals with autism.

4 iii. identify the measurement instruments used.

We expect the findings will highlight the conceptualization of social deprivation in autism research and the levels observed in autism populations, which will inform research contexts and clinical practice of environmental or contextual considerations. Additionally, identifying the ways in which social deprivation is operationalized and measured can facilitate the development of strategies to optimize future research efforts contributing to health systems organization, and policymaking for autism communities.

Methods

A scoping review is a form of knowledge synthesis that addresses an exploratory research question. It aims to map key concepts, types of evidence, and research gaps related to a defined area by systematically searching and synthesizing existing knowledge (Arksey et al., 2005). This design was chosen over a systematic review as this domain has not yet been explored in the autism population. The diversity of measures and definitions used identify social deprivation also presents a research gap best suited to the scoping review design. (Munn et al., 2018). This review used the methodological framework of Arksey and O'Malley which consisted of five steps: i) identifying the research question(s), ii) search strategy and selection criteria, iii) selecting/screening studies, iv) charting the data and v) collating, summarizing, and reporting results (Arksey et al., 2005). An additional step of stakeholder (hereafter 'community colleague') consultation was added to this review from Levac's framework (Levac et al., 2010). At each of the five steps, we sought community colleague consultation to identify knowledge gaps in research, knowledge translation, and real-world settings.

i) Identifying the Research Question

Our research questions were: 1) "What is known about social deprivation investigation among individuals with autism in published, peer reviewed literature? 2) How is social deprivation operationalized, what levels are reported, and what measures are used?

ii) Search Strategy and Selection Criteria

The first author, T.D., conducted an electronic search with the assistance of an academic librarian (J.B.) of four databases: Ovid MEDLINE (1946 – June 2022), CINAHL (1977 – June 2022), Ovid EMBASE (1974 – June 2022), and PsycINFO (1987 – June 2022). These databases were selected based on consultations with an expert librarian and their relevance to our research question and the keywords were selected with the help of the librarian, stakeholders' consultation, and literature review (*Appendix A).*

iii) Selecting and Screening Studies

Using the eligibility criteria (Table 1), the screening process was pre-tested on 30 articles in three random sets, each containing ten articles to ensure consistency and strong inter-coder agreement in applying selection criteria. There were five conflicts in the abstract and title screening, which represents 84% agreement. Conflicts were resolved by discussion with a third author, resulting in 100% agreement. This review was conducted using The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines with the scoping review extension (Tricco et al., 2018). The PRISMA flow chart was used to track the number of articles and exclusion reasons at each stage (Figure 1) (Moher et al., 2009). All articles from the search process were imported into EndNote and duplicates were manually removed. Two reviewers (T.D. and S.N.) independently screened the articles at the title and abstract, and full-text level. References of systematic reviews investigating social deprivation and health outcomes among pediatric populations were hand searched by the two reviewers to retrieve relevant articles. Conference abstracts were scrutinized to find relevant abstracts. Textbooks and gray literature were excluded due to inherent differences with research studies. Any discrepant, divergent opinion during the screening process was addressed by consultation with the research team. Non-English studies with an English abstract were included if they met the above criteria, and were translated by DeepL, which has demonstrated reliability in research settings (Takakusagi et al., 2021). Examples of eligible articles were those that mentioned ‘social deprivation’ by name, and reported on individuals of any age with either with an autism diagnosis (e.g. by clinical tool or self-identified), or were clinically identified as ‘at-risk’ of autism at an early age. Examples of ineligible articles were reviews or editorials; articles that explored another kind of deprivation (ie. neighbourhood deprivation, socioeconomic deprivation); or studies that explored outcomes which did not directly measure outcomes of individuals with autism (e.g. caregivers of children with autism).

Search Terms

Search strategy is detailed in Appendix A. Examples of search terms were ‘social deprivation’ ‘autism spectrum disorder OR Asperger’s Syndrome’, ‘Health Status Disparities’ and a number of named measures and deprivation indices. We reviewed specific social determinants of health factors, and through our research, we refined that list according to the extent of heterogeneity of factors required to answer our research questions. The inclusion of the term ‘social determinants of health’ was not included for this reason, as well as by recommendation of our community colleagues and experts.

Table 1: Inclusion and Exclusion Criteria

<u>(Insert Table 1)</u>

iv) Charting the Data

Using Microsoft Excel, a predefined data extraction form was created, reviewed, and finalized by all authors (Table 1). The following information for each included article was extracted independently by two authors (T.D. and S.N.): first author, year, study aims/objectives, study setting (including country and design), sample size, sample characteristics, index of social deprivation, and key findings pertaining to our research questions (Table 2).

v) Collating, Summarizing, and Reporting Results

Once data extraction was complete, findings were synthesized independently by both authors according to two steps: (1) reporting the findings, and (2) discussing the implications. The study purpose and major findings related to social deprivation were the primary units of analysis. No qualitative articles captured by the search strategy. Quantitative analyses were conducted to describe the study designs, populations, measurement tools and social deprivation levels. An inductive thematic analysis was used to investigate the use and operationalization of social deprivation, as this term has been found to be represented by a variety of definitions, indicators, indices, and settings depending on the context of study.

vi) Community Colleague Consultation

Community colleagues' consultation was added from Levac's guidelines (Levac, 2010) to provide key perspectives to strengthen the review's applicability to the autism community. To reflect on recent recommendations, we used the term "community colleagues" to refer the stakeholders (CDC, 2022). Therefore, we included community colleagues in the childhood disability and autism community at the inception of this project to inform of the conceptualization of social deprivation impact in autism, and knowledge gaps in research and real-world settings.

Four community colleagues were included: 1) an expert librarian who contributed to the methodological development and translation of the research question and search strategy; 2) a front-line community support coordinator for families and children with autism who contributed to the objectives, terms used by families seeking support, inclusion criteria, implications of the findings, and knowledge translation (KT) strategies impactful in community services; 3) a researcher in policymaking and Chair of Diagnosis, Supports, and Services for Autism at the Canadian Academy of Health Sciences (CAHS), who contributed to the research question, selection criteria, the use of language endorsed by the National autism strategy; 4) and a knowledge translation (KT) expert and member the Centre of Interdisciplinary Research in Rehabilitation (CRIR), who contributed insights on KT strategies based on our findings. Between 2-5 virtual meetings ranging between 30-90 minutes were conducted with each community colleague between January 2022 and October 2023. The first author annotated each meeting to summarize recommendations and observe common themes. Community colleagues were asked about their experience and understanding of social deprivation within the autism community, as well as the practical impact and relevance to future research. Of the four community colleagues, two were caregivers of a child with autism, and thus provided first-hand insights describing the lived-experience of participating in their child's care, and witnessing of social deprivation with their children. These insights were important to the inclusion/exclusion criteria and interpretation of results. They also helped address the gap in community-academic partnerships (Meza et al., 2016), as community colleagues voiced practical gaps in representativeness of and research pertaining to social deprivation in the autism community. As participation of community colleagues within the autism community such as caregivers help Examples of recommendations included key words to refine search terms, advocacy to include intervention studies, current consensus on preferred vernacular, KT strategies optimal for families and health practitioners, and negative health impacts where social deprivation is reported in support-related settings, such as diminished mental health, social isolation, increased anxiety, depression, and emergency service use.

Figure 1: *PRISMA Flowchart of all articles captured, screened, excluded, and included.*

(Insert Figure 1)

Results

Search Strategy

searching. After removing duplicates, 3,145 records were reviewed by title and abstract, where 3063 (97.36%) records were excluded. The most common reason for exclusion at this stage was a lack of reporting on social deprivation (n=2366, 77.2%). The remaining 82 records were screened at the full text stage, where 76 records (92.6%) were excluded as they did not describe social deprivation. See Figure 1 for a description of selection decisions.

Study Selection

A total of six records published between 2000-2020 were included (see Table 2). Social deprivation was found to be investigated only among child and adolescent populations, ages 0-18. Five of six studies were conducted in the UK (England, Northern Ireland) and were reported in English. One study was conducted in France and was translated from French to English. The total number of participants with a primary diagnosis of autism across included studies was 18,066. The included articles comprised of three cohort studies (Powell et al., 2000; Warnell et al., 2015; McConkey 2020), one prospective study (Schroder et al., 2015), one prospective data registry (McConachie et al., 2008), and one cross-sectional study (Griffith et al., 2011).

Table 2: *Summary of Included Studies*

(Insert Table 2)

Social deprivation operationalization

Direct operationalizations were notably missing across studies. However, two studies described the construct according to one population- and one individual-level context. Griffith et al. 2011 described social deprivation as an effect that can be quantified using the Index of Multiple Deprivation, which ranks deprivation levels in small neighborhood areas based on census data variables of employment, income level, crime, and education (Griffith et al., 2011). Schroder et al. (2015) investigated an intervention called the Early-Start Denver Model (ESDM), which indirectly conceptualized social deprivation in an autism-specific deficit of social attention, a context observed only among autistic individuals. The remaining four studies indirectly conceptualized the construct by use of the term "social deprivation" in tandem with a report of a population-level measurement instrument or public data source.

Table 3: *Operationalization of Social Deprivation and Context of Measurement*

(Insert Table 3)

Social deprivation levels in autism

Social deprivation levels on area-level indices among autistic children were most frequently higher than the national average in many areas of the UK (McConachie at el., 2008; Warnell et al., 2015; Griffith et al., 2015) (see Table 3). Incidence was not found to be associated with social deprivation (Powell et al., 2000). Griffith et al. (2015) found that 39.6% of students attending ABA therapy in public schools lived in areas of the higher deprivation quartiles (areas of the most socioeconomic disadvantage), while 50.4% of children lived in areas with lower deprivation (or areas of least socioeconomic disadvantage). McConkey et al. (2020) grouped autistic individuals according to students living in areas of 30% greatest and least deprivation between census data from 2010/2011, and 2018/2019. They found that prevalence rates in affluent areas of Northern Ireland saw the lowest increase (1.49% to 2.95%). Conversely, significant associations were found between the increased prevalence of autism in socioeconomically disadvantaged areas, (1.44% to 3.78%) where families in the region with the highest level of social deprivation or disadvantage (e.g. Belfast) saw the steepest increase (1.72% to 6.21%).

Social Deprivation Measurement

Three published area-level measures were used to identify social deprivation among four studies (see Table 2 & 4). Each measure was specific to populations in the UK and included a different number of demographic domains. Two studies used the Townsend Deprivation Index (McConachie et al., 2009; Warnell et al., 2015), Griffith et al. (2011) used the Index of Multiple Deprivation (IMD, 2010) (Griffith et al., 2011), and McConkey (2008) used the Northern Ireland Multiple Deprivation Measure (NIMDM, 2017). One study reported only their census data source and deprivation score from an unspecified index. Each index provided a slightly different scale, number of domains, and difference in interpretation (see Table 4).

Table 4: *Social Deprivation Measures and Interpretations*

<u>(Insert Table 4)</u>

Discussion

This scoping review aimed to explore what is known about social deprivation in the context of autism, specifically regarding the operationalizations reported across studies, the levels of deprivation reported in autism samples, and their measures. Social deprivation was found to be a multidimensional construct that requires further clarity and investigation in autism research. The first and most notable finding was that most studies inferred the operationalization of social deprivation through their choice of measurement instrument and background, rather than using a direct operationalization. However, two important contexts were reported. In addition to the area-level context quantifying socioeconomic distribution described by Griffith et al. (2015), Schroder et al (2015) explored social deprivation at the individual level in an autism-specific context. This conceptualization included the more commonly described attributes of being 'socially deprived' or having limited social opportunity at the individual, which identified a vulnerability to social attention deficits to which autistic children are hypothesized to be predisposed. This study did not include a direct report on social deprivation level, and instead used social behaviours and severity of autism symptomatology as proxy to determine social deprivation levels and was therefore not included in the results section. However this context has not previously been distinguished in social deprivation literature and identifies a knowledge gap in where distinctiveness of terminology could create barriers in autism-specific contexts. These findings contribute clarity regarding the ambiguity of using term 'social deprivation' in both clinical and research settings, and among autistic and typically developing populations. This review suggest that future studies include a direct operationalization of social deprivation that (e.g., general population, or specific to autism); and 3) the measurement instruments with their included domains used to observe its effects.

Secondly, across all autism samples included in this review, levels of social deprivation were identified in children and youth to be higher than the national average in the UK. Where health outcomes of autism occurrence were investigated, McConkey (2020) reported the largest sample size and most recent data collection (2010-2019), finding areas with highest levels of social deprivation in the UK had the most marked increases in prevalence of autism. Regarding government-funded support and allocated to education settings in the UK, children living in more disadvantaged areas—where families rely most on therapy at no additional cost— were found to comprise the minority of the children who received it (Griffith et al., 2015). Despite the low number of studies identified, these findings did identify the disparity of increased social deprivation and gap in ABA therapy offerings within the autism communities of the UK. What remains to be seen are how these data are put to practical use to inform health systems planning to improve health outcomes such as mental health status, emergency service use, or service accessibility. The absence of these outcomes within autism literature was an unexpected finding, given 1) the high rate of psychiatric co-morbidities present among autistic individuals (Guererra et al., 2022, Siminoff et al., 2008); 2) reports of service inaccessibility and mental health severity impacting emergency service use among autistic individuals (Liu et al., 2017; Beverly et al., 2021); and 3) that social deprivation measures were developed for the purpose of quantify these area-level disparities to facilitate health systems planning (Pampalon et al., 2009; Social Deprivation Index, 2018).

Finally, social deprivation was most frequently investigated using an area-level index which weighted various demographic factors within small areas of the UK (see Table 4). describes the measures used in each study and their interpretations. Despite the presence of and many other HIC's, area-level social deprivation was found only to be investigated in the UK. There was no 'gold-standard' instrument, although there were more commonly known indices (e.g., TDI, 2009). Studies explored area-level health outcomes of representativeness of academic participation, sample recruitment, and epidemiological trends.

Intervention studies were included by the recommendation of community colleagues, who highlighted the importance of considering structural and contextual factors related to caregivers, such as mental health distress and lack of proactive interventions, as health outcomes observed with greater socioeconomic disadvantage (e.g., experience of loneliness, increased psychiatric emergency service use) are not limited to the context of autism symptomatology alone. As social and material opportunities of the caregiver are associated with mental health status for both the child and parent (Bambra et al., 2014) colleagues recommended inclusion of resources that proactively increase education and support strategies for children and their caregivers prior to emergency or distress. To that end, the EDSM intervention investigated by Schroder et al. (2015) was included as it demonstrated effectiveness in these areas, developing skills to mediate individual social deprivation impacts in a small sample of toddlers. The finding of early-intervention improvements in autism is consistent with reviews on early intervention studies, where interventions provided at the earliest developmental stages demonstrate high efficacy in reducing socially-related barriers for children with autism throughout development (Andanson et al., 2012).

Consistency of Findings with Existing Literature

Where other area-level deprivation indices are explored in autism, high levels of deprivations are observed alongside prevalence estimates, where multiple studies report that 1) autistic individuals are more frequently found to have higher deprivation levels compared to non-autistic controls, and 2) high deprivation levels were associated with increased prevalence of autism (Li et al., 2014; Delobel-Ayoub et al., 2015; McGuinn et al., 2019). Interestingly, the lack of association between social deprivation status and incidence found by Powell et al. conflicted with a 2014 study where incidence of childhood autism was associated with area- (or neighbourhood) level deprivation (Li et al., 2014). This is likely due to differences in sample size and year of data collection.

One possible reason for the finding of increased prevalence of social deprivation in the autism cohort reported in McConkey (2020), is the release of the Diagnostic and Statistical Manual for Mental Disorders, 5th Edition (DSM-V) in 2013 (APA, 2013), where revisions were targeted toward improving diagnostic accuracy with respect to criteria for autism (American Psychiatric Association, 2013). The DSM-IV (1994) included a group of disorders called "Pervasive Developmental Disorders" (PDD) as the umbrella diagnosis related to neurodevelopmental and social communication disorders. This diagnosis included pervasive developmental disorder - not otherwise specified (PDD-NOS), social communication disorder (SCD), autistic disorder, Asperger's disorder, and childhood disintegrative disorder (American Psychiatric Association, 1994), in addition to early infantile autism, childhood autism, Kanner's autism, high-functioning autism, atypical autism (American Psychiatric Association, 2013). The DSM-V recategorized the above diagnoses to fall under Autism Spectrum Disorder (American Psychiatric Association, 2013; Kim et al., 2015), stating that the factors influencing the rise in prevalence of autism to be 'unclear', as the increased rate could be reflective of the "expansion of the diagnostic criteria of DSM-IV to include subthreshold cases, increased awareness, differences in study methodology, or a true increase in the frequency" (American Psychiatric Association, 2013).

A 2015 study by Kim and colleagues explored these factors by comparing the prevalence estimates in a sample of over 55 000 Korean children aged 7-12 previously diagnosed with PDD-NOS and SCD before 2013, with ASD prevalence estimates after the implementation of the

DSM-V. They found prevalence rates to be “virtually the same,” when accounting for the prevalence rates in the 2013, indicating frequency of autism diagnosis did increase, but it was due to the recategorization of PDD-NOS and SCD (Kim et al., 2015). As the study by McConkey (2020) includes census data from 2010/11 to 2018/19, the timing of this study could influence interpretation of findings in the following ways: 1) the specific area Northern Ireland proportion of children requiring ABA therapy in school settings were among the lower proportion of those who received it; 2) that the autism rate did not increase, but the recategorization of diagnostic criteria brought forward children previously not identified prior to the 2010/11 census who required autism-specific therapy. In either case, this study identified that children with the diagnosis of autism had differing levels of opportunity to receive therapy, depending upon their social deprivation level. This provides a basis to evaluate the distribution inform the education offerings for students, and future policies.

The ambiguity of reporting and lack of distinct operationalization of deprivation terminologies in observed in this scoping review has been a longstanding criticism of deprivation literature beginning in 1995. Gordon (1995) identified a fundamental lack of clarity between indices and definitions, where terminologies were used “loosely…with little reference to their technical meanings” (Gordon, 1995). In research contexts, social deprivation is used interchangeably with many other area-level constructs within deprivation literature. Reviews in other diagnostic categories have echoed this finding, identifying construct ambiguity to have introduced “a level of dubiousness” (Smith et al., 2021) in the selection, establishment, and interpretation of deprivation indices as distinct constructs. The differential inclusion of socioeconomic domains also limits the systematic comparison of this deprivation measures across studies. To reduce this barrier, studies have studies suggest establishing a core set of socioeconomic domains (Smith et al., 2021) to represent this construct.

Implications

One important implication of this review from the area-level perspective is the advantage of investigating social deprivation in academic settings. A considerable proportion of children were diagnosed after starting school (McConkey, 2020; Dillenburger et al., 2015) which implicates academic settings as an important aspect of accessibility to diagnostic services. Despite inherent restrictions of school census on providing descriptive data, this could be a more feasible avenue of data collection. In a broader lens of public health, inclusion of social deprivation measurement in academic or data registry contexts would provide a clearer and current picture of service need and distribution in autism, especially for minority ethnic groups and socially disadvantaged children (Roman-Urrestatazu et al., 2022). To that end**,** the use of publicly available data to support policy development has been impactful in other contexts, where studies in cerebral palsy used this approach to demonstrate associations between level of deprivation and quality of mobility outcomes. Oskoui et al. (2016) were the first to raise the issue of how social-material deprivation may be related to CP mobility status and leisure participation, which has initiated service allocation considerations to guide policy development (Mogo et al., 2020).

Regarding implications in clinical contexts, increased clinical knowledge of the multiple context-dependent meanings of social deprivation could be more easily clarified by discussion. As social deprivation could mean an individual level of social distress in contexts specific to autism, or an area-level impact reducing service accessibility, discussion of these differences in clinical settings could facilitate communicative clarity and the applicability of subsequent recommendations. Secondly, clinical knowledge of domains involved in area-level deprivation could optimize contextual factors related to treatment feasibility and uptake. For example, considerations of employment type and job stability of the caregiver, in conjunction with area code and transportation access are useful when strategizing patient-centered treatment options supporting the unique needs of a family or individual. Telerehabilitation is a prime candidate in this respect, where multiple autism interventions have demonstrated being as or more effective than face-to-face treatment when administered through telerehabilitation (Ogourtsova et al., 2021; Ogourtsova, 2023). Application of telerehabilitation is shown to reduce barriers of time, financial costs, and psychological stress for caregivers and their children, and increase accessibility to services (Ogourtsova et al., 2021).

Excluded studies

Although some included studies used the same measurement instruments as excluded studies, many records were excluded because they did not explore social deprivation by name. This identified an important gap in research reporting styles, where vocabulary is used interchangeably and obscures distinction between constructs. Future studies aimed to capture the wider variety of social and material deprivation are recommended to use ‘area-level deprivation’ to identify studies measuring deprivation using area-based terminologies (see Appendix C). This approach would allow for the comparison of multiple area-level deprivation frameworks and include a wider variety of health outcomes.

Community Colleague Contribution

The participation of community colleagues was a crucial aspect of this scoping review which strengthened the relevance, clarity, and applicability of our findings. Community colleagues made important contributions to elucidate the gap between research efforts and the impacts of social deprivation in real-world settings of support coordination. In the same way that research in social deprivation included interchangeable terms for social deprivation at the area- and individual-levels, colleagues reported the same inconsistencies in clinical settings, where caregivers use interchangeable terms with clinicians, resulting in ill-fitting treatment strategies. This knowledge barrier between caregivers and clinicians would be improved by including education interventions in knowledge translation such as webinars or posters, which present clarifications of terminologies used (e.g. social deprivation vs. social isolation) and the intended contexts (e.g. area-level access to services vs. mental health).

Advocacy of Language

The term 'deprivation' in socioeconomic context may be interpreted as a derogatory inference, negatively highlighting disadvantaged communities, which is not the aim of its use. This review advocates for researchers, clinicians to consider the many interrelated factors involved in social deprivation for the purpose of large-scale health systems intervention. The purpose of using this term is not to identify families as 'deprived', but to promote awareness of potential risk factors that impact the autism community to inform clinical practice and policy. Social deprivation in this sense can be understood as the vulnerability of opportunity or level of precariousness inherent to a set of social and material circumstances.

Limitations

The limitations of this scoping review were firstly in establishing a comprehensive and distinct definition of social deprivation, and secondly in the low number of studies identified.

The process of developing a rigorous and comprehensive search strategy was extensive and required multiple iterations with community colleagues to establish distinctiveness and accuracy.

The use of interchangeable Index of Multiple Deprivation terminologies across both autism and deprivation literature may explain the low proportion of studies fitting inclusion criteria, which limit the generalizability of our findings.

Conclusion

This review is the first to explore social deprivation in autism. Social deprivation was conceptualized as a construct that observed the impacts of numerous social and material factors on health outcomes and levels in autism samples. Importantly, this is also the first review to report the distinction between general population-level and autism-specific social deprivation. As higher-than-average levels of social deprivation were reported for most of the total autism sample in the UK, further investigation is warranted in countries with established indices and available population-level data, to identify the contexts in which this is most impactful. Investigating health outcomes highlighted by community colleagues such as service accessibility, social isolation, and mental health distress are recommended to advance the quality, distribution, and applicability of services. To optimize clarity of social deprivation in research, future operationalizations are recommended to include a description of the level of observation, the population context, and the measures used to capture its effects.

Declaration of Interests

All authors declare no known conflicts of interest.

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Appendix A: Search Strategy created in MEDLINE-OVID

1. Social Deprivation.mp.
2. Psychosocial Deprivation.mp.
3. Social experien* deprivation.mp.
4. ((economic or socioeconomic) adj2 (factor* or deprivation or disparit* or difference*)).tw,kf.
5. ((social or material) adj2 (disparit* or deprivation or marginalization)).tw,kf.
6. ((Area-level or district or neighborhood or neighbourhood) adj2 (disparit* or deprivation)).mp.
7. individual-level deprivation.mp.
8. Health Status Disparities/ or Socioeconomic factors/ or economic factors/
9. social exclusion.mp. or Social Isolation/ or social isolation.tw,kf. or social environment/ or social environment.tw,kf.
10. Mexican Marginalization Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
11. Namibian Indices of Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
12. South African Indices of Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
13. Multidimensional Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
14. Area Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
15. Social Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
16. Neighbourhood Equity Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary

concept word, rare disease supplementary concept word, unique identifier, synonyms]

17. Ontario et Canadian Marginalization Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
18. Canadian Marginalization Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
19. Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
20. (The Material and Social Deprivation Index).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
21. The Canadian Index of Multiple Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
22. Pobal Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
23. All Island HP Deprivation Index.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
24. Northern Ireland Multiple Deprivation Measure.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
25. Scottish Index of Multiple Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
26. Welsh Index of Multiple Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword

heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]

27. (Index of material and social deprivation English Index of Multiple Deprivation).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
28. Carstairs.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
29. Townsend Material Deprivation Score.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
30. Jarman score.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
31. Index of Multiple Deprivation.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
32. Autistic Disorder/ or exp Autism Spectrum Disorder/ or Asperger Syndrome/ or autism.mp. or autistic.mp.
33. or/1-31
34. 32 and 33

Appendix B: Area-Level Deprivation Indices identified among excluded studies

Indices found to be used in area-level deprivation contexts:

1. Canadian Marginalization Index / Canadian Index of Multiple Deprivation
2. Carstairs Deprivation Index
3. English Indices of Deprivation
4. Index of Multiple Deprivation
5. Index of Socioeconomic Deprivation
6. Material Deprivation Index
7. Material and Social Deprivation Index
8. Multidimensional Deprivation Index
9. Neighborhood Deprivation, using SAMS (small area market statistics which are geocoded areas in Sweden)
10. The Neighborhood Deprivation Index
11. The Northern Ireland Multiple Deprivation Measure
12. Scottish Index of Multiple Deprivation
13. The Social Exclusion Index
14. Socioeconomic Status Index
15. Townsend Deprivation Index

Appendix C: Deprivation Keywords According to Context of Use

Deprivation as an Area-Level Exposure:
1. Area (or area-level) deprivation
2. Deprivation (used alone)
3. Material deprivation
4. Multidimensional deprivation
5. Neighbourhood/neighborhood deprivation
6. Socioeconomic deprivation
7. Economic deprivation
8. Social and environmental deprivation
9. Social and material deprivation

Deprivation as Stimulus Deficit:
1. Cultural Deprivation
2. Emotional-social deprivation
3. Institutional deprivation
4. Psychosocial deprivation
5. Sensory Deprivation
6. Sleep Deprivation
7. Social-Experiential Deprivation

Related but distinct terms used to describe effects of social deprivation.
- Social isolation
- Social participation
- Social exclusion
- Social inclusion

Figure 1: Flowchart of all articles captured, screened,excluded, and included

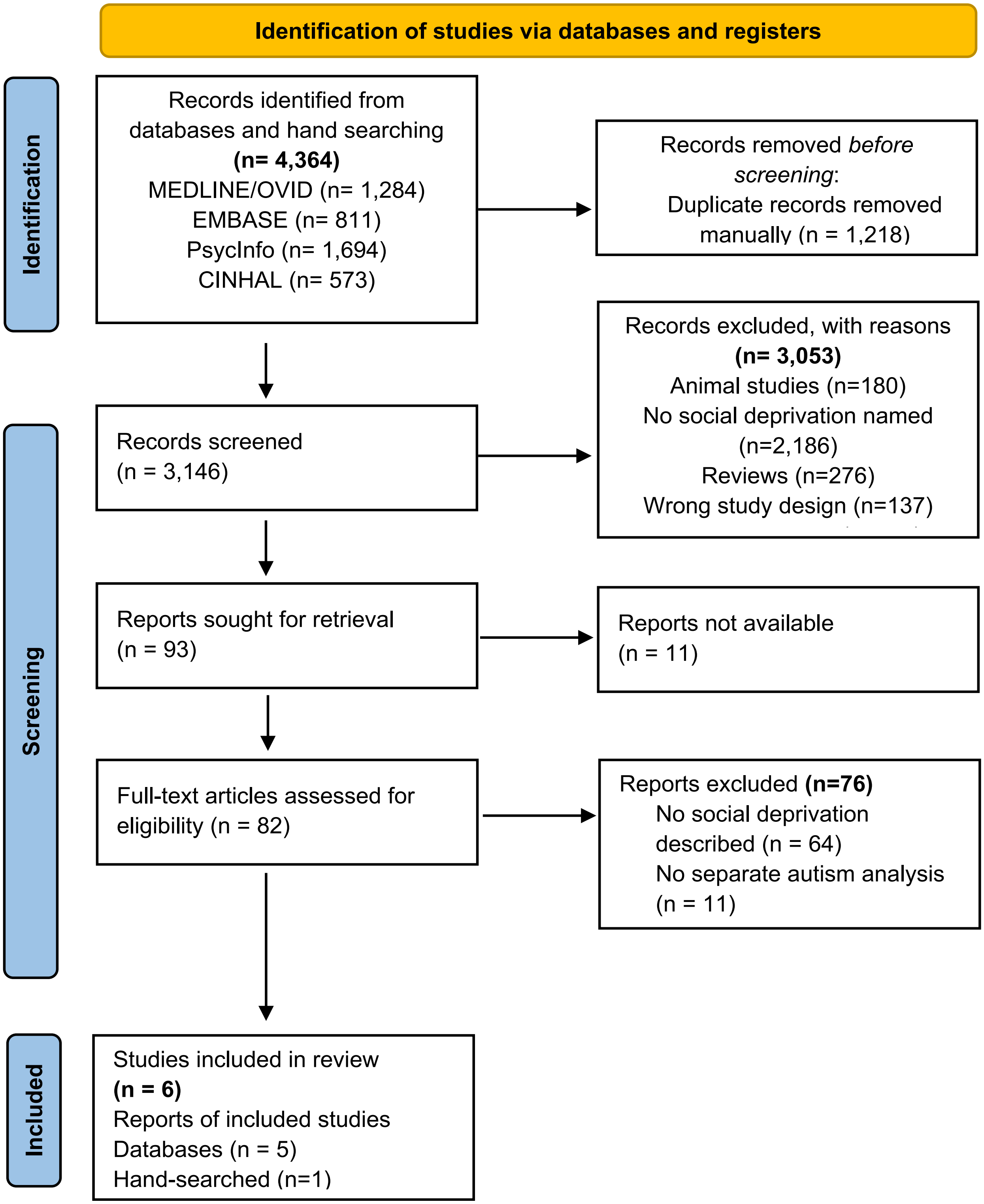

Table 1: Inclusion and Exclusion Criteria

Selection Criteria Domains	Inclusion criteria	Exclusion criteria
Participants' age and gender	- Any age - All gender categories	None.
Autism diagnosis	- Clinically confirmed at-risk of autism, or diagnosed with autism or Asperger's based on DSM-V, ADOS-2, ADI-R Or - Self-reported, self-identified - Studies must have analysis with statistics representing autism diagnoses only	- Studies that group multiple diagnoses with autism with no separated analysis - Studies that explore diagnoses not specified to be autism (e.g., intellectual disability, Rett Syndrome, Pervasive Developmental Disorder, Conduct Disorder)
Study design	- Quantitative And/or - Qualitative designs	- Reviews (e.g., systematic review, scoping review) - Case series/reports - Editorial/ Letter to editor
Language	English or provided English translation of abstract	Studies that did not provide English or French translation
Social deprivation	- Reports with established deprivation indices - Interventions evaluating effects of social deprivation	- Constructs with no data, or report on social deprivation; studies with no use of the term 'social deprivation'

Table 2: Summary of Included Studies

First author (Year)	Index of social deprivation	Key findings
Powel J.E. (2000)	Not reported	Incidence rates were similar in areas, despite differences in social deprivation and proportions of ethnic minorities. The incidence rate in South Birmingham (high ethnic population) was similar to that in Tamworth (small proportion of ethnic residents).
McConachie H. (2009)	Townsend deprivation Index	Social deprivation was significantly greater in non-responders (p<0.001); however, the difference was small (mean 1.77 vs 0.45 for non-responders vs responders, on a scale from 10 (very deprived) to -6.
Griffith G.M. (2012)	Index of Multiple Deprivation (IMD)	Schools offering ABA therapy showed a reasonably even distribution of children living in the lowest two quartiles (more deprived: 39.8%) and the highest two quartiles (affluent localities: 50.4%).
Schroder C.M. (2015)	Proxy measure testing skills in social attention and motivation	1. A highly significant increase in cognitive and socio-emotional abilities 2. A significant improvement in the development of pivotal functions e.g., joint attention, expressive and comprehension language and vocal and 3. A positive effect in operational causality, spatial relations, and action patterns. 4. A reduction in heterogeneity of skills between the cognitive and socio-emotional domain, with significant reduction after one year of care for socio-emotional domain
Warnell F. (2015)	Townsend deprivation index	Lower mean of Townsend score for consenting families compared to non-consenting families with ASD (0.86 vs. 1.61)
McConkey R. (2020)	The Northern Ireland Multiple Deprivation Measure 2017	For pupils living in more deprived areas, the rise in prevalence was more marked (from 1.44% to 3.78%) than for those in less deprived areas (1.49% to 2.95%). The prevalence of autism among the more deprived pupils rose from 1.72% to 6.21%.

Table 3: Operationalizations and Measurement Context

Measurement Context	**Study**	**Operationalization of Social Deprivation**	**Social Deprivation (SD) Context of Measurement**
Evaluation of Intervention	Schroder et al., 2015	"The ESDM aims to decrease the child's 'social deprivation' and its perpetuating impact on the neuronal and psychological development, to rekindle the altered developmental processes during this period of maximal brain plasticity."	SD measured indirectly by social and behavioural skill development increase and autism severity decrease
Occurrence of Autism (Incidence, Prevalence)	Powell et al., 2000	<u>No operationalization or index reported.</u> Area-level proportion of deprivation assumed based on description of data source (Office of Population Censuses and Surveys) and region-based reporting style.	SD measured as a factor in the rate of incidence of autism based on national surveys
	McConkey, 2020	Socio-economic deprivation based on the Northern Ireland Multiple Deprivation Measure 2017	SD measured as a factor in prevalence of autism
Distribution and Representativeness	Warnell et al., 2015	Social Deprivation as measure and described by the Townsend Deprivation Index	SD measured as a factor in representativeness of autism diagnosis between two research databases
	McConachie et al., 2008	Social Deprivation as measure and described by the Townsend Deprivation Index	SD measured as
	Griffith et al., 2011	"[The Index of Multiple Deprivation] IMD ranks indicate the level of social deprivation in the neighborhood based on a variety of UK census data variables (such as rates of local employment, income levels, crime, and education)."	SD measured to observe if area or region of a school influences the distribution of ABA therapy, along with associated outcomes

Table 4: Social Deprivation Measures and Interpretation

Measure	# of Domains	Domain Categories	Interpretation	Scale Range (Minimum to Maximum)	Used in
Townsend Deprivation Index	4	1. Unemployment 2. Non-car ownership 3. Non-home ownership 4. Household overcrowding	Greater deprivation = **greater positive score; average deprivation level is 0** (UK Data Service, 2022)	-6 to 13	McConachie et al., 2009; Warnell et al., 2015
Index of Multiple Deprivation (IMD) 2010	7	1.Income 2. Employment 3. Education 4. Health 5. Crime 6. Barriers to Housing and Services 7. Living Environment	Greater deprivation = **lower rank** (Ministry of Housing, 2019)	0 to 32,482	Griffith et al., 2011
The Northern Ireland Multiple Deprivation Measure (NIMDM) 2017	8	1. Income, 2. Employment 3. Health and Disability, 4. Education 5. Skills and Training 6. Access to Services, 7. Living Environment 8. Crime and Disorder	Greater deprivation = **lower ranking** (Northern Ireland Statistics and Research Agency, 2017)	1 to 890	McConkey, 2020

CHAPTER 4: BRIDGING TEXT

4.1. How did the scoping review answer the research question?

To answer our first research question about population-level impacts of SDOH specific to social deprivation in ASD, we found that few studies investigated this construct, but those that did find that higher social deprivation levels were significantly associated with increased prevalence of ASD, and the availability of publicly-offered therapy. Social deprivation levels also tended to be higher in the cohorts with ASD diagnosis, representing more precarious living areas and socioeconomic circumstances. Autism-specific therapy was also less available to autistic children living in higher deprivation areas. On both counts, this implicates intervention at the policy or health systems level in the United Kingdom to address area-level disparities. Although measures were in place to identify social deprivation as a risk factor in school settings, no studies explored mental health areas such as social isolation, depression, or anxiety according to the social deprivation area, which according to the stakeholders included in our scoping review, is a critical area of need in the ASD community and remains to be addressed.

4.2. How did findings inform the second manuscript?

By exploring social deprivation measures, findings relevant to the second manuscript were the convergence of overlapping domains used across included indices. Although core domains representing social deprivation are not yet established in literature, which is a common criticism which limits comparison across contexts (Smith et al., 2020), the most frequently used domains common to social deprivation were identified. To answer the remainder of the primary research question about individual-level SDOH, the second manuscript was informed by the findings from the first manuscript, and thus incorporated the following elements:

1) Frequently overlapping domains of caregiver education level, employment type/income level, living conditions, and marital status were identified in the AMC registry data set. The attributes of the caregiver were used to create an index which intended to represent the child's level of support in their given environment.
2) Clear operationalization of constructs and their expected relationships were included.
3) The addition of caregiver stress and child mood/mental health were included in the extraction of the subset of AMC registry data, as stakeholders from the first manuscript voiced a knowledge gap in research where social or mental health outcomes were not included, despite being highly impactful to every-day living conditions.

The impact of disability on caregiver resources and mental health was a peripheral finding of the scoping review. However, this is useful in the AMC registry project as the data collected included one item to measure frequency of psychosocial stress on a 5-level scale (Never, rarely, sometimes, frequently, always): *"Do you feel stressed between caring for your child and trying to meet your other responsibilities for your family and/or work?".* In cerebral palsy, an early-onset neuromuscular condition, caregiver mental health status and perception of stress are associated where high stress was observed alongside high levels of depression and anxiety among caregivers and diminished social support (Kouther et al., 2022). In clinical contexts, caregivers often have the lion share of responsibilities (e.g. physical, social, and material support for the child, in addition to organizing and attending appointments, diminished work opportunity, diminished frequency of social relationships), with little energy or resources to pursue social supports for themselves, resulting in higher levels of loneliness and depression (Yang et al., 2022). This context will be incorporated in the next manuscript, to understand the extent to which psychosocial wellbeing of caregivers is an influence, or is influenced, by their child's condition.

4.3. What will be explored in Manuscript 2?

In this thesis there are two contexts of SDOH: the population-level, which includes social deprivation and area-related health differences, and the individual-level, which includes a child's caregiver(s) and their social and material characteristics. The first manuscript described the context of SDOH within an invisible disability that is more commonly diagnosed. The second manuscript compared the importance of individual-level SDOH with personal factors in a visible, rare disease population. As there were numerous factors and associations to measure, structural equation modeling (SEM) was used to answer the remaining research questions about the influence of individual-level SDOH in AMC, and the strongest factors associated with functioning important to children with AMC and their caregivers.

CHAPTER 5: MANUSCRIPT 2

Title: Using a social deprivation perspective to investigate functional outcomes in children with Arthrogryposis: A structural equation modeling analysis

Authors: Tessah J. Dunn, Nancy Mayo, Reggie Hamdy, Haluk Altiok, Lauren Hyer, Michelle A. James, Sarah Nossov, Ellen Raney, Frank Rauch**,** Laurie Snider, Noemi Dahan-Oliel

Abstract

Background: Social determinants of health (SDOH) have been increasingly implicated as non-medical factors associated with adverse health outcomes. Social deprivation is one construct that clusters environmental factors (e.g. caregiver employment, education, marital status) to quantify population-level health outcomes. The International Classification of Functioning and Disability (ICF) framework was applied to the Shriners Arthrogryposis Multiplex Congenita (AMC) registry data. The aim of the study was to estimate the direct and indirect effects of environmental and personal factors on functional outcomes of individuals 8-21 with AMC.

Methods: A cross-sectional analysis was carried out on a cohort of 189, with environmental factors related to the SDOH of their caregivers. Functional domains included standardized measures on the individual's physical, psychosocial, and cognitive function, and parental stress. Structural equation modelling (SEM) was used to describing the direction, strength, and effect of these factors on functional domains.

Results: Only personal factors were associated with the child's health outcomes and parental stress. Type of AMC involvement and biological sex demonstrated a significant effect on pain and peer relationships. Pain exhibited the strongest effect on child health perception. The level of physical function was associated with parental stress, where caregivers reported more stress when their child had more physical limitations.

Conclusions: In the context of AMC, personal factors more important to health outcomes than environmental factors. Clinicians can use these results to inform treatment strategies involving caregiver psychosocial health where greater physical limitations are present, and identify contributing factors to improve peer relationships, pain, and health perception among children with AMC.

Keywords: social determinants of health, AMC registry, cross-sectional design, function, arthrogryposis multiplex congenita, musculoskeletal conditions, rare disease research

Abbreviations:

APPT – Adolescent Pediatric Pain Tool
AMC – Arthrogryposis multiplex congenita
CFI – Comparative Fit Index
CNS – Central nervous system
CG – Caregiver
ESI – Environmental support index
EQ-5D-Y – EuroQOL - 5 Dimensions – Youth version
FAQ – Gillette Functional Walking Subscale
ICF – International Classification of Functioning and Disability
ICF/WC – International Classification of Functioning and Disability with Wilson-Cleary extension
IRB – Institutional Review Board of Research Ethics
MLE – Maximum Likelihood Estimation
PROMIS-SF – Patient-Reported Outcomes Measurement Information System – Short Form
RMSEA – Root Mean Square Error of Approximation
SDOH – Social determinants of health
SD – standard deviation
S.E. – Standard Error
SEM – Structural equation modelling
SRMR- Standardized Room mean square residual
TLI – Tucker-Lewis Index
WeeFIM – The Functional Independence Measure for Children
VAS – Visual analogue scale

Introduction

Arthrogryposis Multiplex Congenita (AMC) is a group of rare conditions affecting mobility and development of joints with a global occurrence of 1 in approximately 3-5,000 live births (1) The diagnostic category of AMC describes over 400 heterogenous, congenital, and non-progressive conditions characterized by the rigid restriction or contracture of two or more joints (2) AMC can also impact other body structures and is thus categorized into three groups of traits according to the location or system of impact: 1) limb involvement only; 2) limb involvement and other systems (e.g., gastrointestinal tract); 3) neuromuscular involvement with central nervous system (CNS) involvement, or intellectual disability (2, 3). Although identification of contractures is commonly established by ultrasound or shortly after birth by a battery of clinical examinations (2), difficulties in establishing treatment strategies arise due to the rarity of AMC and heterogeneity of AMC characteristics (4).

To maximize treatment efficacy, early intervention using a multidisciplinary team approach involving physical therapy, surgical procedures, and assistive devices is required (5). Adults with AMC have been reported to undergo an average of 9 to 10 surgeries in their life time with half of surgeries occurring in childhood (6). Children with Amyoplasia, one of the most common groups of AMC, can expect to need therapy into their teen years (7). Obtaining access to treatment and services is expensive from both direct (e.g., monetary expenses) and indirect costs (e.g. loss of wages when attending appointments). The nature and extent of the services accessed by a child born with AMC can depend on the socioeconomic and sociodemographic profile of the family (8). Therefore, it is relevant to identify which if any of the specific profile variables influence outcomes in AMC.

The complex relationships between a child’s disability and the structure and profile of the family is shown by the biopsychosocial model from the World Health Organization's (WHO) International Classification of Functioning, Disability, and Health (ICF). This model provides a comprehensive framework classifying the relationships between body structures and functions, activities, and participation with complex non-medical and contextual factors of the individual and their environment. Non-medical factors are represented in the ICF by material and social factors and fall under the definition of social determinants of health (SDOH) (9). As the effects of SDOH can account for the quality of up to 55% of health outcomes in populations, they are pertinent to explore (10).Studies in chronic conditions have extended the ICF with the Wilson-Cleary model, augmenting the range of hypothesized relationships in the ICF with non-medical social factors and their contribution to levels of participation and quality of life, which are shown to be influenced by an individual's health perception (11). Previously explored via a scoping review (12), social deprivation is one important population-level construct used to capture SDOH using population-level census data. Social deprivation indices are constructed from multiple metrics collected as part of the census are used to identify regions or neighbourhoods that experience area-related diminished resources that can affect health. Methods used to contextualize social deprivation require national or regional census data, area-specific social deprivation indices, and/or area code. In this study, we used a collection of data points used in social deprivation indices to inform our analysis and create an index representing environmental support.

As AMC is a rare condition, site-specific investigations have limited value owing to small sample size. To address these challenges, a registry to collect clinical and patient-reported outcomes in children with AMC was implemented in 2019 across four Shriners hospitals for Children in Montreal, Canada; Philadelphia, Pennsylvania; Portland, Oregon, and Sacramento, California, USA (2, 13,) In 2021, four study sites were added in Chicago, Illinois; Honolulu, Hawaii, Greenville, South Carolina, and Shreveport, Louisiana, USA. Sites were selected as they had the largest patient pool of children with AMC across the Shriners network (13). The overall aim of the AMC registry was to facilitate research through providing harmonized data from many patients across multiple sites (13). Data was collected through questionnaire, medical chart review, and patient-reported outcome measures (13) The availability of a comprehensive set of data made the possibility of investigating the complex relationships between functional, environmental, and personal factors possible. Structural equation modelling (SEM) is well-suited for this type of multivariate analysis as SEM tests relationships between and among a set of variables that are theoretically linked. SEM uses a combination of correlation, factor analysis, path analysis, and regression with latent variables to test the extent to which the theoretical model underlying the choice of variables fits the data on hand. SEM provides estimates of the strengths of the paths (direct or indirect) between and among of a set of complex variables (14). Using a SEM analysis with the AMC registry data confers the advantage of modelling multiple associations between multiple outcomes.

Objectives

The objective of this analysis to was to identify the extent to which personal and environmental factors are associated with functional outcomes of symptoms/impairments, activity limitation, participation, and health perception among individuals 8-21 with AMC in Canada and the United States. The hypothesis underlying this statistical approach is that indicators from family-related characteristics used in the measurement of social determinants of health will produce a better model fit than personal factors.

Method and Procedures

A cross-sectional analysis was carried out using data from the Shriners AMC registry from January 2019 to December 2022 from the eight participating sites **(13).**

Ethics

Ethical approvals were obtained at both SHC-Canada (McGill University Faculty of Medicine Institutional Review Board A08-M30-19B) and all participating US sites through WCG (WIRB and Copernicus Group IRB #20191755). Administrative site approval was obtained for the registry from the Department of Medical Research at Shriners Hospitals for Children Sponsor (CAN1903). Ethical approval was sought prior to the commencement of data collection. As the collection of patient-reported outcomes was considered minimal risk, the local ethics boards granted a waiver of written consent. As such, only verbal consent was required prior to participation and was obtained by the clinical research coordinator (CRC) assigned to the study either face-to-face or remotely. All data was accessed through password protection and secured through encryption for authorized members of the research team.

Population

Entry into the registry study was open to pediatric patients and young adults 0-21 years of age with documented manifestation of multiple congenital contractures (i.e., two or more joints in different body parts with limitation of movement present at birth) identified as AMC (**15)**. Data were collected at eight Shriners locations (Montreal, QC; Chicago, IL; Greenville, NC; Honolulu, HI; Northern California, CA; Philadelphia, PA; Portland, OR; Shreveport, LA) in English, French, and Spanish to avoid recruitment bias. Primary caregivers of participants of all genders and ethnic backgrounds were invited to provide proxy reports, medical history, and demographic information. Recruitment was performed during hospital visits at the Shriners hospital locations by the clinical research coordinator at each site, or remotely by telephone or videoconferencing using a secure platform (MS Teams).

Participant Data

Inclusion and exclusion to the present study was based on a subset of data from participants between 8 to 21 years of age, as the measures used in this study were developed for children 8 years and over (*see Table 2*). Surgical procedures (braces, casting, assistive devices) are known to impact mobility and influence the scores on physical function in AMC (16). Studies show the average age of physical development on tasks such as ambulation, walking independence, and self-care in musculoskeletal conditions (e.g. Cerebral Palsy) are present on average by the age of 9 years (17). Therefore, the subset of data was chosen to represent the age category with the most complete data that was representative of the child's functioning. For this study, a subset of data from the AMC registry was extracted using the following criteria: the included data must represent 1) social determinants of health domains, 2) individual demographic and diagnostic characteristics of participants, and 3) measures of health outcomes describing level of impairment of body structures of functions, activity limitations, participation restrictions, and health perception.

Statistical Methods

SEM analysis provided greater advantages over multivariate regression by addressing the interrelationships between multiple variables and their simultaneous influence over multiple outcomes (18). It also facilitated the creation of variables to be represented by multiple indicators, thereby reducing random measurement error (19). Both latent (indirectly measured) and manifest (directly measured) variables were included to represent core components of personal and environmental variables, and all outcomes.

Model

As presented in previous studies in chronic conditions (11), we used the integrated ICF/W-C model as a basis by which to categorize all measures and hypothesize anticipate effects (see Appendix B and Figure 2). Figure 1 illustrates the integrated ICF/WC Framework used to situate data according to the individual's personal factors, environmental factors, health perception, and physical/social function.

*Figure 1: Combined ICF Framework with Wilson-Cleary Extension (**11-Mayo et al., 2020**)*

[insert Figure 1]

Figure 2 illustrates how the AMC registry subset of data measures were situated within the rubric of the ICF/WC model, and represents our included variables within the context of structural equation modeling (SEM).

Figure 2: Conceptual Model using the integrated rubric of the ICF/WC to categorizes measurements with identified SEM variables

Personal factors

Three independent variables were included to represent patient-related AMC characteristics: type of contractures, age, and biological sex at birth.

Environmental factors

Five independent categorical variables were used from the AMC registry as family-related factors used in SDOH research to identify the level of environmental support, called the Environmental Support Index. The data extracted from the registry used to represent environmental factors that have been identified in SDOH literature. These data were the categorical variables: level of education (20), employment type (21), housing/living arrangement (22), marital status (23), and parental stress (24). These data were extracted and coded into an index called The Environmental Support Index (ESI). The data comprising the index was interpreted to represent the level of environmental support from caregivers. Informed from literature review, the data were dichotomously coded according to practical or social resources where "1" (e.g. full-time employment; graduate degree) indicated a more advantageous level of support and "0" indicated less advantageous level of resources (e.g. volunteer; high school diploma). A cumulative score was derived ranging from 4 (most support) to 0 (least support). For a detailed description of the ESI, see **Appendix C.**

Functional outcomes

A total of six measures (five patient-reported outcome measures and one demographic questionnaire) were extracted from the AMC registry data. Data from the following measures for ages 8-21 were extracted from the AMC registry.

1. *EuroQOL-5D-Youth* (EQ-5D-Y) is a descriptive measure encompassing five dimensions including mobility, self-care, usual activities, pain/discomfort, and anxiety/depression (25). The EQ-5D-Y has been established for testing among pediatric and adolescent populations (26). Three response levels are included for each of the 5 domains to indicate frequency of functioning or interference: *a lot, somewhat, a little bit.* The 6th domain of health perception was included, where the child provides an overall score on the item "*How is your health today?"* 0-100 using a Visual Analog Scale (VAS) where highest score of 100 indicates best health.

2. *Patient-Reported Outcomes Measurement Information System – Short Form* (PROMIS-SF) subscales were collected for pain interference, mobility, upper extremity, and peer relationship short forms from Pediatric Bank v2.0 (27). This measure has been evaluated in a variety of diverse pediatric populations with chronic conditions as reliable, valid, and sensitive (27).

3. *Gillette Functional Walking Subscale* (FAQ) assesses the child's level of functional mobility by describing levels of mobility based on the environment, terrain, or obstacles, providing one score ranging from 1-10 on walking ability (Ammann-Reiffer et al., 2019). The FAQ was created for children with walking disabilities (28).

4. *The Functional Independence Measure for Children* (WeeFIM) was used to assess children 3-21 years of age by certified personnel. Self-care, mobility, cognition and total scores were computed. The WeeFIM has been widely used and validated among children with Cerebral Palsy (CP) (29).

5. *The Adolescent Pediatric Pain Tool (APPT)* was used to observe pain intensity by location, presenting an ordinal score on a visual analog scale (VAS) indicating 5 categorical intensities of pain from "no pain" to "worst possible pain." This tool is easy to administer to children, and has demonstrated sensitivity, reliability, and validity for acute pain characteristics among children ages 8-17 (30).

6. Child and caregiver data from the *Demographic Questionnaire* was extracted. Caregivers or individuals 18 or older completed the questionnaire, where the age, diagnostic AMC type, biological sex at birth, and living situation was extracted for individuals with AMC; the caregiver data extracted was age, education level, employment type, relatedness (e.g. mother, paternal grandparent, foster parent) marital status, and stress level.

Both PROMIS and EQ-5D-Y are measures designed to capture quality of life (QoL). In this context is defined as the subjective personal perception of every-day living quality observed and experienced by the child, or parent proxy report) (25, 27). However, the items included in the PROMIS and EQ-5D-Y were categorized according to item content rather than categorical labeling, so where items pertained to ICF domains and functions being evaluated, items were grouped and analyzed within that category. Four measures represented physical function. Within two measures, separate domains represented health perception, mood, and cognition. These measures importantly relate to the impact of AMC on mobility, independence, upper extremity use, pain experience, subjective health perception, mental health, and cognition. They are relevant to AMC based on the type of diagnosis, number, and location of contractures, and impact of other systems (e.g. CNS, gastrointestinal issues, oral health).

[insert Figure 2]

Statistical Analysis

The SEM analysis consisted of six steps: model specification, identification, data preparation, screening, parameter estimation, model evaluation, and modification (31, 32). A conceptual model was specified *a priori* by literature review and discussion with expert statisticians and is available in **Appendix C**. The subset of data from the AMC registry was prepared, and the model was programmed using the statistical software Mplus (33). Screening of variables was done by correlation matrix where exogenous variables (personal and environmental factors) were allowed to correlate with all endogenous variables, while endogenous variables were allowed to correlate with one another (e.g. cognition with peer relationships; pain with health perceptions).

Model identification and parameter estimation entailed testing the model if it is over-identified, just-identified, or under-identified. Model evaluation was performed in Mplus to assess the performance of the model, each path's significance, and goodness of fit. Finally, modifications to the conceptual model were made by removing variables if they met the following criteria: 1) their Spearman correlation coefficient with related measures was below

0.5, or 2) the variables did not demonstrate-significance in the path analysis, or 3) the variable reduced the fit of the model. After modifications, the revised model was evaluated once more. The Maximum Likelihood Estimation (MLE) measurement used was loglikelihood to identify the probability parameter of how well the model best fit our observed dataset. Statistical significance of all associations were identified by p-value <0.05. Goodness of fit was assessed using: 1) the Root Mean Square Error of Approximation (RMSEA) of a minimum value at or above 0.60; 2) the empirically validated Comparative Fit Index (CFI) at or above 0.90; and 3) Tucker-Lewis Index (TLI) of 0.60. Descriptive statistics and distributions were derived using SPSS version 29 (34).

Sample Size and Distribution

This project aimed to meet the requirements for 80% power, with a type I error rate of 5%, using a *p*-value of <0.05 to detect significance of association. There were 3 latent variables (Pain, Physical Function, and 10 manifest variables, the minimum sample size to detect an effect was 119 participants using a SEM sample size calculator. The data available for ages 8-21 was 189, therefore this sample size was sufficient for the planned analysis.

Due to the sample size and number of variables analyzed, this study maintained assumptions of normal distribution, linearity, homoscedasticity, and independence of observations. Due to the categorical nature of independent variables, and categorical and continuous nature of the dependent variables, multiple steps were be taken to ensure data is analyzed by a statistical procedure that will be suitable to answer the research question. A combination of histograms and Q-Q plot of predicted and fitted residuals were used to establish linearity of data. All variables exhibited distribution within the range of normality. A detailed description of data preparation, procedures, and analysis are available in **Appendix D.**

Missing Data

Missing data was anticipated to due to attrition as the length of data collection was undertaken at multiple locations over the years 2018-2022. Data were “missing completely at random (MCAR), as missing data occurred for reasons unrelated to the observed or unobserved data **(35)**. Missing data was handled by reporting only the number of participants who have complete data for that outcome. As this study is based on registry data and not evaluating a treatment, imputation of mean scores did not provide an advantage to the analysis.

To identify and perform the SEM analysis, a statistical methods expert (N.M.) was consulted to supervise and participate in the preparation and analysis of the data. Statistical software Mplus was used to create the correlation matrices which demonstrated the associations between each variable and test the significance of the model. SPSS was used to synthesize descriptive statistics, and test normality of distribution and linearity. Mean scores with standard deviation were be used for continuous scales, response frequency per category was used for ordinal scales (e.g. EQ-5D-Y domain scores from 1-3).

Results

Sample characteristics

The total sample was comprised of individuals with AMC (n=187) and their primary caregivers (n=185). The results are presented in Tables 1-3 and Figures 1-4. The mean age for individuals with AMC was 12.3, where 51.3% were male, and 78.6% had AMC impact to limbs only. Most children (70.5%) lived in the home of their two caregivers, where caregiver respondents were mothers (76%) who had a partner (60%). Occupations for caregivers were most often employed full-time (43%), or homemakers (19%). The majority of caregivers were educated after high school and had a college or associate degree (29%) or undergraduate degree (22%). The median level of caregiver stress was 2 (indicating "Sometimes stressed") on a scale from 0-4 (0=Always stressed; 4= no stress). Model fit was good (RMSEA: 0.06; SRMR: 0.67; with an acceptable CFI: 0.95).

Table 1*: Sample Characteristics of Children and Caregivers*

[Insert Table 1]

Summary of functional outcomes

Table 2: *Descriptive Statistics for All Outcomes*

[Insert Table 2]

Model Testing

Paths were allowed to all variables. The hypothesized theoretical model (see Figure 1) did not demonstrate goodness of fit. After removing the ESI from the SEM model, linear regression was used to identify if any of the caregiver variables of education, occupation, marital status, parental stress, and living situation were significantly associated with functional outcomes. Neither SEM nor linear regression of caregiver variables reached significance at the $p<0.05$ level. From the two stages of testing, we concluded the null hypothesis could not be rejected, and our first research question was answered: personal factors were more impactful than environmental variables in this sample.

Model Alterations

Variables that reduced the model fit and were either removed or revised by the consultation with a statistical expert. **Appendix E** details the variables and justification of these revisions. Of note, as the analysis situating parental stress as an environmental exposure failed to reach goodness of fit, this variable was reevaluated by our statistical expert (N.M.) as an outcome, as parental stress could also be an indication of the level of stress a caregiver experiences in response to their child's health and the caregiver's subjective circumstances. It was therefore input in the revised model as a dependent variable. The final conceptual model is described in **Appendix F**.

Final Model

Figure 3 displays the final model with all paths labeled by standardized beta coefficients. Standardized beta coefficients allow for comparisons between measures on different scales. They indicate that for a change (increase or decrease) of 1 standard deviation in the independent variable is associated with a standardized beta coefficient of change in the dependent variable. After revisions, the final demonstrated acceptable goodness of fit using RMSEA, CFI, and TLI, which are described in **Appendix G.**

***Figure 3**: Final Model with all clinically and statistically significant paths and beta coefficients*

[insert Figure 3]

Table 3 presents the SEM model paths by ranked first by standardized beta coefficients with clinically and statistically significant paths. The largest effect sizes identified were the moderate effect of pain (β=0.348) and small effect of mood (β=0.238) on the level of child health perception. The third strongest effect was found where the child's level of physical functioning had a small effect (β= -0.238) on parental stress frequency.

***Table 3:** Model paths of all statistically or clinically significant direct effects by Variable*

[Insert Table 3]

Discussion

The final ICF/W-C model tested by SEM demonstrated good fit with personal factors**.** For children 8-21 with AMC, the strongest effects were observed for variables of pain and mood, where they had a moderate effect on the individual's health perception, and physical function had a small effect on caregiver stress. Cognition and AMC type had a small effect on peer relationships, pain had a small effect on health perception, and pain differed by biological sex at birth.

Overall, participants exhibited highest mean scores and lowest variance on health perception, peer relationships, and cognition. The upper extremity function/self-care items had the lowest scores, accompanied by the highest proportion of variance, followed by lower-extremity/mobility. Small effect sizes were found where the type of AMC (limb only) was associated with better peer relationships than the other AMC diagnostic categories (limb and other systems, limb and CNS). Female participants reported less frequency of pain than male participants. Level of involvement representing greater impact on body structures and functions reported lower quality of peer relationships. AMC type 2 categories were not associated with higher frequency of parental stress. The finding that personal factors were more impactful to functional outcomes could be due to the following: 1) the nature and physical impacts of AMC fundamentally alter body structures and functions to the effect that the opportunities brought about by caregiver resources can help only to a degree; 2) surgery and subsequent physical rehabilitation are the gold-standard, and based upon the individual impact of AMC on the child; 3) the type of data included from the AMC registry was not collected to represent SDOH and therefore may not have sufficiently reflected the construct; and 4) small sample size could have contributed the reduced model fit of the initial SDOH model, making it harder to detect an effect.

Consistency with literature

The majority of primary caregivers (47.8%) reported feeling stressed ‘sometimes’ followed by ‘rarely’(23.2%) between taking care of their family and work responsibilities and taking care of their child’s needs. Psychosocial stress among caregivers of children with musculoskeletal disabilities such as cerebral palsy (CP) have demonstrated similarly higher-than average levels of stress **(36)**, however this is the first study to identify one of the impacts of caregiver stress in the context of AMC. Caregiver stress contributed importantly to this study, in that it was tested as both an exposure and an outcome, and only exhibited statistical significance as an outcome. Although psychosocial stress can in some contexts act as both, literature mor often identifies this variable as an outcome among caregivers of children with disabilities, regardless of the nature of the disability category (37). Furthermore, the association found between lower physical function and higher parental stress is corroborated in recent literature, where a 2021 study identified that caregivers exhibited increased stress specifically in the context of obtaining physical therapy for their child (38). Because psychosocial stress is a subjective experience and can vary by individual, these findings contribute to a more fundamental understanding of specific factors that impact caregiver wellbeing. This study also recommended the development of a parental stress questionnaire tailored to the context of physical therapy components leading to increased stress, to improve efficacy of clinical inquiry and subsequent intervention (38).

The impact of cognition in AMC diagnosis, although rare, is well established in AMC literature where the presence of CNS involvement has often been associated with intellectual disability and cognitive difficulties (39). It is therefore not surprising that the model paths from the AMC types representative of increased system impact (e.g. gastrointestinal, CNS involvement along with joint involvement) and cognition both contributed significantly to the level of participation in peer relationships. Extended beyond physical limitations into social participation restriction, or social isolation, have been reported, where having a rare physical condition not understood by peers contributes to increased frequency of social distress and reduced social engagement (40).

Clinical Implications

Participants who reported greater frequency of pain and lower quality mood also reported lower quality of health perception. This finding is useful in clinical settings, where clinicians can identify rehabilitation strategies that focus on reducing pain frequency while providing psychosocial supports to improve health perception. Where caregivers are concerned, higher frequency of physical challenges (e.g. lower scores on self-care, mobility, and daily activity) can be used by clinicians as a prompt not only to address the child's physical needs, but also to to ask caregivers about concerns related to physical therapy strategies, requirements, and every-day challenges to advance family-centered care..To that end, psychosocial stress has been found in recent literature to be improved by access to emotional support in clinic setting where caregivers can raise current concerns, and social settings such as caregiver support groups (41). Finally, as parental stress did not demonstrate goodness of fit in the first hypothesized model as influencing outcomes, this contributes to the interpretation that 1) the frequency of caregiver stress does not directly impact physical function for their child, and 2) caregivers are indirectly impacted by AMC challenges that can be improved in settings like caregiver support groups. Clinicians can apply these findings to proactively identify and reduce barriers in the process of physical rehabilitation.

Limitations

Limitations in this study included availability of data on SDOH, sample restriction to Shriners locations, sample size, type of data collected, the ESI metrics, and the interpretations of parental stress. As the AMC registry project did not have the goal of collecting data on SDOH or social deprivation, the included response categories, and interpretations of the importance of these factors were limited. Postal code was not available for data extraction, and area-level factors impeded our aim of calculating social deprivation in this sample. As the registry data was collected only from Shriners locations, it is possible there was reduced diversity of included participants on characteristics such as employment type, education, education, as distance from services may have limited participation for families in more rural areas. Additionally, the measurement type used to comprise the ESI may have influenced the reliability of reflecting the construct. Caregiver item responses were categorical, which introduced ambiguity to the statistical representativeness of each domain. For example, the variable 'employment' provided a general domain of employment type and source, but may not completely represent the material environmental in the majority of partnered participants as the second partner's socioeconomic status was not collected. An additional limitation of the testing of SDOH using the ESI was that the statistics for validity or reliability were not measured. Although model fit represents the fit of the sample to the hypothesized relationships, future studies would benefit from including these metrics. Due to attrition in the participants in the data registry, some missing data reduced the sample size. This limitation was addressed by modification of the model to remain consistent with the minimum sample size to detect the effects between all included variables (n=119).

The variables of mood, pain, health perception, and parental stress were limited to one response item representing the entire construct at the time of measurement. Although the model fit was moderate and paths significant, the generalization of the model is limited as single items are susceptible to response bias, reduced reliability, and limited validity of capturing the intended multifaceted construct. Future studies are recommended to investigate psychosocial outcomes using the parental stress index (PSI), or exploring the use of SDOH questionnaires to augment the suitability of treatment for the caregiver and child (42). As an example, a mixed-methods study could administer the PSI to caregivers, and functional outcomes using PROMIS or WeeFIM for AMC child participants, coupled with semi-structured qualitative interviews of both to observe the lived experience of stress in the context of AMC, and associated direction and level of impact between SDOH, and every-day physical and psychosocial functioning. Although one recent study has comprehensively investigated predictors of mental health outcomes anxiety and depression among adults with AMC (43), parental stress was not measured due to the population age range. This provides the opportunity to explore the impact of stress in AMC to facilitate optimized support. Finally, social deprivation was unable to be calculated because area code was not an included data point in the protocol. To that end, eight sites included in the registry would require state-based census data in addition to a sufficiently comparable social deprivation index between Canada and the United States, which at this time are not developed.

Importance of caregivers & future directions

Our findings indicate that caregivers should be considered in the rehabilitation strategies of children with AMC. It is well established that caregivers of children with disabilities play multiple roles in providing support and assistance in daily activities like walking, ambulation, eating, clothing, and physical rehabilitation exercises, in addition to emotional, educational, and social support, leading to increased stress (44**).** As caregiver are the primary point of contact with clinicians and practitioners, recent research advocates appointments as an opportunity to practice family-centered care by including caregivers in rehabilitation strategies, which can facilitate improvements in the needs of the child and their caregiver (45). Although this study did not begin with the objective of highlighting caregiver contribution and indirect impact of level of physical function on stress, this finding emerged and is importantly relevant to practitioners when investigating interventions that include the parent and the child. There are noted limitations to the parental stress measurement as it was only 1 item, however our findings provide the first empirical answer to questions raised in the AMC community such as "is my level of stress making my child's condition worse?" The two stages of the SEM analysis indicated that parental stress was not impactful on any of the child's outcomes; rather, caregivers demonstrate the need for psychosocial support based on greater levels of physical limitation.

Conclusions

These findings contribute to an important gap in AMC research which identifies the extent to which personal factors impact level of functioning, and how level of functioning impacts the quality of the child's health perception and peer relationships, and the frequency of the caregiver's psychosocial stress. Future studies can be informed about which domains to investigate from our final model presenting the strength and direction of association between contextual factors to advance approaches to SDOH in clinical settings. Where improvement of the above domains are targeted, clinicians can use these findings to identify which factors (e.g. type of AMC, biological sex at birth, physical function, cognition, mood, pain) to examine which could be contributing to diminished health status and psychosocial wellbeing. To that end, researchers aiming to investigate SDOH in the context of musculoskeletal disorders will have preliminary findings relevant to environmental factors used in the social deprivation construct, in addition to a clear operationalization, which has been lacking in previous studies. Finally, this research contributes to raising awareness among families of and children with AMC, by providing a statistical model that describes the direction, strength of impact, and factors that contribute to the every-day experiences of pain frequency, peer relationships, health perception, and caregiver stress.

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Conflicts of Interest

The authors have no conflicts of interest to report.

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Table 1: Sample Characteristics of Children & Caregivers

Child Characteristics	**N**	**Mean (SD) or Frequency (%)**
Child Age	187	12.3 (3.4), 8-21.9
Child Biological Sex	187	
Male		96.0 (51.3%)
Female		91.0 (48.7%)
AMC Diagnosis Type	179	
Limb Only		147.0 (78.6%)
Limb+Other systems		17.0 (9.1%)
CNS involvement		15.0 (8%)
Missing		8.0 (4.3%)
Home Residence of Child	185	
1 Parental home, 2 CG		141.0 (70.5%)
1 Parental home, 1 CG		38.0 (19.0 %)
Other		6.0 (3.0 %)
Missing		15.0 (7.5%)

Caregiver Characteristics	**N**	**Mean (SD) or Frequency (%)**
Age at birth of child (years)	185	28.4 (6.1); 16-42
Occupation	137	
Full Time		86.0 (43.0%)
Homemaker		38.0 (19.0%)
Part-Time		25.0 (12.5%)
Self-Employed		20.0 (10.0%)
Disability Benefits		6.0 (3.0%)
Other		8.0(4.0%)
Missing		15.0 (8.0%)
Marital Status	183	
Single		31.0 (15.5%)
Divorced or Separated		22.0 (11.0%)
Partnered		120.0 (60.0%)
Not reported		10.0 (5.0%)
Missing		17.0 (8.5%)
Education	185	
Less than high school		7.0 (3.5%)
High school		41.0 (20.5%)
College or Associates		58.0 (29.0%)
Undergraduate		44.0 (22.0%)
Masters		23.0 (11.5%)
Doctorate		7.0 (3.5%)

Missing		15.0 (7.5%)
Relationship to Child	185	
Mother (birth or adoptive)		158.0 (80.0%)
Father		23.0 (11.5%)
Other		8.0 (4.0%)
Missing		15.0 (7.5%)

Table 2: Descriptive Statistics for All Outcomes

ICF/Wilson-Cleary Domain	Measure	N	Scoring	Mean (SD) or Frequency (%)
Impairments				
Pain	APPT*	158	0-10	8.04 (2.5)
Pain or discomfort	EQ-5D-Y*	156	A lot	5 (2.55)
	EQ-5D-Y*		Some	67 (33.5%)
	EQ-5D-Y*		None	84 (42%)
			Missing	31 (15%)
Pain Interference	PROMIS-SF**		6.25-100	75.59 (24.0)
Worry/Sad/Unhappy	EQ-5D-Y*	156	Very	4 (2%)
	EQ-5D-Y*		A bit	56 (28%)
	EQ-5D-Y*		Not	96 (48%)
	EQ-5D-Y*		Missing	31 (15%)
Activity Limitation				
Cognition	WeeFIM**	163	3.3-100	94.01 (17.4)
Lower Extremity Function	PROMIS-SF**	141	6.3-100	63.50 (25.9)
Problems walking about	EQ-5D-Y*	156	A lot	27 (13.5%)
	EQ-5D-Y*		Some	59 (29.5%)
	EQ-5D-Y*		None	70 (35.%)
			Missing	31 (15%)
Mobility	WeeFIM**	163	0-100	71.9 (30.9)
Self-Care	WeeFIM**	163	0-100	72.6 (29.4)
Problems washing or dressing	EQ-5D-Y*	156	A lot	28 (14%)
	EQ-5D-Y*		Some	59 (29%)
	EQ-5D-Y*		None	69 (34.5%)
	EQ-5D-Y*		Missing	31 (15%)
Overall Independence	WeeFIM**	163	0.93-100	78.3 (22.9)
Upper Extremity Function	PROMIS-SF**	142	0-100	58.6 (29.3)
Walking Ability	FAQ Walking **	170	0-100	62.0 (34.3)
Participation				
Problems with usual activities	EQ-5D-Y*	156	A lot	6 (3%)
			Some	68 (34%)
			None	82 (41%)
			Missing	31 (15%)
Peer Relationships	PROMIS-SF**	142	0-100	80.8 (19.0)

Parental Stress * ***	Demographic Questionnaire	138	2.2 (1.1)	
			Always	0
			Frequently	20 (14%)
			Sometimes	66 (47.8%)
			Rarely	32 (23.2%)
			Never	20 (14%)
Health Perception				
Health Today VAS	EQ-5D-Y*	158	3-100	83.9 (18.4)

Measurement scales indicate high scores = better health for all measures
* indicates a measure that wase reverse-scored
** indicates a measure where raw scores were converted to a score out of 100
EQ-5D-Y scores represent 1=worst health, 3= best health
*** Parental stress item: *Do you feel stressed between caring for your child and trying to meet other responsibilities for your family/work?*
Item responses: *0=Always, 1=Frequently, 2=Sometimes, 3=Rarely, 4=Never*

Table 3: Model paths of all statistically or clinically significant direct effects by variable

Variable Type	Personal Factors	Impairment	Activity Limitation	Participation	Health Perception	Standardized (STDYX) β	S.E.	Est./S.E.	*p*-value
	Biological Sex	Pain				-0.167	0.078	-2.134	0.033*
	Biological Sex			Peer Relationships		0.070	0.084	1.812	0.070
	AMC Diagnosis			Peer Relationships		0.174	0.082	2.121	0.034*
Observed		Pain			Health Perception	0.348	0.074	4.707	0.000*
Observed		Mood			Health Perception	0.238	0.079	3.018	0.003*
Observed		Mood		Peer Relationships		0.163	0.087	1.866	0.062
Observed			Cognition	Peer Relationships		0.188	0.080	2.351	0.019*
Latent			Physical Function	Parental Stress		-0.244	0.082	-2.982	0.003*
Latent			Physical Function		Health Perception	-0.160	0.076	-2.102	0.036*

*indicates significance at the <0.05 level

"Biological sex" was coded 0=Males, 1=Females

AMC Diagnosis was coded 0=Limb involvement only, 1=Limb+Other Systems, Limb+CNS

Figure 1: Combined ICF Framework with Wilson-Cleary Extension (Mayo et al., 2020)

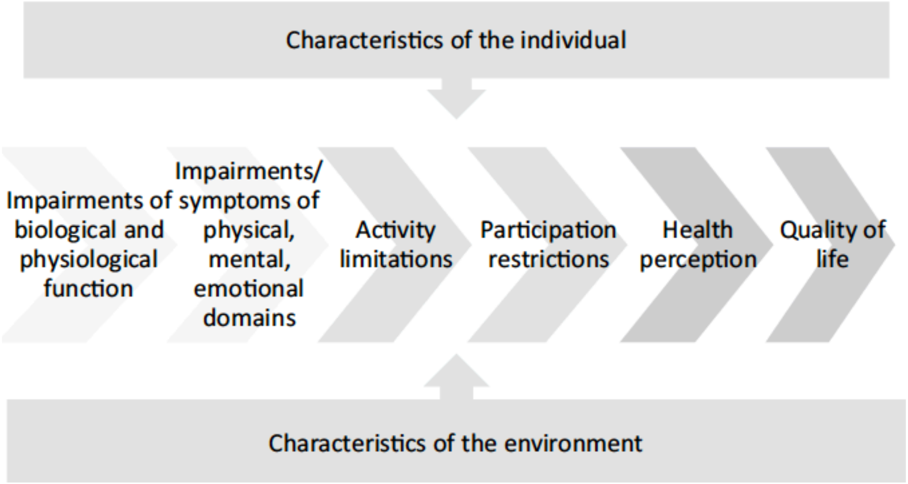


Figure 2: Conceptual Model using the integrated rubric of the ICF/WC to categorizes measures, with identified SEM variables

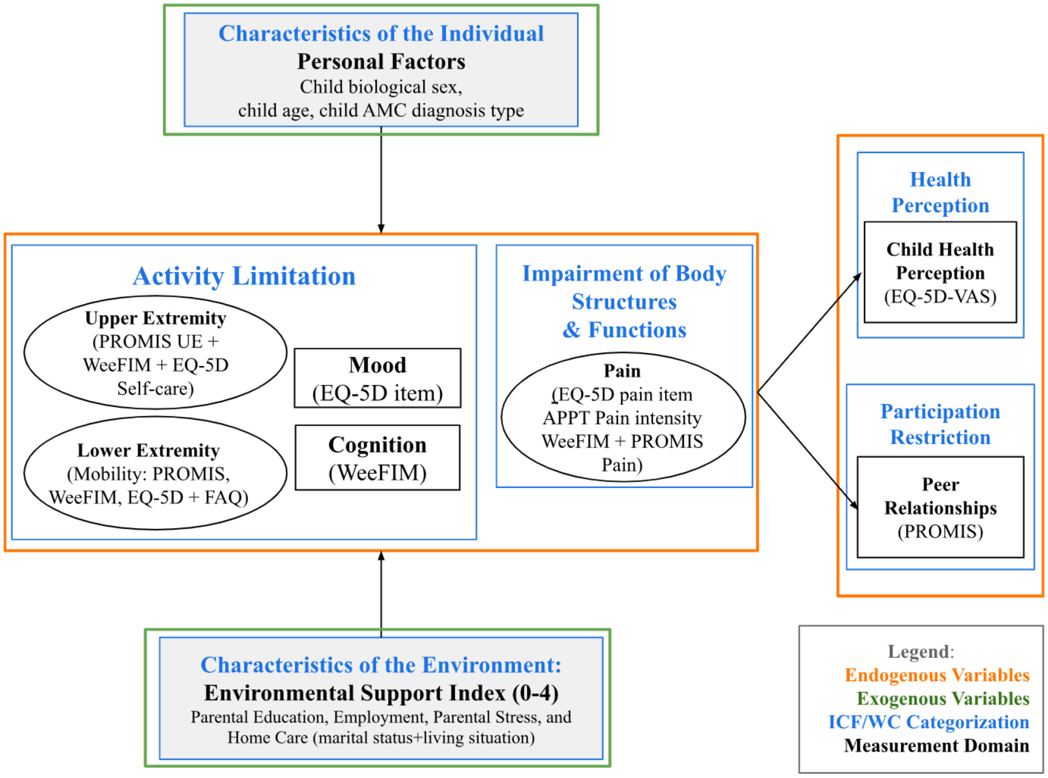
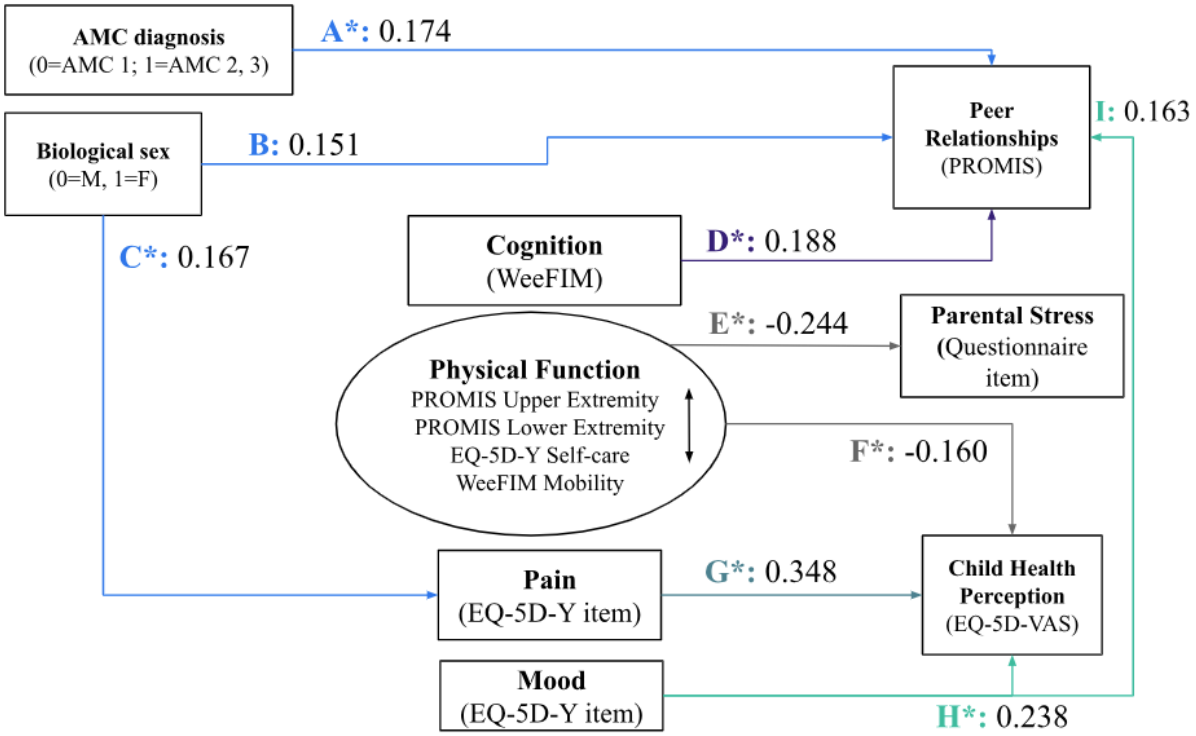


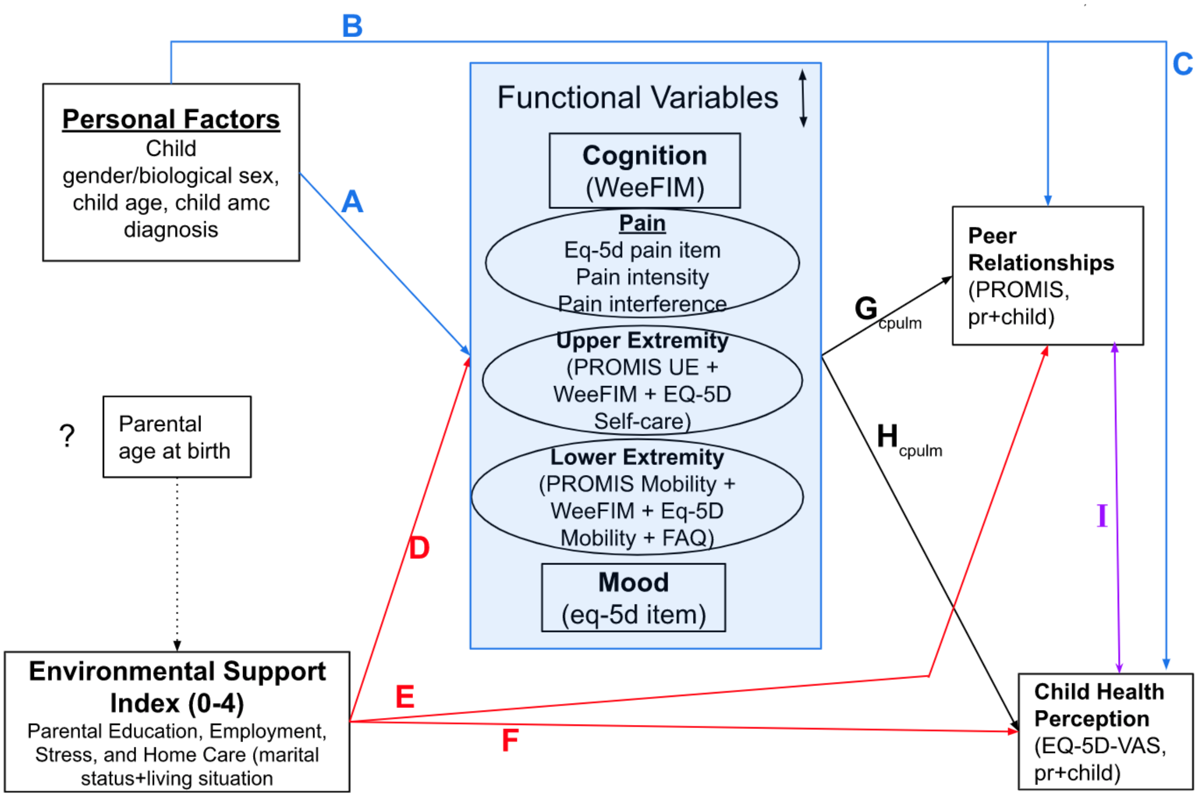
Figure 3: *Final Model with all clinically and statistically significant paths and beta coefficients*



* Indicates statistical significance at *alpha <0.05*
Bidirectional Arrow indicates a Pearson Correlation of 0.5 or higher
Personal Factors= AMC Diagnosis; Biological Sex at birth

APPENDIX A: Hypothesized Conceptual Model & Hypothesis Table

Table A: Initial Conceptual Model



APPENDIX B: First Conceptual Framework

ICF Classification	Domain	Measure	Variable Type
Impairment	Pain	1. EQ-5D-Y: Pain item 2. APPT VAS: Pain Intensity 3. PROMIS: Pain Interference	1 Latent
Activity Limitation	Upper Extremity	- PROMIS: Upper Extremity - PROMIS: Lower Extremity - EQ-5D-Y: Self Care	1 Latent
	Lower Extremity	1. PROMIS: Mobility 2. WeeFIM: Mobility 3. Gillette Functional Walking Status Subscale (FAQ) 4. EQ-5D: Mobility 5. EQ-5D: Daily Activities	1 Latent
	Cognition	1. WeeFIM: Cognition	
Participation Restriction	Peer Relationships	PROMIS: Peer Relationships	1 Manifest
		EQ-5D-Y: Mood	1 Manifest
Health Perception (Wilson-Cleary extension)	Child Health Perception	EQ-5D-Y VAS: How is your health today?	1 Manifest
Environmental Factors	Environmental Support	Demographic questionnaire items: 1. Parental Education 2. Parental Occupation 3. Marital Status 4. Living Situation 5. Parental Stress	4 Manifest
Personal Factors + Health Conditions	Personal factors	Demographic questionnaire items: 1. Age 2. Biological Sex (M=0, F=1) 3. AMC Diagnosis (1=Limb Only; 2: Limb and Other Systems; 3: CNS involvement =0)	3 Manifest

APPENDIX C: Coding and interpretation of Environmental Support Index

Factor	Variables	Response Options	Coding Interpretation
Personal Factors	Child age	8-21	Child's age is interpreted according to physical abilities and social norms and roles in the periods of childhood, adolescence, and early adulthood.
	Child biological sex	Male or Female	In social contexts, social roles of gender perception will be used. • Kedar, 2019 In physical contexts, such as fine motor vs larger lifting, biological sex will be used as physical strength and some motor functions are seen to be different among girls (increased motor function) and boys (increased physical capacity) • Kedar 2019 Biological sex was coded as M=0, F=1.
	Type of AMC diagnosis	1. Limb involvement (limb has a contracture) 2. Limb & other system involvement (e.g. limb contracture + cardiac or gastrointestinal impact) 3. Neuromuscular involvement with central nervous system dysfunction or intellectual disability	The contractures inherent in AMC diagnoses have been split into 3 categories which describe the level of impact. The interpretation of categories 2 and 3 are that they represent an increased impact on more body structures and functions than category 1. • Hall, 2004 Diagnosis was coded as 0= AMC diagnoses 2+3, 1=AMC Diagnosis 1
Environmental Support Index	Parental Employment	1. Full time 2. Part time 3. Self-employed 4. Retired 5. Homemaking	Employment is interpreted as it relates to the frequency of employment opportunities or production of monetary income.

	6. Student 7. Volunteer 8. Social assistance 9. Workers compensation 10. Disability benefit 11. Accident insurance 12. Temporary sick leave	1=Employment full time (1) Self-employed for money, (3) 0=part-time for money (2) not employed for money (4-12)
Parental Education	1. Primary or elementary 2. Middle school 3. Highschool 4. College/associates degree 5. Undergraduate 6. Masters 7. Doctorate	Education is coded as it relates to credentials required for levels of employment: 1= Undergraduate and above (5-7) 0=College degree and below (1-4)
Parental Stress	How stressed do you feel between caring for your family and child?	Parental stress is interpreted as the perception of the parent in meeting both social and material needs of the family and child. The interpretation of this response intensity is that it will represent the caregiver's experience of stress, which has been used as a proxy indicator for material and social support or strain. 1= Never, Rarely, 0= Sometimes, Frequently, Always
Living Environment	<u>Marital Status:</u> 1. Single 2. married/common law 3. divorced /separated 4. Widowed <u>Living Situation:</u> 1. Parental home, 2+ caregivers	Marital status and Living environment were combined to increase sample size. This comprises a composite observed variable which more completely represents the components of a living environment, which include a caregiver (whether single or partnered) and the home in which the child lives. 1= Partnered, parental home 2+ caregivers 0=Single, Parental home single caregiver + Foster home

2. Parental home,
   single caregiver
3. Group Home
4. Foster Home

APPENDIX D: Process of Data Analysis, Interpretation of Variables, Revisions

Phase 1: Preparation of Data: Independent Categorical Regrouping

The first phase of analysis was to identify the distribution of responses for all levels of independent variables. Where response levels are not populated (<2), they were be removed. Where multiple highly related categories are populated, categories were be collapsed in order to present distinct response categories exhibiting normal distribution (e.g. 'education' had 14 response levels, but 3 were sufficient to describe distinct categories. After obtaining the distributions from collapsed categories, all variables representing personal or environmental support factors were recoded into binary variables "0" and "1." To create the environmental support index, variables were recoded with 1 representing the category associated with highest support. A total score of 4 would indicating highest support. Personal variables were binary coded by diagnosis and biological sex. Child age at interview and Parental age at birth were also included as continuous variables to test for significance within the SEM model. Variables and their interpretations are listed below.

Phase 2: Treatment of Dependent Variables

Dependent variables were completed by self- or parent proxy-report for children between the ages of 8-21. All measures were rescaled and/or reversed where applicable to represent a 0-100 score, with 100 indicating best health. The EQ-5D-3L domains were kept as categorical scales with 3 levels and reverse scored, where 1 indicates worse health and 3 represents best health. The EQ-5D VAS was continuous with a scale from 0-100. A total of 14 variables were analyzed from measurement instruments. **Appendix A** summarizes these variables, their measures, and position as manifest or latent in the model.

Phase 3: Correlation Matrix

A Spearman correlation matrix was used to observe the level of association all variables measurements. Spearman correlation was chosen as it best represents ordinal variables. The functional measurements (e.g. measures of upper extremity, lower extremity, pain) that showed a moderate to high correlation were kept for the SEM path analysis, to be tested for their representativeness of construct and goodness of fit as manifest or latent variable.

Phase 4: SEM Path Analysis

The model paths were assessed for significance using the beta coefficient and p-value. Standardized Beta coefficients indicate the level of change expected by standard deviation for every unit of increase in the independent variable. These values ranging from -1 to 1. Statistical significance of the beta coefficient was determined by $p\text{-value} < 0.05$. Goodness of fit was examined for the conceptual model and all non-significant paths were removed or adjusted.

APPENDIX E: Model Alterations

The ESI did not demonstrate statistical significance or importance to any outcomes as hypothesized and was removed from the model. Parent proxy-report was highly correlated with self-reported outcomes and was therefore removed. Parental age at childbirth and child's age at the time of interview were removed as they did not add predictive value to the model. The correlations for the latent variable "Pain" did not meet the correlation cut-off (minimum Spearman correlation of 0.5), likely due to the item differences in description of pain (e.g., pain intensity, pain frequency, pain interference). Only the EQ-5D-Y pain item was kept as it best represented the construct of interest (frequency of pain related to impairment) and did not diminish the model fit. APPT-VAS and PROMIS Pain interference were removed. The variables of Upper Extremity and Lower Extremity were regrouped as Physical Function, using the most highly correlated measures. EQ-5D-Y VAS remained an important path to the model representative of child health perception, along with Peer Relationships.

The model's endogenous variables included two groups: the first group represented singular domains of function: physical function, cognition, mood, and pain. The second group of endogenous variables pertained to complex domains requiring multiple functions: parental stress, child health perception, and peer relationships.

APPENDIX F: Final Conceptual Model

ICF Classification	Domain	Measure	Variable Type
Impairment	Pain Intensity	EQ-5D-Y: Pain item	Manifest
Activity Limitation	Physical Function	1. PROMIS: Upper Extremity 2. PROMIS: Lower Extremity 3. WeeFIM: Mobility 4. EQ-5D-Y: Self Care	1 Latent
Participation Restriction	Peer Relationships	PROMIS: Peer Relationships	1 Manifest
	Cognition	WeeFIM: Cognition	1 Manifest
	Mood	EQ-5D-Y: Anxiety/Depression	1 Manifest
Health Perception (Wilson-Cleary extension)	Child Health Perception	EQ-5D-Y VAS: How is your health today?	1 Manifest
Personal Factors + Health Conditions	1. Biological Sex (M=0, F=1) 2. AMC Diagnosis (1=Limb Only; 2: Limb and Other Systems; 3: CNS involvement =0)	Demographic questionnaire	2 Manifest

APPENDIX G: Fit indices of the final model

Model Fit Parameter	**Value**	**Fit indices cut-off criterion**
Chi-Square Test of Model Fit	52.49	
Degrees of Freedom	34	
P-value	0.022	Non-significant chi-square p= greater than 0.01
RMSEA Estimate	0.06	0.06-0.08=marginally acceptable fit 0.01-0.05 = excellent
90% CI	0.023-0.090	Should not include zero
Comparative Fit Index (CFI)	0.95	0.90-0.95= marginally acceptable 0.96-0.99= excellent
Tucker-Lewis Index (TLI)	0.90	0.90-0.95= marginally acceptable 0.96-0.99= excellent
SRMR (Standardized Root mean square residual)	0.067	0.06-0.08 = marginally acceptable

CHAPTER 6: DISCUSSION

6. Did our findings support our hypotheses?

Our initial hypothesis that SDOH are important and impactful to the health and wellbeing of childhood disability populations was partially supported, in that social deprivation was impactful to ASD cohorts reported in the scoping review as Manuscript 1 but was not statistically significantly associated to outcomes in children with AMC in Manuscript 2. The findings of these two manuscripts support the hypothesis that the contextual factors (environmental and personal factors) are differentially impactful in these two groups of early-onset conditions. In the findings of both manuscripts, environmental factors (attributes of caregivers of children with disabilities) did not clearly demonstrate the expected socioeconomic gradient in health outcomes that have been reported in the SDOH literature, where advantageous material and social advantage is associated with better health (Bonaccio et al., 2020). The disparity between findings in the AMC registry study could be due to methodological limitations from the type of data collected or the categorical nature of SDOH variables in the AMC registry study.

6.1. Manuscript 1

To answer the research question of Manuscript 1, the SDOH risk factor known as social deprivation demonstrated an impact to families of and children with ASD in a six studies (Powell et al., 2000; McConachie at el., 2008; Griffith et al., 2012; Warnell et al., 2015; Schroder et al., 2015; McConkey et al., 2020); and outcomes (e.g. incidence, socioeconomic representativeness of respondents to two ASD databases, distribution of ABA therapy, testing a proactive intervention, and prevalence). The levels of deprivation in the caregivers' area of living identified by the social deprivation indices showed an impact on prevalence (McConkey et al., 2020) and accessibility to therapy among children with ASD (Griffith et al., 2012), indicating that SDOH are impactful in ASD. Children on the autism spectrum are known to have increased vulnerability to physical and social environments, as elements of the diagnosis involve high sensitivity and reactivity to external stimuli (Lai et al., 2014). When considering the sensory issues, limitations in social communication, and rigid behavioural tendencies, the environment acts as a mediator in participation in home, school, and community activities, accounting for up to 65% of variation in health outcomes (Anaby et al., 2014).

6.2. Manuscript 2

In answer to the second manuscript's research question, environmental factors, whether in SEM or regression analysis, did not reach significance with any outcomes when set in the model as an exposure. Personal factors were more important and exhibited best fit within the model, where children with more systems impacted by AMC (i.e., limb+other systems, limb+CNS involvement) reported lower satisfaction with peer relationships, and females reported having less frequent pain in daily activities than males. Regarding the factors that were most highly associated with the outcomes of health perception, and parental stress, effect sizes ranged from moderate to small, where small to moderate effect sizes (ranging from 0-1, small effects=0.1-0.2; moderate effects = 0.3-0.4) (Nieminen, 2022) were observed between: pain on health perception (beta coefficient =0.384), physical function on parental stress (beta coefficient=0.244), and mood on health perception (beta coefficient =0.238). Children who reported having less frequent pain perceived their health more positively than those reporting more frequent pain. Although less pain and better health perception is expected, biological sex at birth had an indirect effect on health perception, as males reported higher frequency pain than females. This finding is contrary to literature among adults with chronic musculoskeletal conditions, where women report higher pain frequency than men (Stubbs et al., 2010). Studies on pain in pediatric disability populations involve a number of qualifiers or contexts such as pain intensity, severity, frequency, quality, and location (Sions et al., 2022).

6.3. Integration of findings from Manuscripts 1 and 2

Two clear findings emerged from both manuscripts: 1) the importance of considering both categories of contextual factors in childhood disabilities, recognizing that the degree of influence by personal and environmental factors can vary according to the characteristic impacts of the childhood condition(s) (e.g. number and severity of systems impacted; visible or invisible disability), and 2) caregivers sit in a unique position both providing every-day resources and support to their children, while simultaneously being impacted and indicating the need for social supports by health care professionals.

6.4. Caregivers

An unexpected finding in both manuscripts was the highlighting of caregivers and the need for psychosocial support for caregivers of children with ASD and AMC. During the stakeholder consultation in the scoping review on ASD (Manuscript 1), a care coordinator for children with disabilities noted the absence of social support in clinical settings, in addition to the lack of representation of both child and caregiver psychosocial health outcomes in the included articles on social deprivation. In the AMC context, the psychosocial health of the child represented by child-reported mood and caregiver stress were among the variables with the strongest effect sizes, with child mood on health perception, and child's physical function on parental stress.

6.5. Novel findings from Manuscripts 1&2

This thesis presented novel findings that contributed to important gaps in childhood disability research. The first was in the scoping review on social deprivation in ASD that included clarifications important to future research. The review was consistent with a previous scoping review on social deprivation explored in musculoskeletal trials (Smith et al., 2020), finding that social deprivation research is limited in systematic and global comparison due to the differences in data indicators used to calculate deprivation across regional indices. To that end, the scoping review in Manuscript 1 also reported inconsistency in terminology, that research efforts can also be limited by lack of clarity or operationalization of social deprivation, as its meaning varied according to its use in autism literature vs. public health use.

Numerous definitions unrelated to population health emerged and were distinguished by this review. Three main categories of social deprivation were observed: i) reduced health or access to therapy based on living area and social opportunity; ii) the neurobiological effects of early-childhood deprivation of social stimuli, which were associated with quasi-ASD symptoms among orphaned or institutionally-reared children (e.g. social experiential deprivation (Kaku et al., 2017), psychosocial deprivation (Merz et al., 2010; Kumsta et al., 2016), social-emotional deprivation (Julian et al., 2016)); and iii) autism-specific social deprivation hypothesized to develop because of ASD, where inattention to social stimuli in early development results in social deprivation experiences in later developmental years (Schroder et al., 2015).

The second novel finding in Manuscript 2, was the use of multivariate structural equation modeling illustrating the direction of multiple relationships not previously established. This study had the opportunity to provide a basis for further studies looking to investigate the intricacies of contextual factors in AMC care and produce qualitative studies that include caregivers and children to advance the applicability of treatment according to the patient's specific context.

The third novel finding was to identify the direction of influence between parental stress and the child's functioning, namely: that the frequency of psychosocial stress of caregivers was strongly associated with the severity of limitations on their child's physical independence (self-care and mobility). Despite the limitations to generalizability of the stress item used to represent this finding, AMC literature has not previously established the direction of impact between caregiver stress and their child's functional outcome. This is the first study to suggest the direction of association, which can be investigated by a mixed-methods study using a complete and validated measure of stress that better represents the entire construct of psychosocial stress, such as the use of the PSI (Berry & Jones, 1995), in conjunction with caregiver and child qualitative reports.

The advantage of the SEM methodology was that it provided a clear answer to this question by determining if parental stress fit as an exposure in environmental factors or as an outcome of the child's health functioning. As parental stress was not significantly associated as an exposure influencing child health, we can conclude with some caution that, based on our analysis, caregivers need not worry about their stress negatively impacting their children's health. Instead, by observing the extent to which parental stress was impacted and by which variable, we can now recommend that health care providers include inquiries about social support for caregivers in cases where AMC physical impacts are most severe.

6.6. How to support caregivers and children

To address the needs of both caregivers and children in treatment contexts, family-centered rehabilitation designs are slowly emerging in child disability contexts such as traumatic brain injury (TBI) (Mast et al., 2014; Kreitzer et al., 2018) and ASD (Hepburn et al., 2016; Ingersoll et al., 2017; Kurvackel et al., 2018), where a child and caregiver simultaneously engage in rehabilitation activities or interventions that are tailored to their respective needs. Based on findings in manuscript two, parent-child interventions are a promising avenue to explore strategies that optimize the time that caregivers and children spend attending appointments.

6.7. Clinical Implications

The findings of this thesis support the need to address contextual considerations of children with disabilities and their caregivers. Specifically, the first manuscript highlighted the importance of considering area of living when evaluating proximity to therapy and autism diagnosis, as prevalence was higher in higher deprivation areas and less schools offered ABA therapy in higher deprivation areas. In clinical settings, this would look like a health care practitioner starting a conversation about the social challenges faced by a family, as there are numerous social and material risk factors that may create barriers given ones' area of residence and living conditions (e.g., proximity to 'service deserts', job instability, access to transportation, access to medical insurance, stigma, crime, domestic violence). In the conditions and resources required to participate in activities, obtain medications or assistive devices, and attend therapy may bring about the need for alternative or more tailored therapies not otherwise considered if contextual factors are not discussed. Screening tools have been developed to support this line of inquiry (Manchada et al., 2015). The goal of these tools is to optimize patient-centered care by providing a series of SDOH-related questions that address social challenges in sensitive and culturally acceptable language (Manchada et al., 2015; Andermann, 2016). The second manuscript highlighted how personal factors can be more influential than environmental factors depending on the physical impact of the condition. It also highlighted how caregiver psychosocial health can be affected by their child's physical limitations, both positively and negatively, providing clinicians with the opportunity to discuss environmental and personal factors with caregiver psychosocial health as a part of the information important to patient-centered decision-making strategies.

To address this gap, the addition of a knowledge translation (KT) infographic poster was chosen to synthesize a visual representation of cumulative relationships observed between contextual factors. The infographic is intended to propose how environmental factors (including SDOH) underlie personal factors of the caregiver and child, and in what order. The gap that was observed by the literature review, scoping review, and cross-sectional study, was that there is no paradigm which comprehensively represents the numerous factors at play, and their support of one another. From the findings of these two projects, the population-level SDOH were observed as contributors that supported individual-level and personal factors of both the parent and the child. From Manuscript 2, the order or impact is reflected in personal factors, where the caregivers wellbeing is at the top, reflecting that parental stress is situated in response to the child's functional outcomes. The proposed model requires validation and testing in both research and clinical contexts to determine the extent to which these considerations are useful in clinical practice, and accurately presented in order of support and cumulative impact.

This infographic contains 10 ascending steps and corresponding questions in two categories: environmental factors (population-level) and personal factors of the child and caregiver (individual-level). Area-based social and material environment at the base as the 1st consideration, leading up to caregiver well being as the 10th consideration. Each step is cumulatively supported by the previous steps. In this way, if there are barriers at one step, the previous steps can be sequentially considered by clinical inquiry to identify where barriers may be present. If there are barriers identified in environmental factors, the population-level contributions should be considered.

Clinical Implications

In the process of considering feasible and effective treatment options that account for contextual factors, telerehabilitation has emerged as a promising option among childhood disability populations (Camden et al., 2020; Ogourtsova et al., 2023). A 2023 systematic review by Ogourtsova and colleagues identified strong evidence and efficacy that telerehabilitation was found to be as effective, or more effective than face-to-face interventions in ASD, attention deficit hyperactivity disorder, cerebral palsy, traumatic brain injury, and intellectual disability (Ogourtsova et al., 2023). Importantly, telerehabilitation interventions have been tested in children and youth with AMC, showing effectiveness and feasibility for patients. A recent study by Gagnon and colleagues investigated the feasibility of implementing a home exercise program among 11 children/youth 8-21 recruited through the Shriners Hospitals for Children-Canada (Gagnon et al., 2021). Participants found telerehabilitation feasible and effective, and were able to reach their individual goals, improving 12 out of 15 goals related to the improvement of pain management, writing endurance, standing, walking, engagement in sports, and daily activities (Gagnon et al., 2021). By considering strategies that integrate a patients' contextual factors (SDOH, social deprivation, the role of caregivers, the child's personal factors and health condition), clinicians and researchers can better support children with disabilities and improve the resources available to both children and their caregivers.

CHAPTER 7: CONCLUSIONS

The findings from the two manuscripts in this thesis demonstrated a family's contextual factors are important to consider when addressing challenges in childhood disability populations. The gravity of environmental factors (which include SDOH) and personal factors can differ proportionally depending on a family's social and materially-related context, as well as by the impact of the condition. On a case-by-case basis, clinicians can ask patients about the impact of the condition in daily activities and independence, as well as the area-related accessibility, to evaluate the proportion of influence between population-level environmental factors (e.g. no specialists in transportation proximity) and personal factors (e.g. severe impact of mental health or limited mobility).

The objectives of this thesis were met by describing the current evidence regarding population-level SDOH risk factor 'social deprivation' where area of residence and level of social deprivation were impactful to the diagnostic frequency and accessibility of therapy to children with ASD. Researchers and policymakers are recommended to investigate population-level barriers in Canada and the United States, as studies in social deprivation were only found in the United Kingdom and France. The objective regarding individual-level influences of SDOH was addressed by the SEM analysis, identifying that children with AMC are impacted more by the personal factors related to their health condition and biological sex than the environmental factors related to SDOH. Despite the lack of importance of environmental factors, it is possible that the data representing SDOH did not adequately reflect the construct. Future research in registry studies are encouraged to screen for SDOH data from validated screening tools designed to capture SDOH using culturally sensitive language, in order to advance the clarity of data representative of SDOH. Future research focused on patient perspectives and policy advancement are recommended to investigate

SDOH using mixed-methods study with a validated social deprivation index, paired with semi-structured interviews that ask caregivers of and children with disabilities to describe their experience of: their contextual factors (personal and environmental), accessibility to services supporting physical and mental health, the barriers addressing SDOH in clinical settings, social stigma, and mental health status (e.g. frequency of stress, mood disorders).

In answer to the ‘what gives?’ in the thesis title, contributions to health outcomes among childhood disability populations were made by: the region or neighbourhood of residence; the resources (social, material, physical, emotional) of caregivers; the personal factors related to the child and the characteristic impact of their health condition(s); and the psychosocial support from caregivers to their children. Health care practitioners are recommended to 1) discuss contextual factors with both the child and caregiver as a way of comprehensively integrating SDOH in order to optimize patient-centered treatment, and 2) include the caregiver in social support strategies to bolster psychosocial health. Future investigations of SDOH in the context of well operationalized health domains that are relevant to childhood disability may support the provision of care that is individualized to the children and their families.

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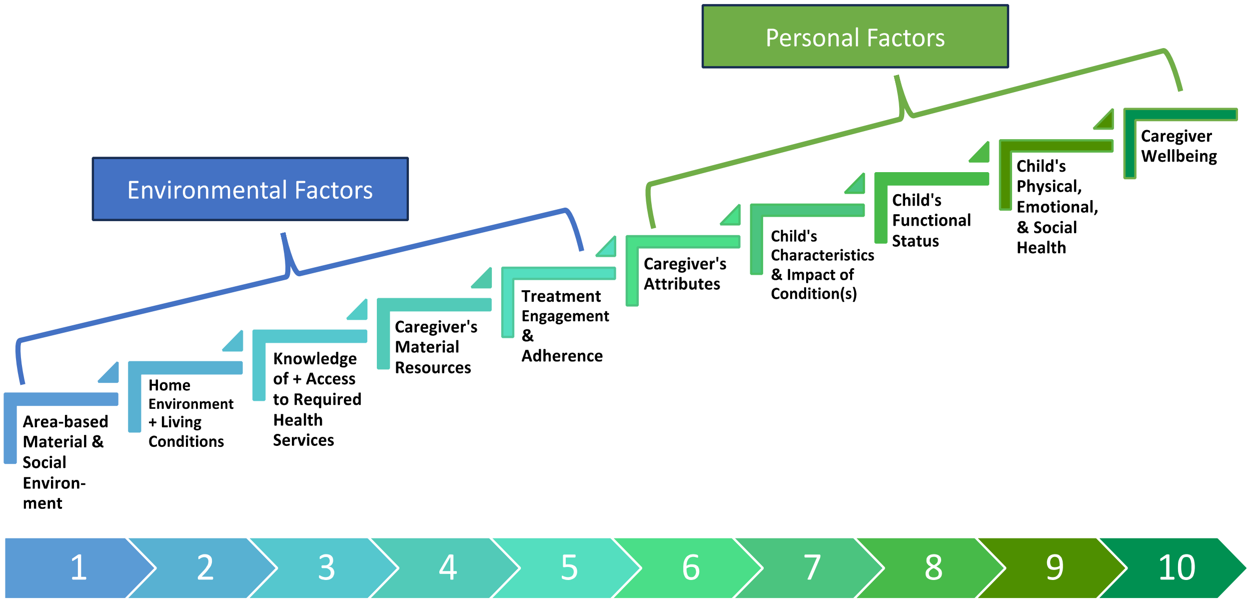
APPENDIX A: 1-page Infographic

Knowledge Translation Infographic: 10 Contextual factors to consider for children with disabilities & their caregivers



1	What are the social and material opportunities available in the area where the family lives?
2	How are the home environment and living conditions supported by the area the family lives in? (e.g. exposure to crime, socioeconomic disadvantage)
3	What is the family's current knowledge and level of access to health services available in their area?
4	What material resources do they have that allow them to access those services?
5	What level of treatment engagement and adherence is recommended by their clinicians, and how does that facilitate or hinder the family's engagement & adherence based on the 4 previous considerations?
6	What attributes does the family have (e.g. ethnicity, education, employment type, temperament, physical health conditions, mental health status) that influence their immediate social and material opportunities?
7	What are the characteristics of the child (e.g. age, sex, temperament) and child's health condition(s)? What needs are required to be met as a result of the health condition(s)? (e.g. assistive devices, surgeries, multidisciplinary treatment)
8	How well can the child function in their current state? Are their specific needs being provided?
9	What is the child's level of physical and social health, and what is required to maintain their health in view of the previous 8 considerations?
10	How can the caregivers' wellbeing be supported in this context? As the caregiver is connected with each of the 9 considerations, do they require support at the individual level (e.g. counselling, support groups) or the area-level (e.g. referrals to services in their area, community-based services, telerehabilitation options, more affordable interventions)?

APPENDIX B: Scoping Review Manuscript Submission

Research in Autism Spectrum Disorders

What is known about social deprivation in autism? A scoping review

--Manuscript Draft--

Manuscript Number:	RASD-D-23-00502
Article Type:	Review Article
Keywords:	Autism spectrum disorders; school-aged children; social deprivation; scoping review; deprivation index; operationalization
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Abstract:	Background: Social deprivation is a multidimensional construct that reflects the interrelated factors comprising the level of social opportunities of a family or individual. As individuals with autism are known to experience challenges in socially related domains, this scoping review aimed to explore what is known about social deprivation in autism. Method: Community colleagues, caregivers, and experts in autism research were consulted to ground the methodology and implications of findings. A literature search conducted in PsycINFO, EMBASE, Ovid MEDLINE, and CINHAL identified 3,146 studies. Results: Six studies met inclusion criteria. Five studies explored area-level social deprivation among children with autism the United Kingdom (UK). The sixth study explored a proactive intervention targeting individual-level skills supporting social functioning in a small cohort of young children at-risk of autism. The term 'social deprivation' was found to be polysemous in both autism and deprivation literature, demonstrating the need for reporting clarity. Deprivation levels were found to be higher than the UK national average, where prevalence increased more dynamically in areas of highest deprivation. Conclusions: To advance research efforts, policy development, and resource allocation strategies, future studies should prioritize: 1) using social deprivation to demonstrate relationships between socioeconomic variation and health outcomes important to the autism community (e.g. social isolation); 2) including community colleagues to inform of current barriers; and 3) clearly describing or operationalizing social deprivation according to the level of observation (ie. area- or individual-level), population context (ie. general or specific to autism), and measurement instrument (ie. questionnaire or census-data).
Suggested Reviewers:	Janine Stichter University of Missouri College of Education stichterj@missouri.edu Dr. Stichter would be a great fit for this review as her research pertains to the individual-level social competence and implementation strategies in education settings for children with autism. She would have specific knowledge of the antecedent behavioural interventions available and how they are reported in literature, and could provide useful feedback regarding individual-level aspects of social deprivation, in additions to interventions that target social improvement for children with autism to strengthen and further situate this review.
	Ailsa Russell

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